Navigating the Swampy Lowlands: Developing Methods for 'Big Picture' Evidence Synthesis

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I also want to acknowledge those who have inspired me by their example and their encouragement. I would not be achieving this PhD without the opportunities they gave or the regard they had for me. I particularly want to thank Professors Alison While, Kate Seers, Iain Chalmers, Mark Strong, Andrew Lee, Jo Thompson Coon and Dawn Craig. My thanks to Eugenie Johnson for her comments on a draft of this thesis.

This PhD represents not only the work written about in this thesis but over twenty years of academic development in the field of evidence synthesis. It is to my family; my husband David, and our four children; Alice, Ben, Grace and Lydia to whom I owe the greatest thanks. My sacrifices have been theirs also. Thank you for being patient, and generous towards me. I love you each to the moon and back and beyond.

For the kindness of colleagues, the love of family, opportunities given and any gift that I possess, I thank a God who I believe gives those good things.

Science is the process of thinking God's thoughts after Him
Johannes Kepler (1571-1630)

Declaration

I, Fiona Campbell, confirm that the Thesis is my own work. I am aware of the University's Guidance on the Use of Unfair Means (www.sheffield.ac.uk/ssid/unfair-means). This work has not been previously been presented for an award at this, or any other, university.

Word Count (14,908) excluding references and text within tables, glossary and abbreviations

Abstract

This thesis aims to demonstrate, with a critical analysis of my publications, my original and substantial contribution to the development of evidence synthesis methodology addressing broad research questions. I use Big Picture reviews as an umbrella term to describe scoping, mapping and evidence gap map methodologies, which I have applied to broad questions in different topic areas. I also aim to demonstrate my original contribution to the topic areas in which I have applied Big Picture methods, a critical reflection of the methods and areas for future methodological research.

Methods

Seven included papers (**Papers A-G**) are used to illustrate how I have interrogated the existing Big Picture review methods, applied them to address a variety of Big Picture review questions, and identified numerous contradictions and inconsistencies within the methods literature and offered solutions to enable others to navigate them. I demonstrate how critical reflection led me to identify limitations when applying recommended methods but also led me to develop novel methods to overcome these limitations. I draw upon Barnett's work (1997, 2015) on 'criticality' as a framework within each chapter to demonstrate how critical thinking and critical reflection has led to critical action. The aim of higher education should be to create learning environments that nurture a critical being; meaning the learner moves beyond thinking and reflection towards action. It therefore is consistent with the aims of this thesis, to show those stages in my own learning and progress as an academic.

Findings

This thesis describes how, while leading, seven reviews using Big Picture methods I have applied critical thinking, reflection and action which has led to my own academic development, and enabled me to make a unique contribution to both Big Picture methods as well as to the topic areas I explored.

The body of work that I describe in this thesis has underpinned a shared understanding amongst methodological leaders in this field of how the methodological approaches of scoping and, mapping reviews and Evidence and Gap Maps have evolved, and how different terminology describing very similar approaches has arisen. I focused leaders thinking

towards a synoptic vision to achieve a consistent approach to review type classification and appropriate use of methodological guidance and reporting standards (**Paper A**).

As well as describing how my reflections have resulted in action, this thesis also affords the opportunity to reflect further on my development as a methodologist and demonstrate how I have applied my own expertise in the reviews. In undertaking a series of reviews using Big Picture review methods I interrogate the methodological guidance underpinning their use, and develop a skill set that has enabled me to apply the methods across a variety of topic areas. These include, the methods of qualitative evidence synthesis (**Paper B**), preterm birth (**Paper C**), mass screening programmes (**Paper E**) and intergenerational interventions (**Paper F**) I present this series chronologically through the different stages of the review process, contrasting the methods used in reviews Big Picture standard systematic reviews of effectiveness and the challenges that these present.

I reflect on approaches to stakeholder engagement, and how this affects effective dissemination of outputs. I also identify challenges encountered and consider how our methods might evolve in order to improve approaches in future reviews (**Paper F**). I describe the particular challenges for locating evidence when the question is broad and the search yield might be unmanageably high for review teams to screen. This work led to further methodological developments, with the creation of a filter to identify studies undertaken in LMIC contexts (**Paper D**). Increasingly, the output from these approaches that commissioners value is the interactive visual map.

I also reflect on how growing methodological expertise has enabled me to cross fertilize knowledge, apply techniques learned in one topic area, and apply them in another. I describe an innovation, introducing evidence and gap map methodology to tumour classification, taking it from the social and environmental sciences and applying it in pathology (**Paper G**). This work has precipitated the development of a new hierarchy of evidence within pathology. A large component of my work has been in training and supporting review groups to undertake these types of reviews and to work closely with knowledge users to ensure our review outputs are meeting a need and filling a gap.

Impact Statement

Identifying the inconsistencies and lack of clarity in the terminology used for these types of review methods has precipitated an acknowledgement that such a lack exists (**Paper A**). I have demonstrated why this lack of clarity has arisen and proposed a route to future clarification. This has led to accepted presentations at key conferences (Cochrane Colloquium 2023, What Works Global Summit 2022, 2023) and ongoing methodological research to help to shape future reporting guidance.

Introducing this method to colleagues working at the WHO IARC (World Health Organisation, International Agency for Research on Cancer) led to a successful grant to map evidence to support tumour classification that informs practice globally. Acting as a methodological expert has provided me with the opportunity of moving methods across topic areas and in so doing has advanced the uptake of evidence in fields where progress has been slow.

Evidence synthesis is a dynamic field of scientific innovation, where methods are evolving and technology is rapidly advancing. This thesis also reveals ongoing questions I am seeking to answer, and a programme of work that is current and live. I intend to continue to contribute positively to the methods of Big Picture evidence synthesis.

Table of contents

List of Tables

List of Figures

Relevant publications and declaration of contribution

	Paper	Role
A	**Campbell, F**., Tricco, A.C., Munn, Z., Pollock, D., Saran, A., Sutton, A., White, H. and Khalil, H., 2023a. Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but different—the "Big Picture" review family. Systematic reviews, 12(1), p.45.	I conceived the idea for the paper, prepared the manuscript, identified collaborators, managed the process of collating feedback, responded to comments from reviewers and I am the corresponding author.
B	**Campbell, F**., Weeks, L., Booth, A., Kaunelis, D. and Smith, A., 2019a. A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance. Journal of clinical epidemiology, 115, pp.160-171.	I secured funding to undertake a methodological study looking at the use of rapid methods in qualitative evidence synthesis. I undertook all stages of the review process and authored the final manuscript.
C	**Campbell, F**., Salam, S., Sutton, A., Jayasooriya, S.M., Mitchell, C., Amabebe, E., Balen, J., Gillespie, B.M., Parris, K., Soma-Pillay, P. and Chauke, L., 2022a. Interventions for the prevention of spontaneous preterm birth: a scoping review of systematic reviews. BMJ open, 12(5), p.e052576.	I was part of the successful team that received NIHR funding under the Global Health Fund. I conceived the idea for the review, as a foundational component of the project. I prepared a protocol, designed the methodology, undertook all stages of the review process, wrote the paper, supervised team members, wrote the original draft, reviewed comments and responded to editorial comments. I am the corresponding author.
D	Sutton, A. and **Campbell, F**., 2022b. The ScHARR LMIC filter: Adapting a low-and middle-income countries geographic search filter to identify studies on preterm birth prevention and management. Research Synthesis Methods, 13(4), pp.447-456.	This was methodological work that evolved from the work described above looking at the evidence for interventions to reduce preterm birth. My co-author led development of the tool, I supported and advised on the process and commented on the manuscript.

E	Foster, C.R., **Campbell, F**., Blank, L., Cantrell, A.J., Black, M. and Lee, A.C., 2021. A scoping review of the experience of implementing population testing for SARS-CoV-2. Public health, 198, pp.22-29.	I was principal investigator for this review funded by Public Health England. I wrote the protocol, designed the methods, managed the project, supervised team members, undertook all stages of the review process, wrote the report which was submitted to PHE. The report that I lead authored formed the basis for this journal article, written by another team member, which I co-authored, checked and commented upon.
F	Campbell, F., Whear, R., Rogers, M., Sutton, A., Robinson-Carter, E., Barlow, J., Sharpe, R., Cohen, S., Wolstenholme, L. and Thompson-Coon, J., 2023b. Non-familial intergenerational interventions and their impact on social and mental wellbeing of both younger and older people—A mapping review and evidence and gap map. Campbell Systematic Reviews, 19(1), p.e1306.	I worked collaboratively with a team at Exeter University on this project. I led the Sheffield arm of this work. Working closely with my colleagues in Exeter, we secured NIHR funding to undertake a body of work which included an EGM and two systematic reviews. We developed methods collaboratively and I co-authored a protocol. I undertook all processes in the review and co-authored the final submitted paper.
G	Indave, B.I., Colling, R., Campbell, F., Tan, P.H. and Cree, I.A., 2022. Evidence-levels in pathology for informing the WHO classification of tumours. Histopathology, 81(4), pp.420-425.	Our team won Horizon funding to undertake EGMs to support tumour classification. I was the lead in the methods processes for the bid. I was instrumental in the initial idea for the project. I commented on and contributed to the writing of the manuscript.

Abbreviations

EBM	Evidence Based Medicine
EGMs:	Evidence and Gap Maps
EPOC:	Effective Practice and Organisation of Care (Cochrane Group)
LMIC:	Low and Middle Income Countries
PICO:	Population, Interventions, Comparators, Outcomes
PHE:	Public Health England
PRISMA:	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PRISMA-ScR:	Preferred Reporting Items for Systematic Reviews and Meta-Analyses for Scoping Reviews
QES:	Qualitative Evidence Synthesis
RCT	Randomised controlled trial
NIHR:	National Institute for Health and Care Research
PRIME:	NIHR Global Health Research Group on Preterm Birth Prevention and Management

Glossary

Big Picture Review:	A term that encompasses those evidence synthesis methods designed to address 'big picture' review questions.
Effectiveness review:	A term that to denote an evidence synthesis method that is addressing a research question that is concerned with the effects of a treatment, intervention or policy.
Qualitative Evidence Synthesis:	A term that encompasses review approaches that use systematic and explicit methods to locate, analyse and synthesise qualitative research studies.
Systematic Review:	Systematic reviews are a methodological approach that aim to use rigorous, transparent and reproducible methods to locate relevant research to answer a particular research questions and to synthesise those findings.

Evidence Synthesis:	This is an umbrella term that covers all types of approaches to evidence synthesis. What is 'evidence' will depend on the review, but in this context refers to research evidence. In some circumstances, expert opinion might be regarded as evidence.
Scoping Review:	A transparent, rigorous and systematic approach to identifying, describing and cataloguing literature available for a particular topic, field, concept or issue. It may seek to identify key concepts, theories or sources of evidence. It is exploratory, not requiring an a *priori* set of codes in order to describe data and may draw upon diverse sources of information *(i.e. primary research, reviews, non-empirical evidence) within or across particular contexts.*
Mapping Review:	Mapping reviews are also a transparent, rigorous and systematic approach to identifying, describing and cataloguing evidence and evidence gaps in a broader topic area. A mapping review typically extracts only descriptive information about the studies and applies predefined codes).
Evidence and Gap Map:	Evidence and Gap maps are described as "a systematic presentation of all relevant evidence of a specified kind for a particular sector, subsector or geography". Evidence and Gap Maps (EGMs) are a systematic evidence synthesis product which displays the available evidence relevant to a specific research question.
Methodologist:	A methodologist is someone who specializes in the methods for studying something and in the context of this thesis, it refers to someone specializing in the study of evidence synthesis.
Information specialist:	Information retrieval specialists and the methods they employ are essential for research projects that require unbiased retrieval of relevant studies or that depend on judicious selection of resources and understanding of sophisticated search syntax.
Horizon:	Horizon is EU's key funding programme for research and innovation

‘In the varied topography of [evidence synthesis], there is a high hard ground overlooking a swamp. On the high ground, manageable problems lend themselves to solution through the application of research-based theory and technique. In the swampy lowland, messy, confusing problems defy technical solution. (Schön, 1987, p. 3)’

Chapter One: Background

Evidence based medicine (EBM), emerged in the early 1990s, and was hailed as a 'new paradigm' in medical science (Guyatt et al, 1992). It had developed from the work of clinical epidemiologists and was inspired by the work of Archibald Cochrane, a physician and epidemiologist. Cochrane was critical of the unquestioned use of untested clinical interventions and practices used by medical doctors and advocated the need to use randomised clinical trial (RCT) evidence to inform understanding of its effectiveness and hence, efficiency (Cochrane, 1972).

This new 'paradigm shift' was designed to challenge an authoritarian or eminence-based attitude in medical care with an evidence-based one (Straus et al, 2011; Bhandari et al, 2004). The research methods it advocated were the use of 'double blinded' randomised controlled trials, with systematic evidence reviews and meta-analyses of RCTs as the highest form of evidence. Whether it truly was a 'new paradigm' is debated (Sehon & Stanley 2003; Solomon, 2010). However, the impact of EBM has been considerable. It has been accompanied by an increased acknowledgment that practices and policies should be supported by scientific evidence. EBM has become a social movement with associated institutions (Cochrane Collaboration, Campbell Collaboration, What Works Centres including the National Institute for Health and Care Excellence (NICE), Social Care Institute for Excellence (SCIE)) and a field characterised by a core of technical guidance and exemplars.

During the past three decades, not only has there been a diffusion of the evidence based practice movement to diverse practice and policy areas, (education, social sciences, criminal justice, international development and the environment), but also an evolution in its core epistemic methods. The hierarchy of evidence, with systematic reviews, which include meta-analyses of double blind RCTs at its pinnacle, and considered the 'gold standard'. This hierarchy has not been replaced, but joined by other types of study design and approaches to synthesis. Synthesis methods have developed to include other study designs and those designs are considered equally valuable in informing decision making (Greenhalgh et al, 2022). There is also a large and growing variety of approaches to

evidence synthesis. A recent catalogue of terms identified 48 distinct review types (Sutton et al, 2019). Moher et al (2019) describe them as a 'family' of evidence synthesis products.

I suggest that multiple potential drivers for these changes have resulted in an increasingly diverse portfolio of approaches to evidence synthesis and a broader view on what constitutes useful evidence to inform decision-making. This includes; recognition that RCTs have significant limitations; they may not be ethical, or logistically feasible, and may have limited external validity (Mustafa, 2017; Cartwright and Munro, 2010; Sanson-Fisher et al, 2007). They may also not be the best design to assess important aspects of effectiveness (such as the potential harms of a treatment) (Cornelius & Philips, 2022).

While RCTs remain a valuable study design for questions of treatment effectiveness, the questions being asked by knowledge users and policy makers may have a different purpose. For example, the research question might seek to understand the factors that influence treatment acceptability, or the diverse intervention options available. Neither of these is best answered with an RCT. Therefore, different types of primary studies may require different types of synthesis. Qualitative evidence synthesis is recognised as valuable in establishing the relative importance of outcomes, acceptability, fidelity, feasibility and equity of interventions (Flemming et al, 2019). This is evident in the changing features of the Cochrane Collaboration Handbook, which now includes guidance on qualitative evidence synthesis (Noyes et al, 2022).

Another factor has been the need for improved understanding of the ways in which context can act as a mediator of effectiveness and the consequent need to understand why things work (mechanisms) and how they work within systems, with impacts that might be both intended and unintended. Evidence synthesis methods are increasingly being developed and utilised to integrate diverse types of evidence and address the complexity of interventions and systems in healthcare (Dixon-Woods et al., 2006a; Flemming et al., 2019). Different approaches, such as; critical interpretive synthesis, meta-ethnography, realist synthesis and systems perspectives, have been proposed and applied to understand the mechanisms, contexts and outcomes of complex interventions (Dixon-Woods et al, 2006; Rycroft-Malone et al, 2012; Jagosh, 2019; Hong et al, 2022; Dixon-Woods et al, 2006b;

Campbell et al, 2018). The synthesis of quantitative and qualitative evidence is particularly useful for understanding the complexity associated with the implementation of complex interventions in health systems (Noyes et al, 2019). These evidence synthesis methods provide evidence-informed theories, explanations and insights into how interventions function in complex environments (Jagosh, 2019; Norris et al, 2019). Addressing these types of questions has again broadened the methods needed to locate, include and synthesise relevant and useful evidence.

Additional drivers include the needs of stakeholders and policy makers, which have played a significant role in shaping the methods used in evidence synthesis. In situations where there is a lack of RCT evidence, the notion of "no evidence" is often unhelpful when making decisions (Yaffe et al, 2012). This is because stakeholders and policy makers require actionable information to inform their decision-making processes. Therefore, alternative types of evidence, such as observational studies or expert opinions, may be employed to provide valuable insights and guidance in the absence of RCT evidence. Policy makers often face time constraints when making decisions, requiring evidence synthesis methods that can provide timely results. As a result, there has been increased use of rapid approaches in evidence synthesis to meet these needs (**Campbell et al, 2021b**; Ganann et al, 2010; Garritty et al, 2021).

The relatively new methodological science of evidence synthesis remains a dynamic one, in which methodologists increasingly explore and test new approaches and where advancing technologies add to the altering landscape (Revaud et al, 2020). It is within this dynamic landscape that the work encapsulated within this thesis has occurred. Between 2018 and 2022 I was increasingly addressing research questions that did not relate to treatment effectiveness, or seek to address a specific question. Questions were very broad and the answers needed required a 'big picture' view (**Paper A**). I became intrigued by the approaches that were the most appropriate: scoping, mapping and evidence and gap maps. As I explored, adopted, applied and critiqued, the methods I found many limitations in existing guidance, a lack of good examples to inform practice, considerable discrepancy within the literature regarding terminology and rapidly evolving technology shaping the availability of tools to support these types of reviews.

That body of work has not only generated this thesis but has also resulted in methodological papers (**Paper A and B**), methodological tools (**Paper D**), guidance for decision makers during the COVID-19 pandemic (**Paper E**), international collaborations, formation of a methods group of experts, the application of approaches to new fields (**Paper G**) and short course training programmes. The following chapters firstly describe the methods used to shape this thesis followed by the findings, which document my original contributions both to the methods of Big Picture review methodology but also to the topic areas which were the focus of the reviews. I conclude by identifying methodological areas of ongoing and future research where I intend to continue making a positive impact.

Chapter Two: Methods for this Thesis

Throughout this thesis I will refer to my seven included papers (**Papers A-G**), using each to show my development as an academic researcher and highlighting where I have made a unique contribution either to the field of evidence synthesis methodology or to the topic area I was investigating. **Paper A** is the key methodological paper that has highlighted the discrepant use of terminology, its origins and a proposal for a way forwards by providing a proposed distinction between scoping, mapping and EGMs and a collective term, Big Picture Reviews. **Papers B-G** are derived from seven reviews, which exemplify scoping, mapping and/or evidence and gap map methods. These reviews (case studies) covered diverse topic areas, and received funding from different sources. They were also undertaken within different time frames and the teams were configured differently. The reviews included in this thesis as published examples are summarised in Table 1. In all cases I led the project, designed the methods, contributed to all aspects of the review process, supervised and trained team members and led or co-led on dissemination of the results.

In addition to the included papers (**A-G**) and the associated reviews, I will also refer to two further reviews and associated publications where it is useful within the text, though these are not included papers submitted with this thesis. This is also illustrated in Table 1. For those still in press details of their status are in Appendix A. In paper (**Paper C**) and Campbell et al (2019, in press), the published papers are derived from extensive unpublished reports. The findings and reflections in this thesis will draw upon elements of these projects that are not described in the publications, such as the reports provided to funders, notes, and protocols.

Table 1: Candidate Papers, and Associated Reviews *

Included Papers	Reference	Title	Review Topic (case study)	Funder	Timeframe	team members
Paper A	Campbell et al, (2023a)	Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but different— the "Big Picture" review family.	Key overarching methodological paper that has emerged from the collective work cited in this paper			
Paper B	(Campbell et al, 2019a)	A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance	Rapid qualitative evidence synthesis	University of Sheffield	2 months	TE, SR (2), IS (International)
Paper C	Campbell et al, (2022a)	A Scoping Review Highlighting an Inverse Pattern of Research with a Lack of Research Evidence from High Burden Settings.	Preterm birth	NIHR	1 year	TE (10), IS (1), SR (1) SH () (International)
Paper D	Sutton and Campbell (2022b)	The ScHARR LMIC filter adapting a low-and middle income countries geographic search filter to identify studies on preterm birth prevention and management	Methodological tool that emerged from the review (CP3) exploring interventions to reduce risk of preterm birth			
Paper E	Foster et al, (2021)	A Scoping Review of the Experience of Implementing Population Testing for SARS-CoV-2.	Screening for COVID-19	Public Health England	3 months	TE, C, SR (2), IS (institutional)
Paper F	Campbell et al, (2023b)	Intergenerational interventions and their effect on social and mental wellbeing of both children and older people – A mapping review and evidence and gap map.	Intergenerational interventions	NIHR	12 months	TE, IS (2), SR (3), SH () (National)
Paper G	Indave et al (2022)	Evidence-levels in pathology for informing the WHO classification of tumours.	Tumour classification	Horizon	3 years	TE (15), SR (2), SH (), IS (1) (International)
Not included but cited	Campbell et al (2019b, in press)	Epilepsy Specialist Nurses The Evidence (ESPENTE): a Systematic Mapping Review	Role of epilepsy nurse specialists[1]	Epilepsy Action	8 months	TE, C, SR (2), IS (institutional)
Not included by cited	Mikton et al (2022)	PROTOCOL: Global elder abuse: A mega-map of systematic reviews on prevalence, consequences, risk and protective factors and interventions	Elder abuse[2]	WHO	1 year	TE (7), C, SR (1), IS, (International)

TE: topic expert, IS: information specialist, SR: systematic review, C: commissioner, SH (stakeholders)

*These examples are referred to in the text of this thesis, and in some instances are the focus of the included candidate papers. In two examples [1,2] the associated papers are under editorial consideration but the report and/ or protocol is in the public domain.

While training and working as a nurse, health visitor and district nurse, in primary and secondary care settings and in both high and low income countries, I learned of Donald Schön's work and his description of the 'expert' practitioner(1983,1987,1992). Schön described the 'expert practitioner' as one who tailors theoretical and research-based knowledge (the 'high ground') to fit the circumstances encountered in specific practice situations (the 'swampy lowlands'). This depiction of professional practice where the expert grapples with the complexities of both (scientifically derived) solutions with practice using professional artistry to move forward as effectively as possible was one that resonated very powerfully. Observing the way a skilled colleague would, for example, relieve a dying patient's pain by gentle repositioning of their limbs and bedding, as well as timely administration of prescribed medicines, taught me more than any textbook. Nor would my nurse colleagues notice or be able to describe the years of learning that such a task would demonstrate. I have returned to Schön's work, and considered it again when preparing this thesis. Although particularly used to describe the work of nurses and social workers, I see similar processes in the work of the expert methodologist. There is a wrestling with the methodology and its underpinning theoretical guidance, the realities of the review itself, and often the need for 'professional artistry' to move forwards and to apply lessons learned.

Criticisms and limitations of Schön's work may extend to considering its questionable application outside of the health and social care context and application to the development of research methodologies. Neither, does it capture the notion of acting as an agent of change. I have therefore also drawn upon the work of Barnett (1997, 2009) and Davies and Barnett (2015) to provide a framework for the development of 'criticality' within Higher Education. These are particularly valuable in the context of demonstrating my own development in critical analysis, reflection and action.

Barnett's work brings together three domains of critical practice: critical analysis, critical reflexivity and critical action. His model of reflective theory felt particularly appropriate to this thesis as it describes what must be the purpose of Higher Education. If Higher Education solely comprises of critical thinking, but does not lead to critical reflexivity and critical action, then our society becomes incapable of change and thus diminished: "Critical

being has to be the business of higher education" (pg. 7 Barnett, 1997). As well as being particularly relevant for supporting the framework for a product of Higher Education, the journey from critique to seeking to make change particularly resonated with the evolution of this work.

Critical analysis (Critical thinking and critique)

Barnett describes critical thinking as the cognitive acts undertaken by individuals. It is collaborative in character, developing through sustained interchange around collective standards. Disciplines, including evidence synthesis, contain their own critical standards through which they interrogate the world. Critical thinking is criticism within the discipline, conducted according to its values and procedures; critique is a form of criticism about the discipline itself.

Critical self-reflection

Critical self-reflection points to the ability to move oneself forwards. The student interrogates her own thinking or her actions, recognising that other thoughts or actions might be even more worthwhile. In the process, new thinking and new acts may emerge. The self-reflection is accompanied by self-criticism.

Critical action

Critical actions are that form of criticality which finds expression in direct engagement with the world. Critical action is an intrinsic, aspect of criticality. Action is regarded as important for not only encouraging students' personal individual critical comprehension of, and reaction to events, but as a justification for political and social change. Critical pedagogy would never regard thought as sufficient if it did not lead to challenging and transforming institutions, ideology (including research ideology), and society. Higher education, therefore, has the potential for acting as a transforming device in society.

As a structure for the thesis, I use components of the systematic review process; formulating the question, selecting the method, engaging stakeholders, locating the evidence, making sense of the evidence, and creating an evidence resource as chapter headings. This structure is not exhaustive, for example, there is not a chapter on quality appraisal. However, it provides a loose chronological structure to the thesis. Within each

chapter, I then explore each component using the three critical processes described by Barnett (Figure 1).

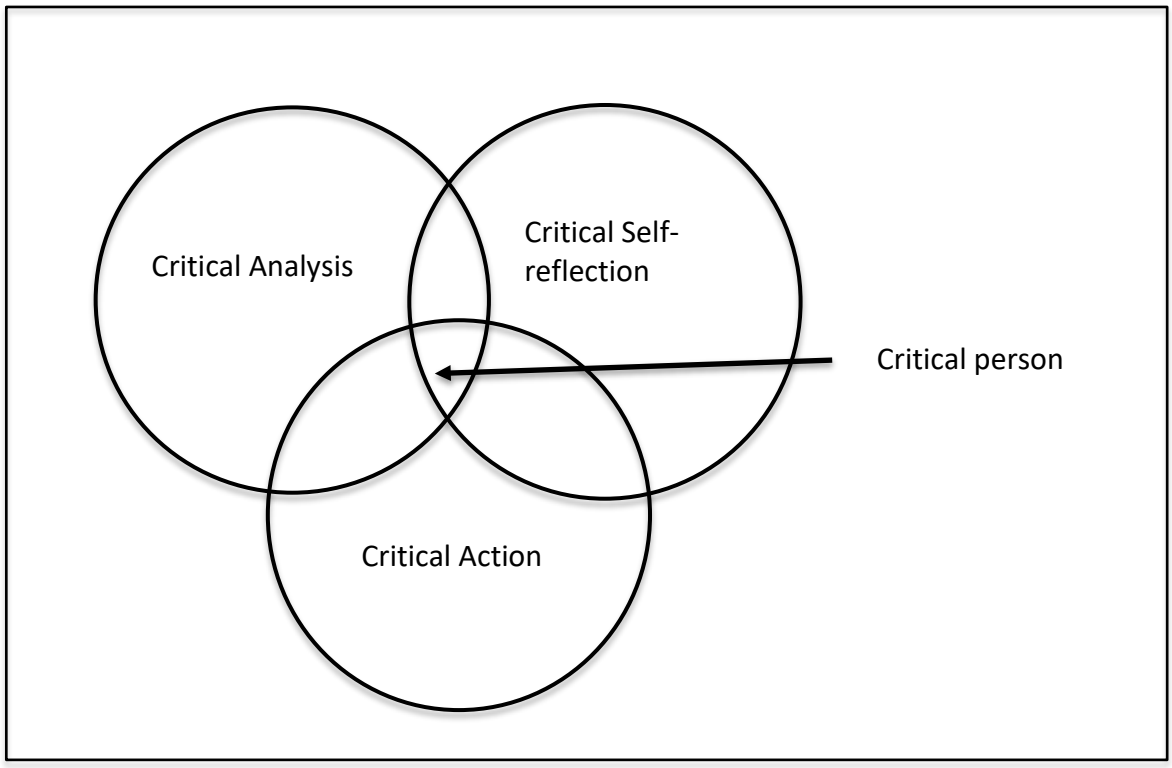

Figure 1:The intersection between critical reason, critical self-reflection, and critical action (Barnett 1997, in Davies and Barnett 2015 pg 17)

Thesis Aims and Questions

This thesis aims to demonstrate:

- A critical use of scoping, mapping and EGM methodologies across a variety of topics, identifying the particular challenges of these methods and how I have successfully addressed these and where my reflections highlight the need for further innovation (critical thinking and reflection
- My original contribution (critical action) to scoping, mapping and EGM methodologies, and also evidence synthesis more broadly, as a result of using these methods in the included case studies (**Paper A and D**)
- My original contribution (critical action) in the topic areas, which are a focus of the included case studies (**Paper B, C, E, F**)
- To show how expertise as a methodologist has led to innovations (critical action) in the use of scoping, mapping and EGMs in new fields (**Paper G**)

The thesis questions are laid out in the table below (Table 2).

Table 2: Thesis questions, associated chapters and included papers

To what extent does the PICO framework support question formulation in scoping, mapping and EGMs? (**Paper B and E**)	Chapter 3
Why have differences in terminology arisen and do they matter? What might be the way forward? (**Paper A**)	Chapter 4
What was the impact of stakeholder engagement on the review findings and what did I learn for future reviews. (**Paper F**)	Chapter 5
What are the challenges in locating evidence for scoping, mapping and evidence and gap maps? How challenges can lead to innovation (**Paper C and D**)	Chapter 6
How can the processes of data extraction and data coding be best managed in scoping and mapping and evidence gap maps, learning the hard way. (**Paper B, C, E, F**)	Chapter 7
Why the visual outputs of evidence and gap maps address a knowledge user need (**Paper F and G**)	Chapter 8

Chapter Three: Question Formulation

Paper B
Campbell, F., Weeks, L., Booth, A., Kaunelis, D. and Smith, A., 2019a. A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance. *Journal of clinical epidemiology*, *115*, pp.160-171

Paper E
Foster, C.R., Campbell, F., Blank, L., Cantrell, A.J., Black, M. and Lee, A.C., 2021. A scoping review of the experience of implementing population testing for SARS-CoV-2. Public health, 198, pp.22-29.

Critical analysis

Good science begins with a well-defined research question and the question is key to choosing a suitable study design. The PICO was adopted as a key part of systematic review methodology (O'Connor et al, 2008), used to structure and refine the review question (Buckley et al, 2016), shape the development of the search strategy, underpin the inclusion and exclusion criteria for the review, and help determine the logistics of the review. As the application of systematic review methods have been applied to and adapted for other types of questions, beyond intervention effectiveness, the PICO remains the framework that much of the guidance and checklists continue to use e.g. PRISMA (Page et al, 2021).

The limitations of the PICO framework, even within its original paradigm of medicine, have been discussed in the literature (Eldredge and Nogar, 2022; Huang et al, 2006). Within a clinical medical setting many evidence based questions are not treatment oriented questions, but belong in the domains of diagnosis, prognosis and epidemiology. Key aspects of the research objectives are not addressed using the PICO framework and some elements of the framework might not be relevant. The need to adapt PICO has been shaped by those factors described in Chapter One, with a growing diversity of types of review questions, a broader inclusion of types of evidence and different methods of synthesis. The challenges of using PICO for reviews of complex interventions (Squires et al, 2013) and in qualitative evidence synthesis (Cooke et al, 2012) have been documented. Critiques of the PICO framework has led to the development of new frameworks to guide question refinement. Davies (2011) describes 12 variations of the PICO framework, where the framework has been modified and, in most cases, to incorporate other features of a research question. A more recent review (Booth et al, 2019) identified 38 different

question formulation frameworks. I have presented these in Table Four, grouping those where the components of the question formulation frameworks are the same or similar (derived from Booth et al, 2019).

For scoping, mapping and EGMs, the guidance on question formulation and the development of alternatives has been more limited. The Arksey and O'Malley (2005) methodological guidance for scoping reviews, suggests using PICO. They recommend that reviewers "consider which aspects of 'facets' of the research question are particularly important, for example the study population, interventions or outcomes" (pg. 23).

More recently the Joanna Briggs Institute (JBI) recommended the use of the PCC framework (Population, Concept, Context) for scoping reviews (Peters et al, 2020). Concept refers to the concept of interest and can be more broadly defined. It can refer to types of interventions, but also may be definitions or a study design. Context varies depending on the objective of the review and may be a geographic location, a particular country or region, or a specific setting (such as schools). Despite the development of PCC, many scoping and mapping reviews still use PICO but adapt it (Konlan et al, 2022, Milne-Ives et al, 2022, Wirawan et al, 2023). Guidance for EGMs recommends the use of the PICOS framework with 'S' representing study design (White et al, 2020).

Critical Self-Reflection

In 2017- 2018 I led a team undertaking a scoping review looking at the methods used in rapid qualitative evidence synthesis (QES) (**Paper B**).

Our research objectives were to:

1. identify existing methodological guidance for the conduct of rapid QES; and
2. identify examples of rapid QES and describe the methods used.

Then, in 2020, I led a team undertaking another scoping review to support decision making on population level screening, during the early stages of the COVID-19 pandemic, (**Paper E**).

Our research objectives were to:

1. identify the variety of ways mass screening for COVID-19 was being and had been undertaken;
2. describe their processes; and
3. describe any evaluation of their accessibility, acceptability and impact on equity.

These two reviews illustrate the challenges of using the PICO or PCC frameworks in the context of scoping, mapping and EGMs. The diversity of topics, and objectives in these types of reviews can be so diverse that no single mnemonic is likely to completely 'fit'. However, in Big Picture reviews, clear communication within the internal review team and externally with stakeholders makes clear articulation of the review question and its components particularly important. It is evident from Table 3 that we adapted, and added, to the recommended frameworks as needed. In case study E (**Paper E**) (Foster et al 2021) we were interested in many types of interventions, with a common purpose (mass screening). We were not able to pre-specify all types of interventions. The purpose of the review was also exploratory, seeking to identify the various ways in which mass screening had been undertaken.

In the review exploring methods of QES (**Paper B**), we returned to the initial question formulation multiple times during the review process. We found, for example that papers described as systematic reviews often lacked methodological criteria that we felt would be an essential component of a systematic review. We also found QES that had used a qualitative approach in its synthesis of quantitative studies. We had not anticipated these a priori and had to adjust our inclusion and exclusion criteria to make these decisions clearer. In this case, the criteria we used to develop our search, was developed and refined as decisions on inclusion and exclusion became unexpectedly opaque and as differences emerged within the team during independent and blind screening processes. Table 3 illustrates how the PICO and PCC related to the two reviews (**Paper B and E**)

Table 3: Applying PICO and PCC

	Paper B (QES)	**Paper E (COVID-19)**
P Population	N/A	symptomatic and/or asymptomatic individuals for COVID-19
I Intervention	N/A	providing or accessing a population testing point using an antigen or antibody test in any setting, using any testing modality
C Comparator	N/A	N/A
O Outcome		Accessibility, barriers, facilitators, equity
C Concept	Qualitative evidence synthesis – examples and guidance	N/A
C Context	N/A	Studies from high or high middle income settings

In our review examining methods for mass screening (**Paper E**), our review methods were iterative. We changed the focus slightly as we progressed, for example, by excluding papers describing testing of passengers at ports or borders. Our focus widened to consider mass screening in the context of other epidemics where there was potential for a pandemic. Neither the review team, nor our funders, knew what the range of interventions might be at the outset. Our team developed the inclusion criteria informed by the protocol. Decisions were made in collaboration with the review team, topic experts and our commissioners.

As stated, the PICO (or alternative) has several purposes in the review process: to define the question; to inform the search; and criteria to determine inclusion during screening. As I have illustrated in both of these case studies, we used our PICO framework very flexibly and we also had several stages of refinement of our question and its scope once the extent or limitations of initial searches and screening began to reveal more about our phenomenon of interest.

This iterative and exploratory approach has also been described by Sager and Pistone (2019) who suggest that the methods for scoping reviews would benefit from a pronounced role of 'informalities'. They draw upon concepts within science and technology, where informal and formal methods exist in a dynamic and creative interplay. Formalities (rules,

guides and frameworks) are used but do not entirely control actual practices, since experts adjust standards and adapt situations in different ways according to the perceived needs of the review. It is therefore more apt to describe the process as a 'dance of (in) formalities'. This aligns well with the 'swampy marshlands of practice' that Schon describes and one which the expert 'artistically' navigates.

In these case studies (**Paper B and E**), our approach was indeed a 'dance', an interplay between formal approaches, informal judgements, and negotiations. In this context, a 'PICO' becomes a shorthand for an approach allowing a team to select of those elements in Table 3 that are best suited to the review question; a 'pick and mix' rather than a prescribed mnemonic. Different components may be introduced in an iterative and negotiated way as the searches progress, the landscape becomes clearer, and further refinement of the scope becomes feasible.

Critical action

These two case studies, and the associated papers (**Paper B and E**) made unique contributions to the knowledge in the fields they were investigating. In March 2020, at an early stage of the COVID-19 pandemic in the UK the team I lead were commissioned by Public Health England (PHE) to undertake a review that would identify different approaches to mass testing, their accessibility, acceptability and cost. Although our findings were limited by a lack of evidence available, our team were able to show that drive-through testing centres were the most common testing modality evaluated and these provided a rapid method of testing whilst minimising resource use. These findings helped inform national testing policies as part of the pandemic response efforts to minimise health, social and economic harms (**Paper E**).

Our scoping review of methods used in undertaking rapid QES (**Paper B**) was also the first methodological study to explore why, and how rapid QES was being applied. Our team were also interested in locating guidance to support these methods. The findings were consistent with other investigations of rapid approaches, showing a lack of guidance and no standard approach to rapid QES (Abou-Setta et al, 2016). The most notable feature they shared was that in the synthesis they sat in the 'mostly unchanged concept's' end of the

continuum of conceptual innovation that occurs during the synthesis process (Thomas et al, 2017).

Another aspect of evidence synthesis methodology uniting these reviews (**Paper B and E**) was rapid approaches to synthesis. In (**Paper E**) the review was undertaken within a six-week timeframe, and in (**Paper B**) rapid methods were the focus of the work. This has supported an ongoing programme of research and interest in rapid review methods, where I have been leading and teaching on the only rapid review methods course in the UK. It has also resulted in the first prepared guidance on rapid approaches in QES (**Booth et al, in press**) and rapid scoping, mapping and EGMs (**Campbell et al in pressA**) as part of a series in rapid review methods (Garritty et al, 2021).

Table 4: Components of the Question Formulation Frameworks (derived from Booth et al, 2019), grouped based on the similarity of the component but the original name is maintained

Question Formulation Components											
A	**B**	**C**	**D**	**E**	**F**	**G**	**H**	**I**	**J**	**K**	**L**
Population	Context	Problem	Concept	Intervention	Comparison	Mechanisms	Outcomes	Professionals involved	Stakeholders	Time	Study design
Patient	Environment	Issues	Construct of interest or measurement	Type of measurement instrument		Models/theories	Themes			When	
Who	Location	Condition	What	Service		How	Measurement properties			Duration	
Person							Impact				

Chapter Four: Choosing a method

Paper A
Campbell, F., Tricco, A.C., Munn, Z., Pollock, D., Saran, A., Sutton, A., White, H. and Khalil, H., 2023a. Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but different—the "Big Picture" review family. Systematic reviews, 12(1), p.45.

Critical Analysis

The first three Big Picture reviews I undertook (**Paper B, C** and Campbell et al (in press B)) made me aware that there were inconsistencies in the definitions and accompanying guidance for scoping and mapping reviews. This was not resolved by looking at other published examples. This led me to try and explore whether these differences mattered, the roots of the methods, and what might be a useful way forward in order to support greater consistency in reporting and also enhance clarity of methodological approaches. This chapter describes that exploration, the results of that thinking were published in **paper A.**

Why terminology matters

I would argue that terminology in research methodology does matter for the following reasons. The evolving 'family' of evidence synthesis types presents greater challenges in selecting the most appropriate tool for the task (Moher et al, 2015). Forty-eight different terms now are used to describe different approaches to evidence synthesis (Sutton et al), and 41 of these are embedded in the 'Right Review Tool' (https://whatreviewisrightforyou.knowledgetranslation.net/) (Amog et al, 2022) which can support reviewers in the selection of the correct methodology. However, as Grant and Booth (2009), many of the labels used fall short of being mutually exclusive and there is a lack of unique distinguishing features for most of the common review types. Where there is lack of clarity and overlap in approaches, selecting the correct type is challenging.

A lack of consistency and uncertainty in terminology leads to complicated, and potentially unresolvable, peer reviewing processes, with researchers, funders, peer reviewers, publishers and knowledge users potentially adhering to a 'preferred' terminology. This has implications for appropriate benchmarks for quality, use of publishing guidance and

reporting standards. Different expectations may evolve, when terminology is unclear making reviews difficult to commission and deliver.

Critical Reflection

Scoping reviews and mapping reviews – how are they used in the literature

Within the published literature, the terms scoping reviews and mapping reviews appear to be used in three different ways:

1. interchangeably (the same type of review in purpose and method);
2. complementary (each describes a feature of the same method); and
3. different (different types of review in purpose and method).

Interchangeably: Where the terms refer to the same type of evidence synthesis product

There are examples within the literature where 'mapping' and 'scoping' are used interchangeably, referring to the same type of review methodology (Colquhoun et al, 2014, Shemilt et al, 2014; Peters et al, 2020). This approach is also used in the PRISMA Extension for Scoping Reviews (PRISMA-ScR) (Tricco et al, 2018). Examination of published reviews does not reveal differences in method between these approaches (Khalil et al (in press)).

Complementary: Where the definitions refer to review processes that are complementary.

Some definitions suggest that mapping is a specific approach to scoping or scoping being the purpose of a mapping review. For example: "Scoping reviews can usefully map the evidence in multiple ways" and "scoping reviews are a way of mapping the key concepts" (Fernandez-Sotos et al, 2019 and Lukersmith et al, 2016). It has also been suggested that the inclusion of the term "mapping" in the method description implies the incorporation of a geographical mapping exercise or the charting of data in a visual format, such as a table or other visual representation.

Different: Where the terms refer to different types of evidence synthesis product.

Grant and Booth (2009) and Sutton et al. (2019) make a distinction between mapping and scoping reviews. According to these authors, scoping reviews are a preliminary assessment of the potential size and scope of available research literature, aiming to identify the nature

and extent of research evidence, including ongoing research. Mapping reviews, on the other hand, are differ from scoping reviews because the subsequent outcome may involve further review work or primary research, and this outcome is not known beforehand. Scoping reviews, within this definition, are not usually regarded as a final output in their own right due to limitations in their rigor and duration, which may introduce bias.

Gough et al. (2012) suggest that the term "scoping review" often describes a rapid and usually non-systematic approach to describing the nature of the literature on a topic area, sometimes as part of planning for a systematic review. It has also emerged within the systematic review literature to describe the preliminary work undertaken with information specialists in planning the review, by getting a sense of the size of the literature, identifying key terms and theories, and potentially consulting with clinical experts [2].

Bragge et al (2011) suggests yet another alternative view that scoping reviews can be distinguished from mapping by the inclusion of research results in the description of relevant evidence, whereas maps simply describe what is there without collating and summarising the results of the studies.

It is evidence that even where the types of products are seen as different, there is not a consistent agreement on what those differences are. Nevertheless understanding why they are considered different is important in considering what is lost, if the terms are amalgamated and used interchangeably.

Finding the roots…

In exploring the terminology and its roots, I suggest different terms have emerged to describe two similar methodological approaches as they have arisen from different academic traditions. These disciplines have different epistemological foundations upon which these are built. Scoping reviews tend to cite the framework defined by Arksey and O'Malley (2005) and later enhancements by Levac et al (2010) with their roots in sociological sciences. In contrast, the term evidence mapping was used first by Katz et al (2003) and has roots in the natural sciences. This was the term adopted by the EPPI Centre (UCL, London) in an early publication of a mapping review and is the term used by the

Centre for Environmental Evidence for the environmental sciences (Haddaway et al, 2016). The approach to evidence mapping accompanied by a visual EGM has been developed by several agencies (Saran and White, 2018), including the International Initiative for Impact Evaluation (3ie) (Snilstveit et al, 2013) in the field of international development and subsequently adopted and adapted across multiple sectors through the Campbell Collaboration. These include, for example, transport (Malhotra et al, 2021) , youth violence, disability (Saran et al, 2020) and elder abuse (Mikton et al, 2022). These roots are summarised in Table 5.

Table 5: Summary of the different roots and institutions that use mapping and scoping reviews

	Scoping Review	**Mapping Review**	**EGM**
Academic roots	Social sciences and health	Environmental science and health	International development
Guidance for methods	PRISMA ScR and JBI Arksey and O'Malley, 2005 Levac et al, 2010, Peters et al, 2015, Peters et al, 2020, Tricco et al, 2018	James et al, 2016, Katz et al, 2003	Campbell Collaboration White et al, 2020 Snilstveit et al, 2013
Identifies gaps in the research	Yes	Yes	Yes – using a pre-specified framework
Visual and interactive web based gap map	No – but may contain within text tables and diagrams	No – but may contain within text tables and diagrams – and may be produced with an EGM	Yes

Critical Action

Suggested approaches for distinguishing between mapping reviews and mapping reviews with EGMs and scoping reviews

The creation of two terminology (scoping and mapping) to represent techniques that share similar aims and procedures demonstrates that, in many ways, the labels employed are formed by the researcher's academic background rather than any fundamental distinctions in the approaches.

Going forward, it is worthwhile to consider if this is truly relevant. As I have demonstrated, the terms 'mapping' and'scoping' are frequently used interchangeably. In this study, I argue that, while there is significant overlap between these methodologies, there is importance in distinguishing between scoping reviews, mapping reviews, and EGMs. They might also be regarded complimentary, and a review may contain components of both 'mapping' and 'scoping'. Each technique in this family of 'broad approach reviews', however, has a common goal: to enlighten a broader study topic rather than to answer a narrowly focused subject. Following that, the approaches differ in part to address the nature of the research question, research objectives, issue area, the depth required for data extraction and the expertise of the review team.

I propose that a useful distinction is to see mapping, scoping and EGMs sitting within the same family of types addressing broad questions, but sitting on a spectrum in some of their underpinning epistemologies, concepts and hence objectives. This is illustrated in the Figure 2 on the following page.

	Scoping Reviews	Mapping Reviews	Evidence and Gap Maps (EGMs)
Purpose	Clarifies and identifies key concepts/definitions, characteristics or factors related to a concept.	Collates, describes, and catalogues the available evidence related to the question of interest.	Systematic evidence synthesis product which visually displays the available evidence and identifies research gaps relevant to a specific research question.
Question	Narrow focus to a broad question; what are the definitions for a particular concept?	Broad question: what do we know about a topic?	Very broad question, includes all relevant evidence of a specified kind for a particular question
Evidence Source	Identifies and maps evidence irrespective of source.	Identifies and maps evidence irrespective of source. Generally > 80+ studies	Identifies and maps evidence irrespective of source. Generally > 80+ studies
Extraction	Extensive and detailed data extractions	High-level with pre-defined codes for extraction.	High-level with pre-defined codes for data extraction
Analysis	Inductive (needs to be developed) or deductive (pre-determined) analysis (may include basic qualitative content analysis)	Deductive summary of high-level data with pre-defined codes.	Deductive summary of high-level data dependent on framework.
Presentation of results	Visual summaries must be accompanied by a descriptive synthesis	Visual summaries with or without EGMs	Visual, interactive online output placed on a web-based platform, such as a funders webpage.

Figure 2: Commonalities and Differences in Approaches in 'Big Picture' Reviews

Scoping review: A transparent, rigorous and systematic approach to identifying, describing and cataloguing literature available for a particular topic, field, concept or issue. It may seek to identify key concepts, theories, sources of evidence. It is exploratory, not requiring an a *priori* set of codes in order to describe data and may draw upon diverse sources of information *(i.e. primary research, reviews, non-empirical evidence) within or across particular contexts.* The approach can be iterative, and either inductive or deductive (Munn et al, 2022). The nature of the 'cataloguing' and coding may be in response to what is found within the literature. Scoping reviews can also be used to identify concepts and clarify terms in the literature. In contrast to a mapping, review the process of coding is predefined. Within a scoping review, the data extracted may be textual and descriptive, allowing for example an analysis of concepts. It may include both predefined coding, and also exploration of themes (for example, Kelly-Blake et al, 2018) In contrast, along a continuum, mapping reviews address broader questions, use predefined coding and adopt less in depth data extraction.

Mapping Review: Mapping reviews are also a transparent, rigorous and systematic approach to identifying, describing and cataloguing evidence and evidence gaps in a broader topic area. They aim to collate, describe and catalogue the available evidence relating to the question of interest (James et al, 2016). They answer these types of questions; 'what do we know about a topic', or 'what and where research exists on a particular area'. A mapping review typically extracts only descriptive information about the studies and applies predefined codes ('surface view data' – see figure 4 in chapter 7). In this sense, they may be informed by an 'aggregative' logic. A mapping review may or may not be accompanied by an EGM, but provides visual summaries in the form of tables and graphs within the text. These types of review may well have broader focus than a scoping review, with more limited data extracted from the included papers, when compared with a scoping review.

Evidence Gap Maps

EGMs are described as a systematic presentation of all relevant evidence of a specified kind for a particular sector, subsector or geography (Snilstveit et al, 2017). EGMs are a systematic evidence synthesis product which displays the available evidence relevant to a

specific research question. EGMs consist of primary dimensions, or a framework (rows and columns), and secondary dimensions or filters, enabling exploration of the map using a particular focus (e.g. looking at particular populations or study designs). It creates a visual, web-based, interactive output (White et al, 2020).

This type of evidence synthesis uses a deductive approach with a pre-specified framework to classify the data and identify gaps in the literature. This is one of the major differences between mapping with an EGM and scoping reviews. For the latter, either an inductive or deductive approach may be used to identify relevant data elements, so the framework for classification of the data and identification of gaps does not need to be pre-specified. Evidence gap maps may accompany a mapping review as a visual representation of the included studies or can stand independently from an accompanying mapping review.

The defining feature of this 'subgroup' within the family of evidence synthesis review types, is the addressing of a broad research question and objectives. They adhere to the principles of rigour and transparency that give users of evidence synthesis confidence in the reliability of the results of the review.

While the literature is inconsistent in its definitions of these types of reviews, and different reviews use different terminology to describe methods that appear very similar, many of these differences reflect the different research traditions and adoption of terms within organisations undertaking these types of syntheses.

I argue (**Paper A**) that there is value in having these distinct terms to describe the different approaches within this group of review types. Table 6 illustrates the different aims of scoping, mapping and EGM reviews. Scoping reviews allow an inductive, in-depth approach to open questions, usually including fewer studies and a greater level of data extraction. Both mapping reviews and EGMs, address closed questions, with pre-specified items defined and able to be coded. EGMs offer a visual, interactive output for users to locate evidence. Their predefined framework offers a rigour to locating gaps in the existing literature and displaying these differences, which is unique to these approaches.

Existing guidance for the conduct and reporting of scoping reviews, also applies to mapping reviews (Peters et al, 2022). Further development is needed in the methods of preparing a coding framework, particularly when the mapping review includes development of an interactive EGM. Current models of good practice exist, though current guidance and reporting standards are absent.

Table 6: Examples of Review Aims

Type of approach	**Aim**
Scoping review	To report in detail the methodology employed to identify relevant theories and provide a list of agreed criteria for judging the quality of theories(Davies and Barnett, 2015) To document and describe the evidence base relating to stakeholder involvement in systematic reviews and to use this evidence to describe how stakeholders have been involved in systematic reviews(Pollock et al, 2018)
Mapping Review	To review empirical evaluations of individual-level interventions intended to improve mental health or well-being for vulnerable adolescents.(Vojt et al, 2018) A mapping review of research on gambling harm in three regulatory environments (Baxter et al, 2019)
EGMs	To identify what has been published on micronutrients and depression and identify gaps in the evidence and collections suitable for meta-analysis (Campisi et al, 2020). Identify and map the available evidence on the effects of food systems interventions on food security and nutrition outcomes in low- and middle-income countries (L&MICs); and Identify potential primary and synthesis evidence gaps (Moore et al, 2021)

Chapter Five: Stakeholder engagement

Paper F
Campbell, F., Whear, R., Rogers, M., Sutton, A., Robinson-Carter, E., Barlow, J., Sharpe, R., Cohen, S., Wolstenholme, L. and Thompson-Coon, J., 2023b. Non-familial intergenerational interventions and their impact on social and mental wellbeing of both younger and older people—A mapping review and evidence and gap map. Campbell Systematic Reviews, 19(1), p.e1306.

Critical Analysis

As researchers, there is both a moral and legal imperative to empower those influenced by our research to have a stake in the conduct of that research, and that imperative is enshrined in policy (NIHR, 2018; Denegri et al, 2015; Richards, 2017). Stakeholders are those who directly or indirectly impact the research or are impacted by its findings or the challenge the research seeks to explore (Oliver et al, 2021). Stakeholder engagement ensures that research addresses the challenges that matter, in ways that are acceptable, reduces research waste, improves translation of research into policy and practice and ultimately leads to improved benefits for society, systems and individuals. (Wiles et al, 2022; Brett et al, 2010; Shippee et al, 2015).

However, there is, uncertainty about how it should be best undertaken and in a manner that leads to genuine partnerships involving a true diversity of stakeholders rather than a few selected individuals (Hubbard et al, 2007). There are also suggestions that power inequities and discrimination have not been adequately prevented which has led to criticism of exclusivity and tokenism (Ocloo et al, 2021). There is a lack of evidence about how stakeholder involvement has changed reviews, and still limited evidence of how it brings benefits in evidence synthesis outputs and dissemination. There is also a lack of consistency in the use of language or standard framework to guide practice (Shippee et al, 2013; Wiles et al, 2022) or understanding how it has impacted the research (Morley et al, 2016).

Much of the critique and guidance regarding stakeholder engagement focuses on its value in primary research, but there is a growing body of research exploring its role in evidence synthesis and resources to support it (Langer et al, 2020; Haddaway, 2019). A recent review

(Agyei-Manu et al, 2023) recommended that future research should focus on using existing frameworks to help describe and/or report the best approaches and methods for stakeholder engagement in evidence synthesis.

The role of stakeholder engagement in conducting scoping reviews receives brief mention in the recent updated JBI guidance (Peters et al, 2020) . Arksey and O'Malley (2005) describe consulting with stakeholders as an optional final step regarding the review findings. Tricco et al (2016) found that only 6% of 494 scoping reviews included in their review included some form of knowledge user engagement. This appears to be shifting with recent publications advocating stakeholder engagement in scoping reviews and aspiration for a model of co-creation and not merely consultation (Pollock et al, 2022). James and Haddaway (2016) recommended the involvement of 'relevant' stakeholders who should be consulted for their expertise to help shape the scope and ensure the relevance of the systematic map. Relevant stakeholders included: review commissioners; policy makers; practitioners; non-governmental organisations; levy boards; scientists and research funding bodies (no non-professional users were listed). James and Haddaway (2016) warn that stakeholders may have strongly vested interests in the topic and care must be taken to avoid any resultant bias to the Big Picture review process. Clearly a model of 'co-creation' is difficult to align with one in which stakeholders are an asset but also a risk to the validity of the review findings. However, there remain, few examples of how stakeholder engagement is operationalised and how it impacts the findings in scoping and mapping reviews.

Guidance for evidence and gap maps makes the need for stakeholder engagement clear in the development of the matrix for the EGM. White et al (2020) describes stakeholder consultation as important in determining the scope of the map, developing the framework, and interpreting the findings. However, they also urge caution, "stakeholder consultation will often create pressure for more categories as they want to see "their interventions" named. But this pressure needs to be weighed against the disadvantages of a cumbersome framework" (pg1). The Campbell guidance is clear on when to involve stakeholders, but does not give clear guidance on how this should be undertaken. Miake-Lye et al's (2016) review of evidence maps does not explore the role of stakeholder engagement in the

development of evidence maps, so again there are few examples of good practice that can be drawn upon and, where stakeholders are included, the processes are poorly reported.

It was against this background of limited examples and guidance, which advocated for, but also warned of the potential negative effects of stakeholder engagement, that we undertook our evidence and gap map.

Critical Reflection

In July 2021, I led the Sheffield arm of a programme of work looking at intergenerational programmes and how they impact children and/or adolescents and older people's health and wellbeing, as well as their impact on the communities in which they are delivered. Intergenerational programmes and activities can take many forms and are delivered in many settings, very often by third sector organisations (**Paper F**). Although evidence suggests that intergenerational activity can have a positive impact on participants (e.g. reducing loneliness and exclusion for both older people and children and young people, improving mental health, increasing mutual understanding and addressing important issues such as ageism) commissioning decisions are complex due to the apparent wealth of options but limited and varying resources with which to provide them. There is also a lack of evidence about their effectiveness, transferability of effects across settings, and cost-effectiveness.

The objectives of the review are described in detail in **paper F**, but in summary, we aimed to identify and bring together the evidence on; the use of intergenerational practice, identifying the nature of the evidence, the approaches used in intergenerational practice and gaps in the evidence.

As part of this project our team undertook an EGM and two systematic reviews. This chapter focuses on involvement of stakeholder's EGM (**Paper F**).

There are numerous frameworks for supporting, evaluating and reporting stakeholder engagement in research, though this literature is diverse and theoretically heterogeneous (Greenhalgh et al, 2019). For the purposes of this chapter, I am using the Six-Step

Stakeholder Engagement Framework (Tomlinson & Parker, 2021) because of its focus on stakeholder engagement in the context of systematic reviews. I am applying this retrospectively; our team did not use the framework to guide our practice. The framework consists of six steps, spanning the process of stakeholder engagement from the planning of engagement to its evaluation and maintenance (see Figure 3).

Figure 3: Six-Step Stakeholder Engagement Framework (Tomlinson and Parker 2021)

Step 1: Be clear about the purpose for stakeholder engagement

Involvement of stakeholders can occur at any stage in a systematic review, and the ACTIVE framework uses a simplified categorization of the review process to assist with locating where stakeholder engagement can occur. Some of these can be anticipated, but others

might arise during the review process itself (Tomlinson& Parker, 2021). Our team planned and described the areas where stakeholder engagement would inform our EGM and mapping review. These included the following:

- Including stakeholders in the development of the research project at the outset, creating an advisory board and inviting stakeholders to be named as co-applicants and co-authors on our grant application.
- Ensuring the research questions were priority areas.
- Reviewing and co-authoring the protocol.
- Informing the search strategy by identifying and reviewing search terms.
- Providing key texts.
- Knowledge of relevant websites and sources of grey literature.
- Informing the creation of the framework (rows and columns) and filters for the EGM.
- Identifying the priority questions for the systematic review.
- Identifying conferences, organisations, websites, and key contacts to ensure our findings and the map could be widely disseminated.
- Contributing to plain English summaries.

Step 2: Reflect on previous stakeholder engagement and consider capacity

Previous stakeholder engagement activity had increased my awareness of the challenges of including children and young people, including: identifying potential stakeholders; safeguarding; costs; and adapting material for a wide age range. .As researchers, where our research will impact children we have an ethical responsibility to ensure they are treated with equity and that barriers to their involvement do not prevent them from being included (International Charter for Ethical Research involving Children 2022) (Powell, 2016). In order to ensure that children and young people were engaged in our review we recruited CHILYPEP, a Children and Young People's Empowerment Project, dedicated to raising the voices of young people, to the project. I knew that this would involve additional costs and built this into my funding application.

Step 3: Identify relevant stakeholders

The stakeholders included in this programme of research were identified and selected based on their expertise or experience. We involved our stakeholders in the process of identifying other key stakeholders. I have categorised our stakeholders using the Cochrane Knowledge Translation Framework (Cochrane 2017).

Consumers and the public: those participating or eligible to participate in an intergenerational intervention.

Practitioners: of health or social care, in this instance those providing or facilitating intergenerational interventions.

Policymakers and healthcare managers: individuals and organisations responsible for purchasing social care interventions and making higher level decisions about social care availability and advice.

Researchers and research funders: those involved in designing, conducting, commissioning and carrying out research.

Table 7: The IGEN Stakeholder group

Consumers	Ronald Amanze Laura Abbott—CHILYPEP – representing children and young people Ellie Robinson-Carter Peter Daniels
Practitioners	Aideen Young - Centre for Ageing Better Sally Pearse—Sheffield University. Members of the 'Only Connect!' The group includes local, national and international members from the care sector, local government, academia, people living with dementia, schools and leading organisations involved in providing intergenerational activities. Members of the group also facilitated discussion of the project with older people, people living with dementia, and young people with experience of taking part in intergenerational activities. Girish Vaidya—Sheffield Children's NHS Foundation Trust;
Policy makers and managers	Kelvin Yates—AgeUK Cornwall; Debbie Hanson—Sheffield City Council; Rachel Staniforth—Public Health Kerry Albright—Unicef;
Researchers	**methods:** G.J. Melendez Torres—University of Exeter; Dylan Kneale—UCL; Ruth Garside—University of Exeter; **older people:** Claire Goodman—University of Hertfordshire; Iain Lang—University of Exeter; Vicki Goodwin—University of Exeter; Jo Day—University of Exeter; Tracey Howe—Cochrane Campbell Global Ageing Partnership **children:** Nathan Hughes—University of Sheffield; Hannah Fairbrother—University of Sheffield

Step 4: Connect with stakeholders

Our team convened three virtual whole project meetings to include stakeholder members, which assisted with understanding and presentation of the evidence in the EGM. On-line break-out rooms and other methods of sharing ideas and suggestions such as a JamBoard, were used to ensure that as many views and perspectives were captured as possible. Large meetings were followed up with smaller meetings and/or phone calls where necessary. Between meetings, people were involved through email, telephone and video conferencing, depending on the nature of the involvement and the preference of individuals.

As described above, stakeholders played a very significant role in designing the matrix that forms the framework for the EGM. The resultant framework then informs our coding (data extraction) form and shapes the entire output of the review. In order to engage our stakeholders in the process of designing the map, I needed to ensure they had a good understanding of what an EGM was. Our meetings required careful preparation to ensure stakeholders were empowered with the knowledge they needed in order to engage with the process of creating the matrix for the map. Our team used small group work as well as presentations to achieve this. The resulting framework included the use of Kaplan's levels of engagement(Kaplan, 2004), which was a direct result of our stakeholder engagement.

Step 5: Report stakeholder engagement

Our stakeholder engagement was documented in both our protocol and published review. However, going forwards this could be improved by adopting the GRIPP 2 guidance for reporting stakeholder engagement (Staniszewska et al, 2017) and providing an in-depth description of our processes and the impact stakeholders made on shaping the review.

Step 6: Evaluate and maintain stakeholder relationships

These are some lessons learned that I will take forwards into future reviews.

Stakeholder engagement led to the adoption of a matrix for our EGM that we would not have envisaged without their involvement. It led to a significant impact of our work on the

uptake of our findings and the utilisation of the output of this review. Having presented our map and described our findings at the 'Only Connect' conference in 2021, our team received invitations from groups working in the field of Intergenerational Practice in Scotland, Australia, Spain and the USA. The map is hosted by 'Generations Together', Scotland. This embracing of the review findings and the EGM is a direct result of stakeholder engagement.

However, there are lessons to learn. The level of stakeholder engagement was not consistent from all stakeholders. Early engagement was not maintained, by some stakeholders, while others became more engaged. This was particularly so for our children and young people. Although accessibility was addressed by running meetings at different times, attendance waned during the project from some of our stakeholders. Stakeholders were kept informed of progress via newsletters, podcasts, emails and presentations, no 'in-person' events that may have reduced the ability to build relationships.

I have applied the Six-Step Engagement (Tomlinson E, 2021) retrospectively but it would have been more useful to have incorporated this into our review at the protocol stage.

Critical Action

I have described in this chapter the process of stakeholder engagement that was adopted for our EGM (**Paper F**) and reflected on both the limitations and the positive impact of our methods. Stakeholder engagement has been described as occurring at different levels from a level where engagement with stakeholders consists of stakeholders being given information (minimal) but with no role in contributing, to co-production where stakeholders are equal partners in the research (Oliver et al, 2008, Pollock et al, 2019). Petkovic et al (2023) further operationalised these levels into two categories: 'advice/feedback' and 'decision making'. My own progress in learning to meaningfully undertake stakeholder engagement reflects shifts we have seen within our field. Co-production in research as a goal does not sit comfortably with the 'optional' involvement of stakeholders or suspicions about their influence which are embedded in existing guidance. My earlier work (**Papers C and E**) included stakeholder involvement, but the depth of engagement varied considerably between stakeholders, and would be best

classified as ‘advice/feedback’. **Paper F** marks a development towards ‘decision making’, however, it remains an area where I am continuing to explore our methods. As highlighted by my experiences in the Intergenerational Interventions review (**Paper F**) areas of further methodological research I see as priorities are; improving our approaches to engaging children and young people; adapting our methods for the needs of the very different groups labelled as ‘stakeholders’; and developing educational tools to facilitate engagement and involvement, particularly in EGMs.

Chapter Six: Locating Relevant Evidence

Paper C
Campbell, F., Salam, S., Sutton, A., Jayasooriya, S.M., Mitchell, C., Amabebe, E., Balen, J., Gillespie, B.M., Parris, K., Soma-Pillay, P. and Chauke, L., 2022a. Interventions for the prevention of spontaneous preterm birth: a scoping review of systematic reviews. BMJ open, 12(5), p.e052576.

Paper D
Sutton, A. and **Campbell, F**., 2022b. The ScHARR LMIC filter: Adapting a low-and middle-income countries geographic search filter to identify studies on preterm birth prevention and management. Research Synthesis Methods, 13(4), pp.447-456.

Critical Analysis

The aims and purposes of the review will inform the methods used in searching for the evidence. Searches may seek to be 'exhaustive', and locate all potentially relevant research, particularly when the purpose of the review is unbiased aggregation. However, for reviews that aim to generate theory or seek important themes, the aim of searching may be 'purposive', seeking to locate exemplars to provide sufficient breadth and representation to address the question that underpins the review (Brunton et al, 2017). The aim of all reviewers is to avoid the potential for selection bias in the search, which risks trustworthy findings. Critical to high quality and reliable review findings is the involvement of information specialists within the review to lead the process of locating the evidence (Rethlefsen et al, 2015, Aamodt et al, 2019, Meert et al, 2016; Metzendorf, 2016).

In Big Picture reviews, the emphasis is on being as exhaustive as possible (White et al, 2021, James et al, 2016, Peters et al, 2015, 2020 White et al, 2020) but also with the caveat that it must also be manageable (Arksey and O'Malley, 2005, Snilstveit et al, 2016). Discerning what will need to be done to make the review 'manageable' will not be entirely known at the outset of the review, and accounts for the often more 'iterative' processes that a Big Picture review might include. Sager and Pistone's (2019) paper is helpful in exploring the 'iteration' that they undertook in their scoping review.

The selection of broad terms and less clear boundaries of relevant evidence can result in very big record sets, which are costly in terms of the time resource needed to screen

results. Screening the results of a search to identify relevant studies is time consuming (taking one researcher a day to screen between 500-800 titles and abstract) and at full paper stage (800 per day) (Haddaway and Westgate, 2019). Managing large sets of search results, particularly within a rapid review context, or where there are resource limitations requires pragmatic decisions. Collaborative discussions between commissioners, reviewers, stakeholders and information specialists are usually necessary to consider ways that the search yield can be made more manageable (Pandor et al, 2019).

Critical Reflection

A particular challenge for Big Picture reviews is that database searches can often yield a large number of titles and abstracts that require screening, ideally by two reviewers blind to the results of the other with any discrepancies resolved by discussion (Shemilt et al, 2014).

Ways in which a review team can address this challenge involves close involvement of the information specialist and include (stage one) approaches during the database searches and (stage two) approaches during the screening of titles and abstracts.

- Stage one approaches include applying restrictions and developing search filters.
- Stage two approaches include machine learning, or rapid approaches which require a modification of an accepted methodological approach, such as single reviewer screening or partial checking of screening agreement between reviewers.

Stage one approaches

Search strategies might include restrictions that limit the retrieval of papers not published in English, publications outside of particular date specifications or by document format (Lefebvre et al, 2022). These might be approaches considered when there is a limit on time or resources, or when the search yield is particularly high. Whenever a limitation is imposed, it needs to be justified to avoid risk of bias. For example, a date limitation should not be arbitrary but based on a known factor that might mean that search yields prior to a particular date are less relevant in meeting the review objectives. Various date fields are also made available by database providers, (such as the create date, last update date,

publication date). The inclusion of non-English studies is recommended to minimise the risk of language bias, but where this is justified, it is recommended that this is not imposed by limiting the search but by including language as an eligibility criteria during study selection (Pieper and Puljak, 2021).

Search filters are pre-made search strategies designed to retrieve particular types of study designs, or topics from bibliographic databases. The use of search filters can reduce the search yield by identifying studies with appropriate study designs. For example, filters exist for identifying systematic reviews, randomised and non-randomised studies and qualitative research across several databases (Glanville et al, 2019).

Stage two approaches

Another route to managing the large yield in configurative reviews is the use of machine learning and/or text mining. Identifying software to support the study selection process can be identified using the Systematic Review Toolbox (Marshall C, 2022). Currently 46 software tools (accessed June 1, 2023) have been identified that can assist at the screening stage of a review. The use of machine learning and automated tools has been shown to reduce the workload involved with selecting studies significantly (Thomas et al 2017). Adopting automation can reduce the need for manual screening by at least 30% and possible more that 90% (Shemilt et al., 2014). There can be cost of up to a 5% reduction in sensitivity (O'Mara-Eves et al, 2015), with a small proportion of relevant studies being missed.

Another approach that might be adopted, and one that is often used in rapid reviews (Abou-Setta et al, 2016), is the use of single reviewer screening. It is recommended that two reviewers screen all titles and abstracts, to reduce the risk of relevant reports being discarded (Waffenschmidt et al, 2019). It may be that a more nuanced approach is needed in making decisions about how to undertake this stage of the review. It may be that in some types of reviews, it is easier to overlook potentially relevant studies. An example might be where much of the literature is 'low level' evidence, and establishing study design is difficult. It may be a field where terminology is ill defined. Another factor, and one that is particularly relevant to configurative reviews, is that missing a relevant study may not

have the same significance in terms of the purpose of the review than when the purpose is to synthesise the included studies. In the context of a meta-analysis, missing a relevant study may have implications for the review findings. In a large mapping review that seeks to identify trends and patterns in the use of terms, for example, missing a relevant study is less likely to influence the overall findings of the paper. These more nuanced factors may inform decisions on whether two reviewers should independently screen all the search results.

One step that is particularly important however, is the need to test the inclusion and exclusion criteria. This task to be undertaken by more than one member of the review team and allows ambiguity in the terms, and judgements regarding inclusion and exclusion to be tested and discussed.

Critical Action

In 2018, I led a team undertaking a mapping review of interventions delivered in low and middle-income country (LMIC) settings to prevent spontaneous preterm birth (PTB) (**Paper C**). The research was commissioned by the National Institute for Health Research (NIHR), funded Global Health Research Group on Preterm birth prevention and management (PRIME). PRIME brought together a group of interdisciplinary researchers form the UK, South Africa, and Bangladesh to address the challenges of PTB in LMICs, where its prevalence is highest (Chawanpaiboon et al, 2014). The mapping review aimed to identify and describe the quantity and quality of evidence that have sought to explore the effectiveness, safety and acceptability of interventions to prevent PTB. Based on the existing evidence, the review would identify research gaps in LMIC contexts to inform future research and identify areas for potential further research synthesis. Search results for scoping reviews looking at maternal health interventions in LMIC contexts, typically find yields of between 45,000 to 50,000 title and abstracts (Chersich et al, 2016). This equates to approximately four months full time work for two reviewers to complete the screening with rigour.

For the mapping review, it was particularly important to identify literature relating to LMICs in this context, as some interventions that have been used in developed world contexts

may be harmful in LMIC settings and conversely there may be interventions that are even more effective in LMICs than in other settings (Jobe et al, 2019). Therefore, our team sought to use an LMIC geographic search filter to ensure the results of our search were relevant to the LMIC setting. The Cochrane Effective Practice and Organisational Care (EPOC) Group's LMIC geographic search filter was selected. The filter has several versions available for different databases: Ovid MEDLINE< PubMED, Ovid EMbase and CENTRAL.

During the searches, our team found that the Cochrane EPOC LMIC geographic search filter did not retrieve a known study of interest. The reasons why this indexed study had not been retrieved with the Cochrane EPOC LMIC geographic search filter were explored. We found that our missing study contained no MeSH headings relating to LMICs; neither the generic 'Developing Countries' MeSH heading nor any specifically named countries were present. India was mentioned, though only in the address (institution) of one of the authors. In the Cochrane EPOC LMIC geographic search filter, the countries are searched for in title, abstract, country of publication, headings, or author keywords, so 'institution' was added to our version of the filter. Possible adaptations to the filter were investigated to ensure this study was retrieved and potentially further relevant studies (**Paper D**).

The resulting 'ScHARR LMIC filter' is a non-validated first generation geographic search filter and is listed on the ISSG website, has been used in other published systematic reviews (Lam et al, 2023, Karamagi et al, 2023) and included by methodological guidance (https://epoc.cochrane.org/lmic-filters). The development of a search filter is a particularly valuable additional tool for use in Big Picture reviews. Frequently Big Picture reviews seek to explore and demonstrate geographical coverage of studies within a topic area. This tool will greatly enhance the efficiency of the review process for those reviews where a focus on LMICs is necessary.

Chapter Seven: Making sense of the data

Paper B
Campbell, F., Weeks, L., Booth, A., Kaunelis,D., Smith, A., 2019a. A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance. *Journal of clinical epidemiology*, 115:pp160-71.

Paper C
Campbell, F., Salam, S., Sutton, A., Jayasoonya, S.M., Mitchell, C., Amabebe, E., Balen, J., Gillespie BM., Parrisy K., Soma-Pillay P., Chauke L., Narice B., Anumba, D.O., 2022a. Spontaneous Preterm Birth Prevention. A Scoping Review Highlighting an Inverse Pattern of Research with a Lack of Research Evidence from High Burden Settings. *BMJ Open,* May

Paper E
Foster, C.R., **Campbell, F**., Blank, L., Cantrell, A.J., Black, M. and Lee, A.C., 2021. A scoping review of the experience of implementing population testing for SARS-CoV-2. Public health, 198, pp.22-29.

Paper F
Campbell. F., Whear, R., Thompson-Coon, J., Sutton, A., Rogers. M., Barlow. J., Robinson Carter, E., Sharpe, R., Chhen, S., Wolstenholme, L., 2023b Intergenerational interventions and their effect on social and mental wellbeing of both children and older people – A mapping review and evidence and gap map. *Campbell Systematic Reviews*, *18* (2) May

Critical Analysis

The work of turning the results of the screening process and resultant studies into something that provides a useful description of the evidence feels akin to unravelling a large, tangled knot of wool of various colours into something that can then be usefully reworked into a beautiful Fair Isle blanket. The quality of the final research output depends largely on the excellence of the process of making sense of the data, which must **be clear, accurate, in relevant format, appropriate, rigorous and systematic (Sutcliffe et al., 2017).**

Achieving Accuracy and Rigour

As is any scientific endeavour, researchers bring their own biases, and this applies to scoping, mapping and EGMs as in any other approach to evidence synthesis.

"Our own subjective ways of looking at the world may lead us, perhaps unwittingly, to producing an accurate but skewed representation of a research field. Just as cartographers have used a number of different approaches for producing a flat paper-based representation of the globe, so reviewers privilege different dimensions when mapping their field" (Sutcliffe pg. 136).

Recommended guidance (Li et al, 2022) advocates that the process is undertaken independently, by two reviewers and that the results are compared, with differences identified and resolved. This is also recommended in guidance for Big Picture reviews (White et al, 2020; Peters et al, 2020). Other measures to ensure the process is clear and appropriate include development and piloting of tools that reviewers will use for this purpose. A range of software might be used, to facilitate the process of checking reviewer decisions or support the presentation of the results (examples include EPPI-reviewer (Thomas et al, 2022), Rayyan (Ouzzani et al, 2016) and Covidence (Veritas Health Innovation, 2023)

Ensuring appropriateness and clarity

Which details are extracted and coded depends is driven by the review objectives and questions (Pollock et al, 2023). The information will include bibliographic (e.g. authors), management (e.g. on request), process (decisions regarding exclusion) and substantive (e.g. study design, findings, condition) information. All reviews will extract bibliographic, management and process details. However, the nature and depth of the substantive data extracted/coded varies considerably between Big Picture reviews and other reviews. I contrast these two approaches as 'surface view' (sometimes called 'high level') and a 'deep dive' data extraction characteristics of systematic reviews, where integration and synthesis of the study results is the objective.

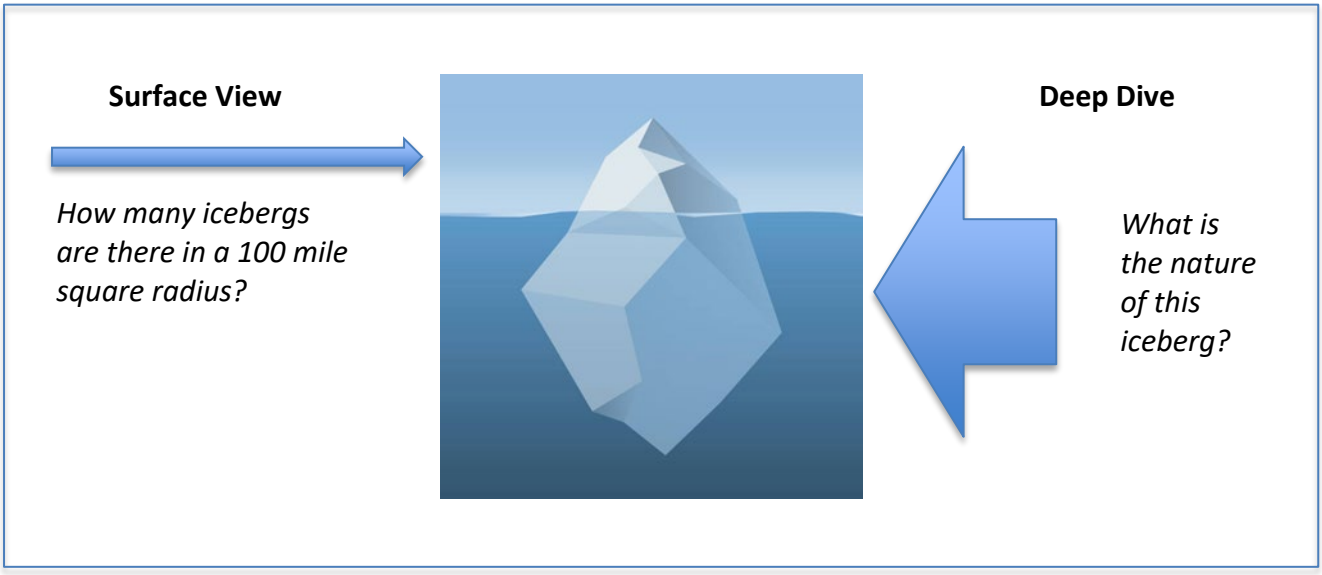

Figure 4: Surface View versus Deep Dive Approaches to Data Extraction

Critical Reflection

However, even in Big Picture reviews differences exist in the depth of data that is extracted/coded and I suggest this can be viewed as a continuum (see Figure 5), with scoping reviews extracting more in-depth data, such as how the role of an epilepsy nurse specialist is described (**Campbell et al, 2019b; Campbell et al in press B**) to 'surface view' data extraction, such as whether a study included an evaluation of mental wellbeing in children (**Paper F**). Data extraction/coding tools are likely to have a combination of 'surface view' and 'deep dive' details, but the presence of one deep dive question will move it along the continuum towards a 'deep dive' classification.

Data extraction might be iterative in Big Picture reviews (**Paper A**; Pollock et al, 2023; Bradbury-Jones et al, 2019), with refinements occurring during the data extraction/coding process, and new categories being added as the included studies are reviewed.

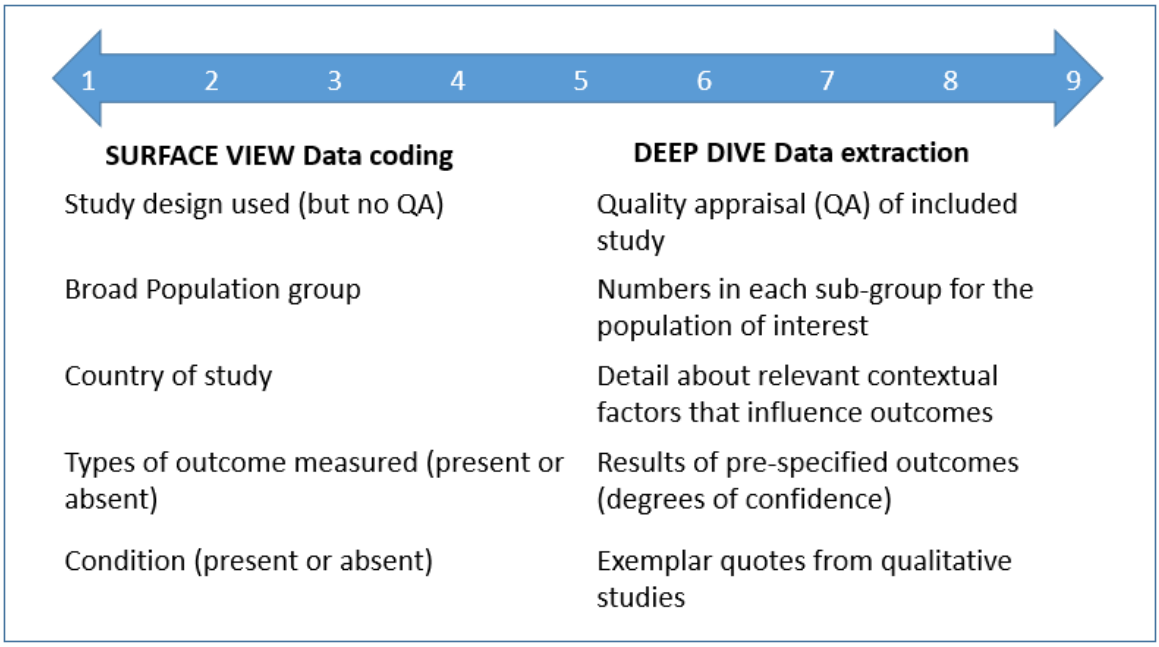

Figure 5: The Continuum from Surface View versus Deep Dive Approaches to Data Extraction

Critical Action

The task of data extraction/coding in the case studies included in this thesis varied in the number of studies that were included in reviews, ranging from 15 to 500, and in the depth of detail that was required in each case. The task was also influenced by the resources available in terms of the team size, expertise within the team and the use of software to support management of the process.

The depth of detail that needs to be extracted will have implications for the time resource needs of the review. For example in one review examining the role of the epilepsy nurse specialist (**Campbell et al 2019b and in press B**) our team extracted details on the nature of the epilepsy nurse specialist role. In another review (**Paper B**) we looked in depth at the methods used as described within the paper. In both cases, the time spent in each included paper was considerably longer than in the case study reviews that focused on intergenerational practice, and elder abuse, where the information we needed was 'surface view' and easy to locate within the paper.

The methods of designing our pro forma also differed. In the case study looking at the role of epilepsy nurse specialists we began with an initial conceptual framework drawn from the literature (Epilepsy Action, 2010) which was then tested, clarified, and extended during the process of the review. In contrast, in the review looking at preterm birth, intergenerational interventions and elder abuse, the data coding frameworks were developed and tested with the aim of no changes occurring during the data coding process. Any changes risked the teams having to repeat the process once again.

Table 8: The task of data extraction/coding

Review	**Number of Included Studies and study type**	**On Deep Dive – Surface View Continuum**	**Resources (team – resource for data extraction) (software used)**
Qualitative evidence synthesis methods (**Paper B**)	n=15 qualitative evidence synthesis	5	2 reviewers data extraction, 3rd reviewer to review differences Excel (Microsoft, 2018) data extraction sheet
Role of epilepsy nurse specialist (**Campbell** et al in pressB and **Campbell** et al, 2019b)	n=118 any study design	4	2 reviewers data extraction 3rd reviewer to review differences Excel (Microsoft, 2018)data extraction sheet
Preterm birth (**Paper C**)	n=139 systematic reviews and 1372 primary studies	3	6 reviewers EPPI-reviewer (Thomas et al, 2022)
Elder abuse (Mikton et al 2022)	n=111 systematic reviews	3	6 reviewers EPPI-reviewer (Thomas et al, 2022)
Intergenerational Interventions (**Paper F**)	n=500 any study design	2	4 reviewers EPPI-reviewer (Thomas et al, 2022)
Mass Screening COVID-19 (**Paper E**)	n=22 any study	5	2 reviewers 3rd reviewer to review differences Excel (Microsoft, 2018) data extraction sheet

Team working is an often-overlooked critical element in the successful completion of a review, particularly when there are tight time frames to work within, which was the case in our case study looking at mass screening. Indeed, the need for good team working is something I believe is not only critical to the successful completion of the commissioned task but also the wellbeing of individuals within the team. Such is its importance that I have built good team working practices into the short course I developed and co-lead with Andrew Booth on Rapid Review Methods.

The process of data extraction/coding requires good management of the included studies, the data and workload allocation, as well as managing the checking and resulting

corrections. In two of the case studies (preterm birth and elder abuse), I was working with large international teams. The challenges of managing large teams of inexperienced reviewers were considerable and my role involved training the team as well as undertaking the review. In order to improve the management of the task, I used EPPI-Reviewer (Thomas, et al, 2018) a software designed to support the review (Action, 2010), which facilitated the process of managing a team, allocating extraction/coding and identifying and correcting differences.

Often due to the size of the coding task, and limited resources, the process of data extraction or coding in scoping reviews is undertaken by a single reviewer (Tricco et al, 2016). A practical challenge was that errors and differences in coding decisions appeared frequently, even in surface level coding, with team members missing codes or miscoding. All of the coding was therefore undertaken in duplicate with reviewers blind to the decision of another reviewer.

One of the most time consuming data extraction case studies (preterm birth) involved tabulating all of the included studies in each review, and the setting in which they were undertaken. Nevertheless, it allowed us to present an original paper, showing that only 3% of all studies undertaken to test treatment effectiveness are undertaken in those setting where preterm rates are highest (**Paper C**). This work has been used to support awareness of the absence of interventions that are effective in reducing the risk of preterm birth, the greatest cause of death and disability in children globally (Cohen 2023). A further important finding in this review was how frequently information about study populations and settings was missing, or ignored in the analysis and synthesis by systematic reviewers.

I have shown how the process of data extraction/coding is, in the reality of Big Picture reviews a 'swampy lowland' where the methods and processes are tailored differently for each review depending on; the topic area, review objectives, size of the included evidence base, resources available, needs of the team, and the resources available.

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Chapter Eight: Creating an Accessible Evidence Resource

Paper F
Campbell, F., Whear, R., Rogers, M., Sutton, A., Robinson-Carter, E., Barlow, J., Sharpe, R., Cohen, S., Wolstenholme, L. and Thompson-Coon, J., 2023b. Non-familial intergenerational interventions and their impact on social and mental wellbeing of both younger and older people—A mapping review and evidence and gap map. Campbell Systematic Reviews, 19(1), p.e1306.

Paper G
Indave, B.I., Colling, R., Campbell, F., Tan, P.H. and Cree, I.A., 2022. Evidence-levels in pathology for informing the WHO classification of tumours. Histopathology, 81(4), pp.420-425.

Critical Analysis

Many factors influence if, and how evidence informs decision-making. These factors are numerous and relate to the nature of the evidence itself, how it is produced and disseminated, and to policy or practice contexts. These factors include limited access to research, lack of timely findings, mismatch between research and research user timelines, low research user skills, costs of better engagement, resistance to change and lack of political will; and, vested and conflicts of interest. A considerable body of work that has explored these in depth in wide ranging fields and policy areas including. Nursing (Hannes et al, 2007; Retsas, 2000), education (Gorard et al, 2020; Thomas et al, 2019), policy (Andermann et al, 2016; Oliver et al., 2014), medicine (Sadeghi-Bazargani et al, 2014; Zwolsman et al, 2012) program management (Spallek et al, 2010 Cross et al, 2023; Humphries et al 2014) health care managers (Barends et al, 2017; Tricco et al, 2015), environmental policy and practice (Hofmann et al, 2022) and child health (Zdunek et al, 2021) are just some examples.

Recommendations for the ways in which researchers can therefore aid the uptake of evidence into policy are also numerous, though the evidence evaluating them is largely weak (Cairney et al, 2023; Oliver and Cairney, 2019). Donnelly et al (2018) has described four features that make evidence synthesis more useful for policy, it must be; inclusive, rigorous, transparent and accessible. Cairney (2016) argues that those who produce evidence need to better understand policy-making contexts and highlight the importance

of ‘relational’ interventions and the need to build relationships and trust in order to influence change.

Thus, the first step in being able to use research evidence for improving population health is ensuring that evidence is available at the right time and in the right format and language so knowledge users can take the evidence into consideration alongside a multitude of other factors that also influence decision-making. What is more likely to work for both policy and practice is the engineering of high quality evidence into a more usable format and presenting it actively or iteratively via a trusted conduit.

Clear guidance exists to support the reporting of evidence synthesis Page et al, 21). However, it is increasingly expected that reviewers will go beyond publishing their work in academic journals and seek to engage in more effective dissemination using a range of outputs (social media, blogs, policy briefs). This has been an area where my practice as a researcher has developed as I have sought ways to reach a wider audience, using for example, podcasts, animations and blogs.

Critical Reflection

In scoping and mapping reviews, presenting results and generating an evidence resource introduces challenges. Reviewers must describe, summarise, interpret, and seek to show patterns and findings - from what is often a large number of included studies and a large quantity of data. Tables and graphics are frequently used to ease interpretation and transfer messages visually as well as in an accompanied text (Pollock et al, 2023). The resulting evidence resource is a text-based report, usually supported by graphics. These review types do not provide a synthesis of findings but rather a descriptive and holistic picture of the evidence.

Evidence and gap maps differ from scoping and mapping reviews, being defined by their visual and interactive output, though the accompanying report is very similar to scoping or mapping reviews (**Paper F and E**). Like scoping and mapping reviews, EGMs are descriptive and do not include a synthesis of study findings. Their interactive features enable user’s direct access to summaries or abstracts of included studies. The graphical display provides visual information regarding the distribution of evidence and location of gaps.

Since 2010 there has been a steady increase in the number of EGMs published (Miake-Lye et al, 2016). It is the visual, interactive features of these types of approaches that I believe make them a particularly valuable addition to the range of evidence synthesis methods methodologists have at their disposal. They are a progressive step in terms of designing outputs that address some of the barriers in knowledge translation. They provide accessible, visual and interactive evidence resources. They also address one of the realities of policymaking environments, as decision makers are often asking multiple questions and looking for contextually relevant data. An interactive map addressing a broad topic, with filters that allow for a more tailored exploration of existing evidence may be a factor that is leading to the increased popularity of EGMs.

Critical Action

Of the included Big Picture reviews referenced in this thesis, five have included undertaking an EGM (**Paper C, F and G**). A further study (**Campbell et al., 2019a**) and **Campbell et al**. (in press B) also included undertaking an EGM. In this review, I led a team looking at the roles of epilepsy nurses specialists (ENS). Our commissioners (Epilepsy Action) wanted a creative tool that could be embedded on their website providing users access to relevant evidence to support commissioning decisions and also guide future research priorities. There were limited publicly available tools to create interactive visual tools and those that were available had limited adaptability. For example, the 3ie tool did not allow me to change the columns from 'outcomes' to elements of the ENS's role. As a result, we worked with a team of final year Computer Science students who developed a bespoke tool that allowed us to create an EGM which accompanied our report (see Figure 6)(**Campbell et al**, **2019b**, (**Campbell F, in press B**)).

The technology evolved quickly, however, and the EPPI Centre developed functions within EPPI Reviewer and an adjunct tool (EPPI Mapper) which I then used in undertaking subsequent EGMs.

With each subsequent map, I have been able to add additional features as the technology has advanced. This has included, for example, being able to embed information about the map, creating a template that makes generating the map less time-consuming and an

increase in the number of segmenting filters that can be used in the map. I have also increasingly found that the team working elements in managing coding across a large team greatly enhanced by the collaborative functions within EPPI Reviewer. Figure 8 shows a more recent map created in EPPI-Reviewer (Thomas et al, 2022) and EPPI-Mapper (Digital Solutions Foundry and EPPI Centre (2022), (**Paper F**). With each review, I learned ways to design the matrix and filters to ensure we were meeting the objectives of the review questions. This included, for example, identifying when and how we would use quality appraisal, where study design might serve as a better descriptive indicator for a body of knowledge and where the type of evidence synthesis was the most useful type of descriptor to use.

I was able to use EPPI-Reviewer (Thomas et al, 2022) and EPPI-Mapper (Digital Solutions Foundry and EPPI Centre, 2022) to create demonstration EGMs that then led to an international group of pathologists, epidemiologists and the WHO Tumour Classification team to successfully apply for an Horizon grant (101057127). I am supporting the consortia by guiding on the methods and training the review team. A demonstration of the types of EGMs that are emerging is shown in Figure 7 (**Paper G**). The purpose of these maps is to highlight gaps, and to indicate where the evidence underpinning tumour classification is very limited or weak. It will also be a resource to support the maintenance of the WHO Guidance on Tumour Classification, an internationally recognised standard informing decisions about treatments for cancer globally.

The types of evidence that I included in **Papers C, F, G** and Mikton et al 2022 differed. In **Paper C** and Mikton et al 2022, the maps only include review level evidence while in **Paper F and G** the maps include all types of study design as well as systematic reviews. These decisions were driven by the purposes of the maps and impacted on the nature of the gap analysis we could undertake. EGMs identify 'absolute gaps' where few or primary studies exist and 'synthesis gaps' where there is a concentration of eligible studies but no recent high-quality systematic review.(Snilstveit et al, 2017) The gap analysis of **Paper F** guided the focus of the subsequent systematic reviews that were undertaken (Whear et al, 2022, **Campbell et al, 2023c**) by clearly identifying the gaps where there was a cluster of eligible studies but no existing systematic review.

However, there are important limitations to these types of approaches. They frequently do not include risk of bias assessment, and therefore even a populated cell is not a guarantee that there is sufficient high quality evidence or that the evidence indicates effectiveness. These types of outputs should indicate where there is a need for an evidence synthesis but not replace it. Some maps are not accompanied by any evidence of the methods used or a link to a protocol (**Campbell et al, submitted**) and existing reporting guidance for scoping reviews (Tricco et al, 2016) does not consider the process of matrix development in EGMs.

These types of outputs will rapidly date due to the breadth of a topic and are likely to have a 'shorter shelf life' but there are still only a few examples of living EGMs. In order for EGMs to be up-to-date and therefore useful requires complex decisions driven by considerations relating to the number of new studies and existing studies, how dynamic the field is in terms of new research, and the sensitivity of the context in which evidence is informing decisions. Ravaud et al (2020) argue that the future of evidence ecosystems lies in creating accurate, concise, living evidence platforms and there are examples of these currently in use (https://pathwaystowork.acf.hhs.gov/). EGMs, and the development of living EGMs are a type of Big Picture review that will continue to grow in popularity and change expectations of what our evidence synthesis outputs might look like.

Finally, although I perceive that as an evidence resource they address some of the barriers to uptake by decision makers, this has not been evaluated and nor do we know how accessible they are to the public. These are challenging reviews to undertake, in part because they require intense training, both of the stakeholders so they can meaningfully engage with the process of the review, and for reviewers undertaking the reviews. I am maximising the skills I have learned in training and supporting review teams and stakeholders to deliver training on short course programmes, and at Cochrane Colloquia.

Evidence and gap maps are the most recent addition to the Big Picture review family, but the field is moving quickly. There is a need to ensure that visual appeal does not detract from transparency in methods to ensure reliable, useful and trustworthy outputs are produced.

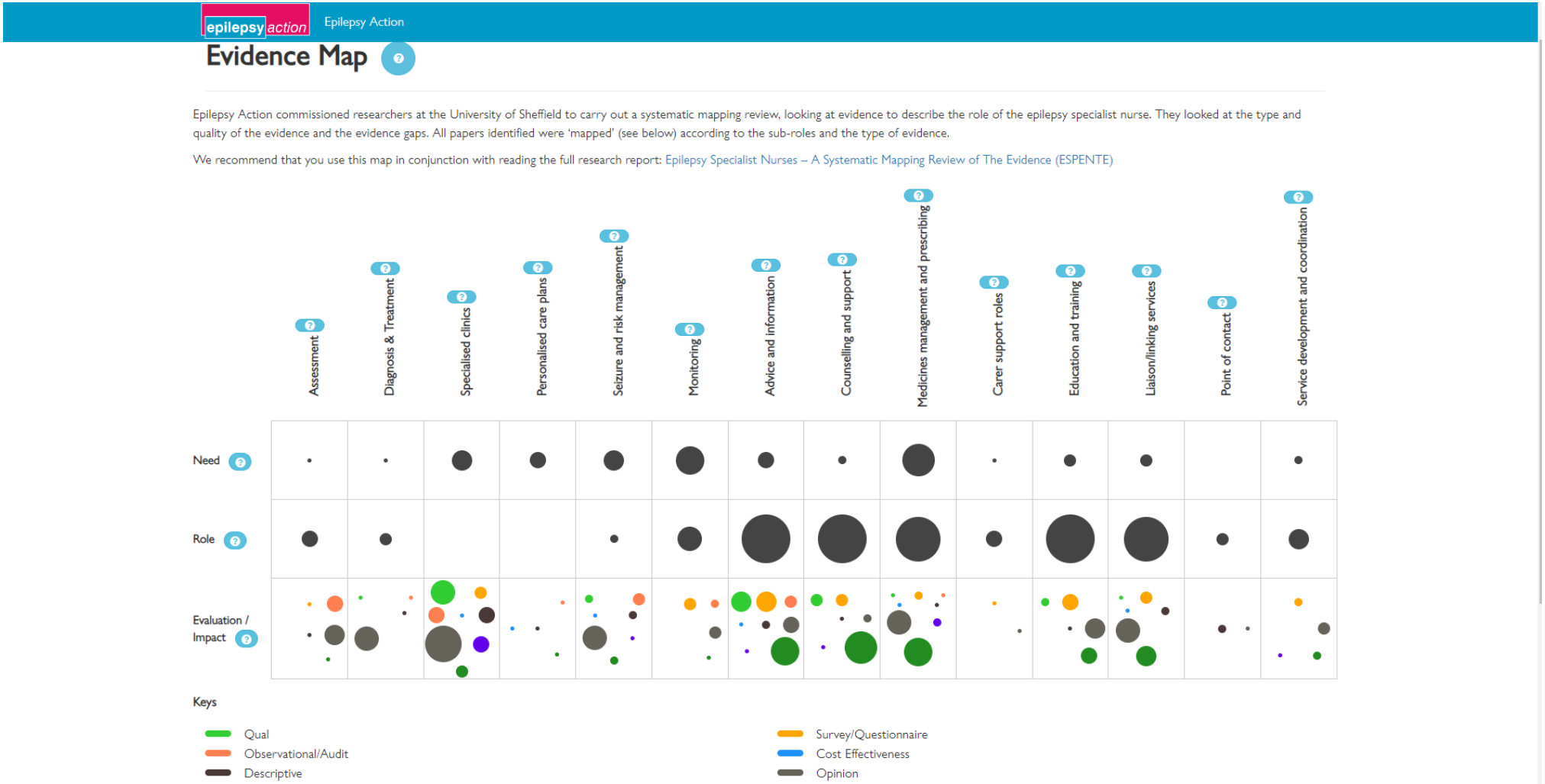

Figure 6: Visual of the Interactive Evidence and Gap Map examining the role of Epilepsy Nurse Specialists [ESPENTE - Evidence map]

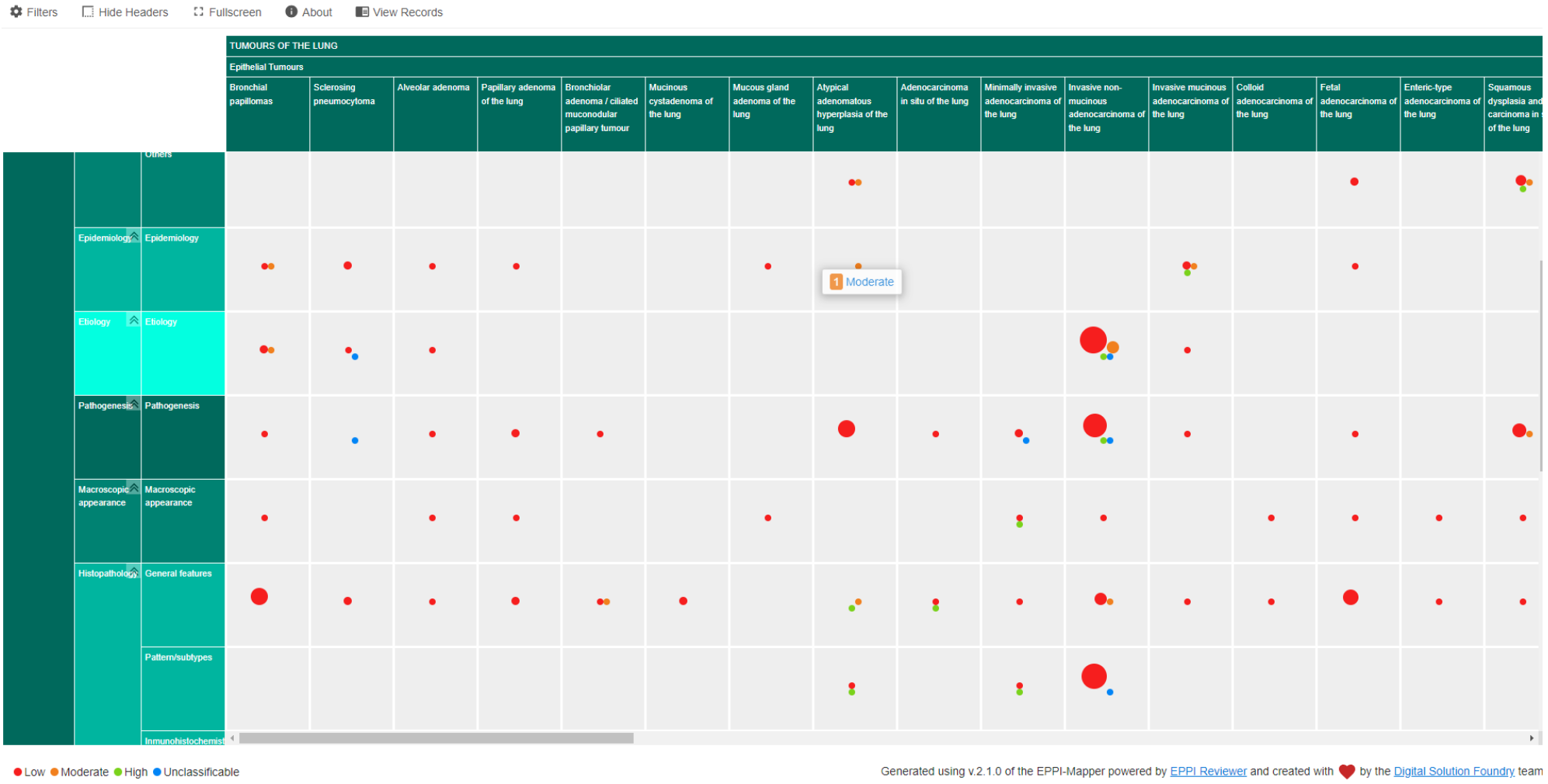

Figure 7: Visual of the WHO Evidence and Gap Map examining the evidence to support Classification of Lung Tumours

Non-familial Intergenerational Interventions and their Impact on the Social and Mental Wellbeing of Younger and Older People a Mapping Review and Evidence and Gap Map

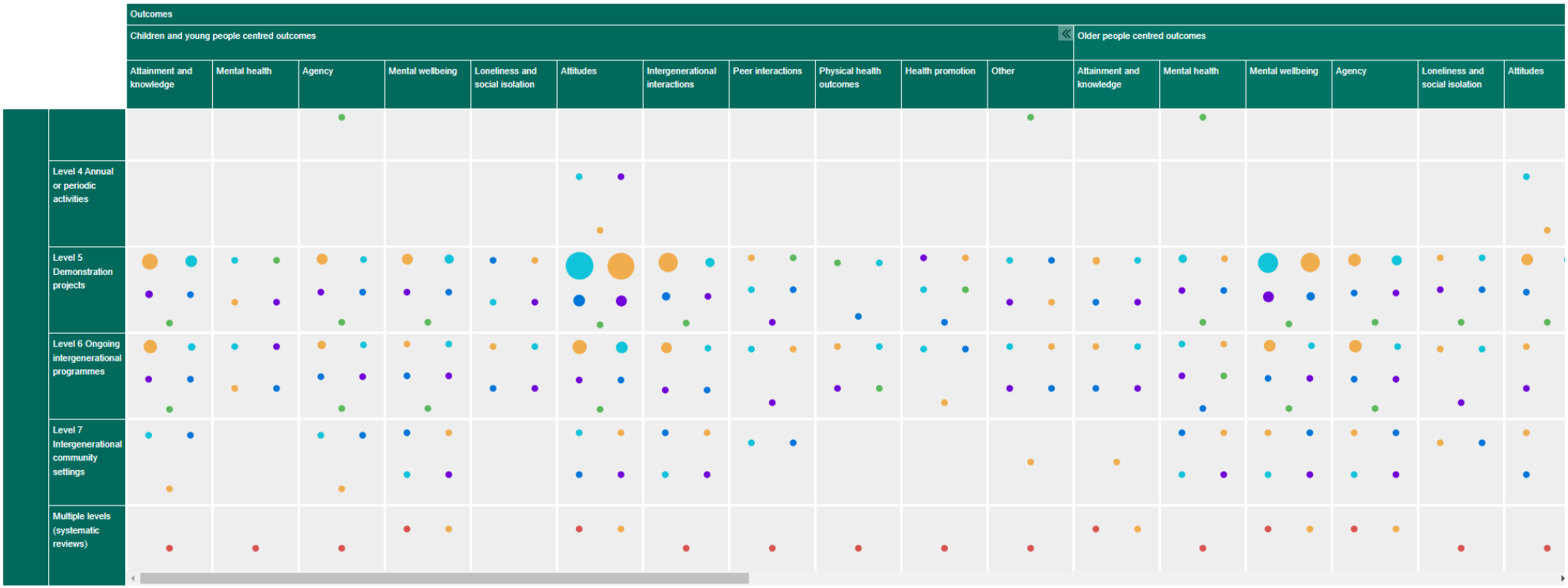

Figure 8: Visual of the Evidence and Gap Map of Intergenerational Interventions [EPPI-Mapper (wiley.com)]

Chapter Nine: Conclusions

How has my work contributed to the methodological development of Scoping, Mapping and EGM Methodology?

The evidence syntheses presented in this thesis, demonstrate how as I have sought to address broad research questions and apply scoping, mapping and EGM methodological guidance. In so doing, I have found contradictions and a lack of clarity in the methods literature, an absence of good examples to follow and limited software to operationalise commissioner expectations of visual and interactive EGMs.

These early findings led me firstly to explore the published literature, in greater depth, and to chase the roots of the definitions and guidance that was available. My conclusions from that work led to conversations with methodological experts within this field and the formation of an international group of methodologists whose focus remains collaborating on ongoing methods research. Even within the group, there was an absence of consensus on how the terminology was both being, used, and crucially how it should be used. These discussions led to the development of a proposed distinction between the approaches that would form an agreed position going forwards. This led to the key methodological paper that was the first to seek to explore the roots of the different terms and propose a method for categorising these outputs: (**Paper A**) *'Big Picture' Reviews: Scoping, Mapping and Evidence Gap Maps, The Same but Different*. The extent to which these distinctions will be adopted going forwards will rely on influential review organisations such as the Cochrane Collaboration implementing these definitions.

The application of scoping, mapping and EGMs presented some particular challenges, distinct from those I had experienced while undertaking systematic reviews addressing questions of effectiveness. I address these reflectively throughout this thesis by drawing on the experiences of undertaking the reviews that are the focus of (**Papers B, C, E and F**). These include exploring the challenges of question formulation when the research question is broad and the standard PICO framework is not applicable. The need for a more 'pick and mix' approach to selecting the elements of the framework in scoping and mapping reviews

may be a more useful approach for reviewers refining their research question, developing appropriate search terms, and clarifying their inclusion and exclusion criteria.

In the EGM looking at intergenerational interventions (**Paper F**), I have been able to demonstrate how stakeholder engagement directly impacted the use of the EGM by practitioners and decision makers. We also demonstrated the challenges of seeking to engage all stakeholders and, particularly, the views of children and young people. Scoping, mapping and EGMs present particular challenges for locating evidence. The iterative approaches needed and the need to find ways to reduce the search yield without compromising the review objectives led to the development of a new search filter to identify studies undertaken in LMIC contexts (**Paper D**).

My growing expertise enabled me to apply these methods across very different topics. The ability to cross-fertilise knowledge from topic to topic and then from field to field is a particular advantage of the role of the methodologist. I was able to lead the application of EGM methodology beyond social sciences, to pathology (**Paper G**). The broadening of the application of these methods into the medical sciences was an innovation I led with colleagues at the WHO IARC. They sought to find ways to demonstrate how evidence is used and applied in the classification of tumours. I developed a prototype map to demonstrate how EGM might be used to address the problem they were seeking to solve. This work led to the award of a 2.8 million Euro grant to support the development of EGMss across all tumour types. We have undertaken a Delphi study to explore how a traditional hierarchy of evidence can be adapted for pathology. Again, as a method, we are seeking to ensure that our outputs are relevant to knowledge users and our map frameworks are informed by those who subsequently use them.

Ongoing research, Publications in Progress and Implications for Future Research

In order to describe and chart the methods used in mapping reviews and EGMs I am working on two reviews examining the methods in mapping reviews and EGMs (**Campbell et al, submitted**, Khalil et al, submitted). The methods used in scoping reviews have been previously reported (Tricco et al, 2016). I draw a distinction between how the methods have been applied, and how we recommend they might be applied going forwards. For

example, one of our early findings in our review of EGMs is that many provide no information regarding plans to update them. Current practice, highlights a need for recommendations on how EGM methods should be reported, including how they are maintained as an up to date and relevant resource.

While there is methodological guidance for scoping reviews, which also includes mapping reviews (PRISMA-ScR), this does not apply to EGMs. The technology to support the use of EGMs, but there is a danger that the EGM is treated as a standalone output, without the protocols or supporting methods linked. This review of the methods used in EGMs provides a basis for ongoing work to support the development of guidance for EGMs.

Good working examples of co-production with stakeholders in Big Picture Reviews is limited, and we are preparing materials to support reviewers undertaking evidence and gap maps as the involvement of stakeholders requires a certain level of preparatory training to facilitate their engagement.

The review exploring methods of scoping reviews within the context of the COVID-19 pandemic meant we adapted our approaches to ensure that we achieved our review within a very tight timeframe. This work, along with the methodological research I have undertaken in rapid qualitative synthesis has paved the way for taking a lead in writing the methods for rapid scoping, mapping and EGMs as part of a suite of publications giving guidance on the use of rapid methods (**Campbell et al in press A**).

The large number of included studies in Big Picture reviews, often means there are methodological 'shortcuts' applied. Frequently this results in single reviewer screening and data coding or extraction. We know very little about the impact of errors or bias on the findings of these types of evidence synthesis outputs.

The impetus to create 'living' evidence synthesis is particularly important in Big Picture reviews. Their breadth of focus means that, in most circumstances, these types of review become out of date more quickly than a review with a very narrow focus. We are currently preparing a methodological paper (Rogers et al in preparation) from our Intergenerational

Interventions EGM demonstrating an approach to streamlining our search methods to facilitate time efficient updating of the review. This work will inform the methods we adopt in maintaining the EGMs that will support tumour classification (**Paper G**).

This thesis aims to navigate the chasm between the high ground occupied by methodological guidance and the complex reality of the swampy lowlands where we conduct Big Picture reviews with ambiguous research questions, uncertain parameters, and vast amounts of data. It explores the challenges of staying up to date, presenting findings creatively, and engaging stakeholders in the process. In this ever-evolving landscape, where technology plays a critical role, I hope to have provided an overview of the bigger picture while keeping a clear view of the intricacies involved.

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Appendices

Appendix A: Relevant publications currently in press, waiting for editorial approval, or for reviewer comment

Relevant Submitted Publications

Paper	Current Status
Campbell F, Wong R, Llewellyen JL, Bond M. Protocol: Methods used in the Development, Production and Updating of Evidence and Gap Maps: A Scoping Review	In press
Campbell F, Pollock, D, Sutton A, Tricco A, Khalil H. Guidance for Rapid Scoping, Mapping and EGMs. BMJ Evidence Based Practice. Campbell et al	Waiting for editorial decision
Campbell F, Sworn K, Dickinson J, Booth A. ESPENTE A Mapping Review of the Role of Epilepsy Nurse Specialists	Waiting for editorial decision

Ongoing Work and Papers in Preparation for Submission

Paper	Current Status
Campbell F, Chambers D, Wong R, Llewellyen JL, Paisley S, Meier P. Employment and Health: A mapping review and EGM	Paper in preparation
Mikton, Christopher, Campbell, Fiona, Beaulieu, Marie, Yon, Yongjie, Cadieux Genesse, Julien, St-Martin, Kevin, Byrne, Mark, Phelan, Amanda, Storey, Jennifer, Rogers, Michaela Elder Abuse: A mapping review and EGM	Paper in preparation
Nethan S, Perez de Oliveira M, Lauby-Secretan B, Campbell, F Oral Cancer: A mapping review and EGM of Primary Prevention Interventions	Paper in preparation

Appendix B: First Author Permissions

Publication

Publication
Indave BI, Colling R, Campbell F, Tan PH, Cree IA. Evidence-levels in pathology for informing the WHO classification of tumours. Histopathology. 2022 Oct;81(4):420-5

Statement

Contribution of the candidate: The candidate conceived the original idea, in discussion with the lead author (Iciar Indave), of using evidence and gap map methods to support the rigorous and transparent use of evidence to inform the WHO Classification of

Tumours. She prepared initial prototypes of frameworks to test the ideas with leading experts (Ian Cree, Puay-Hoon Tan, , Armon S, Colling R, Chechlinska M, Kowalewska M, Pollán M, Holdenrieder S,). She was a partner in preparing a successful grant, led by Iciar Indave and funded by EU Horizon (3.2 million Euros). The grant is supporting an international consortium to undertake a body of evidence and gap maps that will provide the foundation for evidence informed practice in the classification of tumours. One of the first of these evidence and gap maps is in development and led by Javier del Aguila Mejia. In both of the manuscripts she provided detailed comments and all authors approved the final draft. She is leading the training in evidence synthesis and evidence and gap map methodologies across the consortium

Endorsed: Prof. Ian A Cree

11 July 2023

Iciar Indave, MD, MPH, PhD
Scientific agent
Support to practice sector
Public health unit
Tel. (351) 211 21 03 17

European Monitoring Centre for Drugs and Drug Addiction (EMCDDA)
Praça Europa 1, Cais do Sodré, 1249-289 Lisbon, Portugal • www.emcdda.europa.eu

Fiona Campbell
Senior Lecturer in Evidence Synthesis
Evidence Synthesis Group
Population Health Sciences Institute
Newcastle University

Subject: PhD thesis University of Sheffield entitled *'Navigating the Swampy Lowlands: developing Methods for 'Big Picture' Evidence Synthesis'.*

I, Blanca Iciar Indave, am hereby giving my permission to include the following two publications below as part of this thesis:

Indave BI, Colling R, Campbell F, Tan PH, Cree IA. Evidence-levels in pathology for informing the WHO classification of tumours. Histopathology. 2022 Oct;81(4):420-5.

del Aguila Mejía J, Armon S, Campbell F, Colling R, Chechlinska M, Kowalewska M, Pollán M, Holdenrieder S, Tan PH, Cree I, Ruiz BI. Understanding the use of evidence in the WHO Classification of Tumours: a protocol for an evidence gap map of the classification of tumours of the lung. BMJ open. 2022 Oct 1;12(10):e061240.

Contribution of the candidate: The candidate conceived the original idea, in discussion with the lead author (Iciar Indave), of using evidence and gap map methods to support the rigorous and transparent use of evidence to inform the WHO Classification of Tumours. She prepared initial prototypes of frameworks to test the ideas with leading experts (Ian Cree, Puay-Hoon Tan, Armon S, Colling R, Chechlinska M, Kowalewska M, Pollán M, Holdenrieder S,). She was a partner in preparing a successful grant, led by Iciar Indave and funded by EU Horizon (3.2 million Euros). The grant is supporting an international consortium to undertake a body of evidence and gap maps that will provide the foundation for evidence informed practice in the classification of tumours. One of the first of these evidence and gap maps is in development and led by Javier del Aguila Mejia. In both of the manuscripts she provided detailed comments and all authors approved the final draft. She is leading the training in evidence synthesis and evidence and gap map methodologies across the consortium.

By signing below, I give my full consent

14th July, 2023
Iciar Indave

Publication

Foster, C.R., **Campbell, F**., Blank, L., Cantrell, A.J., Black, M. and Lee, A.C., 2021. A scoping review of the experience of implementing population testing for SARS-CoV-2. Public health, 198, pp.22-29.

Statement

Dr. Clare Foster (PhD)
Public Health Specialty Registrar
Centre for Pandemic Preparedness
UK Health Security Agency

Contribution of the candidate: The candidate led an application to undertake the review. She designed the methods and prepared the study protocol. She led the review team and undertook all aspects of the review process. She wrote the initial un-published report of the review which was sent to the funders. Prior to preparing the published paper an update of the review was undertaken and Clare Foster wrote the draft of the published paper. The candidate commented extensively on the draft and all authors approved the final draft of the published paper.

Best wishes,

Clare Foster

Appendix C: Included Papers; url links and hard copies

Paper A: Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but different— the "Big Picture" review family | SpringerLink

https://link.springer.com/article/10.1186/s13643-023-02178-5

Paper B: A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance - ScienceDirect

https://www.sciencedirect.com/science/article/pii/S0895435619300162?casa_token=mvE1KGzf_9MAAAAA:qYdIJIYPxttGUWXEHwXMkhjYUtEOSoE3r7DbS-E6dVJi9ouyuAAYBjuVhcy-eMANALH1Mr2FQA

Paper C: Interventions for the prevention of spontaneous pretem birth: a scoping review of systematic reviews

https://bmjopen.bmj.com/content/bmjopen/12/5/e052576.full.pdf

Paper D: The ScHARR LMIC filter: Adapting a low- and middle-income countries geographic search filter to identify studies on preterm birth prevention and management (wiley.com)

https://onlinelibrary.wiley.com/doi/pdfdirect/10.1002/jrsm.1552

Paper E: A scoping review of the experience of implementing population testing for SARS-CoV-2 - ScienceDirect

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Paper A

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Mapping reviews, scoping reviews, and evidence and gap maps (EGMs): the same but different— the "Big Picture" review family

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Abstract

Scoping reviews, mapping reviews, and evidence and gap maps are evidence synthesis methodologies that address broad research questions, aiming to describe a bigger picture rather than address a specific question about intervention effectiveness. They are being increasingly used to support a range of purposes including guiding research priorities and decision making. There is however a confusing array of terminology used to describe these different approaches. In this commentary, we aim to describe where there are differences in terminology and where this equates to differences in meaning. We demonstrate the different theoretical routes that underpin these differences. We suggest ways in which the approaches of scoping and mapping reviews may differ in order to guide consistency in reporting and method. We propose that mapping and scoping reviews and evidence and gap maps have similarities that unite them as a group but also have unique differences. Understanding these similarities and differences is important for informing the development of methods used to undertake and report these types of evidence synthesis.

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Introduction

Evidence synthesis(defined broadly as the rigorous collation, evaluation and analysis of literature, studies, and reports) is increasingly viewed as critical to inform decision making in policy and practice. Over the past three decades, as various methods of evidence synthesis have emerged and evolved, the systems and labels used to categorize different review types have proliferated. A recent catalog of evidence synthesis approaches and terms identified 48 distinct review types [1]. Moher et al. (2015) [2], describes them as a "family" of evidence synthesis products that have arisen in response to policymakers and other stakeholders needs for diverse forms of information. This growth reflects the increased value placed on evidence synthesis to inform decision making, and we now see evidence synthesis used to address a broader range of research questions beyond effectiveness, along with tailored approaches (in terms of methods and

products) to evidence synthesis as appropriate for different research needs, purposes, situations, and audiences [3].

Examples of approaches that are increasingly seen in the published literature are scoping reviews, mapping reviews, and evidence and gap maps (EGMs). Scoping reviews, mapping reviews, and EGMs are relatively new approaches that rarely appeared before 2009 [4, 5]. Scoping reviews, evidence maps, and evidence and gap maps have been grouped together as "Big Picture" approaches due to their shared purpose and approaches. These Big Picture reviews can be contrasted with systematic reviews (addressing interventions, diagnostic test accuracy, prognosis, etc.) as they have a broader scope as compared to the (normally) narrower scope of classic systematic reviews. There have been consistent yearly increases in the publication of scoping, mapping, and evidence and gap maps [6]. Despite this, there remains confusion as to their application, meaning, and whether differences exist between them. This commentary aims to clarify these approaches, identify any differences between them, and provide recommendations for reviewers.

Terminology matters

This growing and evolving family of evidence synthesis types presents some challenges [7].

Firstly, there is the challenge of choosing the correct approach, particularly when terms are used inconsistently in the literature. The selection of an appropriate review approach will ensure the correct methods are employed using the appropriate standards for both its conduct and reporting. Indexing and wider dissemination can be challenging for researchers when there is ambiguity in terms [8, 9].

Scoping reviews and mapping reviews—how are they used in the literature

Scoping reviews, mapping reviews, and evidence maps are terms that are not used consistently in the literature, with different terms used to describe similar approaches and review objectives. The same term is also used to describe different approaches and review objectives. Within the published literature, the terms scoping reviews and mapping reviews appear to be used in three different ways. Firstly, the terms "mapping" and "scoping" reviews are used interchangeably, referring to the same type of review methodology [5, 6, 10]. This approach is also one that is used in the PRISMA Extension for Scoping Reviews (PRISMA-ScR) [11], providing guidance to inform reporting standards [12]. This may therefore have been influential in increasing the use of the term scoping review over the use of the term mapping review. Examination of published reviews does not reveal differences in method between these approaches (Campbell et al., 2022 publication in press).

Secondly, we see the terms used as complementary to the other. Some definitions tend to use the terms in a way which suggest that mapping is a specific approach to scoping—or vice versa. For example, "scoping reviews can usefully map the evidence in a number of ways" [13] and "scoping reviews are a way of mapping the key concepts" [14]. Lukersmith et al. (2016) [15] and Fernadez-Sotos et al. (2019) [16] suggest that the term map is a descriptive term used to describe one of the purposes of the scoping review. A mapping review may also scope the literature. It has also been suggested that when the term mapping is included in the description of the method that the review will incorporate a geographical mapping exercise or charting of the data in a tabular or any other visual format that can plot or portray the data.

Finally, we see scoping and mapping used to describe different types of evidence synthesis, and a distinction is made between mapping and scoping reviews [1, 17]. These authors suggest that scoping reviews are "preliminary assessment of potential size and scope of available research literature which aims to identify nature and extent of research evidence (usually including ongoing research)". It also is a term that has emerged within the systematic review field to describe the preliminary work undertaken with information specialists in planning the review, by getting a sense of the size of the literature, to identify key terms and theories and potentially clinical experts [18]. Within these definitions, mapping reviews are distinguished from a scoping review because the subsequent outcome may involve either further review work or primary research and this outcome is not known beforehand. For the purpose of this paper, we will refer to these as a scoping exercise instead of a formal scoping review methodology. Scoping exercises within this definition would not usually be regarded as a final output in their own right, primarily because of limitations in their rigor mean that they hold the potential for bias.

Gough et al. (2012) [19] suggest that the term scoping review often describes a more rapid, and so usually non-systematic, approach to describing the nature of the literature on a topic area, sometimes as part of planning for a systematic review compared with a standard systematic review. It is also important to note that there are published rapid scoping reviews where streamlined methods are used, but transparency and rigor are maintained to produce quicker results for decision-making purposes. Examples of these types of rapid scoping reviews include rapid responses to policy questions during the COVID-19 pandemic [20, 21].

An alternative view of the difference comes from Bragge et al. (2011) [22] who suggests that a scoping review is distinguished from mapping by the inclusion of research results in the description of relevant evidence, whereas maps simply describe what is there without collating and summarizing the results of the studies.

So, even where the types of products are seen as different, there is not a consistent approach in this difference. Nevertheless, understanding why they are considered different is important in considering what is lost, in terms of an apt descriptor, if the terms are amalgamated and used interchangeably.

Historical origins

One reason that the terms scoping and mapping have emerged to describe two similar methodological approaches addressing broad types of research questions lies in the academic traditions from which they derive and the epistemological foundations upon which these are built. Scoping reviews and scoping review methodological guidance [12] tends to cite the framework defined by Arksey and O'Malley (2005) [23] and later enhancements by Levac et al. (2010) [24]. These approaches have their roots in sociological sciences. In contrast, the term evidence mapping was first used by Katz et al. (2003) [25] and has roots in the natural sciences. This was the term adopted by the EPPI Center in an early publication of a mapping review and is the term used by the Center for Environmental Evidence for the environmental sciences. The approach to evidence mapping accompanied by a visual evidence and gap map has been developed by several agencies (see Saran and White, 2018) [26], most notably by the International Initiative for Impact Evaluation (3ie) [27] in the field of international development and subsequently adopted and adapted to a wider a range of sectors through the Campbell Collaboration. These include, for example, transport [28], youth violence, disability (Saran et al. [29]), employment (Campbell et al. [30]), and health and elder abuse [31] (Table 1).

Suggested approaches for distinguishing between mapping reviews and mapping reviews with EGMs and scoping reviews

The emergence of two terms (scoping and mapping) to describe approaches that have much in common in terms of their objectives and methods suggests that the terms used will be shaped more by the academic background of the researcher than by inherent differences in the approaches.

Currently, as we have shown, there are many instances where "mapping and scoping" are used interchangeably. We argue, in this paper, that while there is considerable overlap between these approaches, there is value in creating a distinction between scoping reviews, mapping reviews, and evidence gap maps. They also could be considered complementary, and a review may have elements of both "mapping" and "scoping." Each approach, within this family of "broad approach and exploratory reviews" however has a shared objective which is to overview a wider research/topic area, rather than to address a tightly focused question. The methods thereafter diverge in part to address the nature of the research question, the research objectives, the topic area, the depth required for the data extraction, and the expertise of the review team.

We propose that a useful distinction is to see mapping, scoping, and EGMs sitting within the same family of types addressing broad questions but sitting on a spectrum in some of their underpinning epistemologies, concepts, and hence objectives (Fig. 1).

This is illustrated in the figure below:

Scoping review

These review types have been variously defined and described in the literature as described above. To address the confusion in this field, a recent formal definition of scoping reviews has been proposed, describing scoping reviews as follows:

It is a type of evidence synthesis that aims to systematically identify and map the breadth of evidence available on a particular topic, field, concept, or issue, often irrespective of source (i.e., primary research, reviews, non-empirical evidence) within or across particular contexts. Scoping reviews can clarify key concepts/definitions in the literature and identify key characteristics or factors related to a concept, including those related to methodological research [32].

They can be more exploratory than mapping reviews and EGMs, not requiring an a priori set of codes in order to describe data and may draw upon a range of sources of information *(i.e., primary research, reviews, non-empirical evidence) within or across particular contexts.*The approach can be more iterative, inductive, or deductive [32]. The nature of the "cataloging" and coding may be in response to what is found within the literature or using pre-defined categorization codes. Scoping reviews can also be used to identify concepts and clarify terms in the literature. In contrast to a mapping review where the process of coding is predefined. Within a scoping review, the data extracted may be textual and descriptive, allowing for example an analysis of concepts and categories using simple content analysis. It may include both predefined coding and also exploration of themes (for example, Kelly-Blake et al. 2018 [33]). In contrast, along a continuum, mapping reviews will address broader questions, use predefined coding, and adopt less in-depth data extraction.

Table 1 Summary of the different roots and institutions that use mapping and scoping reviews

	Scoping review	Mapping review	EGM
Academic roots	Social sciences Arksey & O'Malley 2005 [23] Levac 2010 [24] Khalil et al. 2016 [4] Peters et al. 2020 [12]	Public health, Biomedical sciences, Environmental science James 2016 [18]	International Development 3ie Snilstveit et al. 2013 [27] Saran & White 2018 [26]
Research concepts	*Inductive and *Deductive *Configurative *Aggregative	Deductive Aggregative	Deductive, inductive Aggregative
Guidance for methods (and reporting)	JBI (PRISMA ScR) [5–8]	SCIE, Campbell Collaboration (PRISMA ScR)	Guidance: Campbell White et al. [34]
Identifies gaps in the research	Yes	Yes	Yes—using a pre-specified framework
Visual and interactive web-based gap map	No—but may contain tables and diagrams within text	No—but may contain within text tables and diagrams—and may be produced with an EGM	Yes

* Aggregative synthesis: where the synthesis is predominantly aggregating (adding up) data to answer the review question

* Configurative synthesis: where the synthesis is predominantly configuring (organizing) data from the included studies to answer the review question

Aggregation and configuration fall on a continuum and all reviews are likely to both aggregate and configure data to some extent [35]

* Deductive reasoning: a pre-existing theory or framework that must be tested

* Inductive reasoning: an unknown theory or framework that needs to be developed

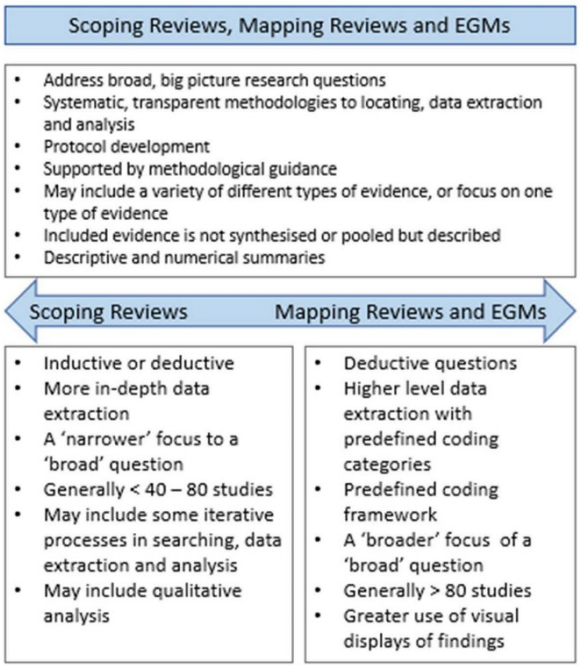

Fig. 1 The Big Picture review family (commonalities and differences in approaches)

Mapping review

Mapping reviews are also a transparent, rigorous, and systematic approach to identifying, describing, and cataloging evidence and evidence gaps in a broader topic area. They are to collate, describe, and catalog the available evidence relating to the question of interest [18]. They aim to answer the question "what do we know about a topic," or "what and where research exists on a particular area." A mapping review typically extracts only descriptive information about the studies and applies predefined codes (high level data). In this sense, they may be informed by an "aggregative" logic. A mapping review may or may not be accompanied by an EGM but provides visual summaries in the form of tables and graphs within the text [36]. These types of reviews may well have broader focus than a scoping review, with more limited data extracted from the included papers.

Evidence and gap maps

Evidence and gap maps are described as "a systematic presentation of all relevant evidence of a specified kind for a particular sector, sub-sector, or geography". Evidence and gap maps (EGMs) are a systematic evidence synthesis product which displays the available evidence relevant to a specific research question. EGMs consist of primary dimensions or framework (rows and columns) and secondary dimensions or filters, enabling exploration of the map using a particular focus (e.g., looking at particular populations or study designs). It creates a visual, web-based, and interactive output [34].

This type of evidence synthesis generally uses a deductive approach with a pre-specified framework to classify the data and identify gaps in the literature. However, if no suitable framework is available, then the research team can develop their own by drawing on the range of resources, such as strategy documents, policy document, and funder reports. This is one of the major differences between mapping with an EGM review and scoping reviews (for the latter, an inductive or deductive approach may be used to identify relevant data elements so the framework for classification of the data and identification of gaps does not need to be pre-specified). Evidence gap maps may accompany a mapping review as a visual representation of the included studies or can stand independently from an accompanying mapping review.

Purpose

All three of these approaches are characterized by seeking to address a broader topic area rather than a specific intervention or exposure. They are an appropriate tool if the research question is one in which multiple dimensions need to be considered, for example, multiple interventions, outcomes, or types of evidence. They do not aim to synthesize data but rather describe, categorize, and catalog findings. They aim to do so by applying defined methods to ensure transparency and rigor in the process of identifying, screening, data extraction, and interpreting findings. By addressing a broad topic area these approaches support the following purposes [3]:

Knowledge generation to support broad research questions and objectives such as the following:

- What types of evidence are available in a given field?
- How are concepts or definitions used within the literature?
- How and where research is conducted on a certain topic?

The type of broad research question will inform the choice of approach. Scoping reviews are more likely to address open questions and the concepts may be emergent such "how is a key term used within the literature," in contrast a mapping review may address more closed questions such as "how often the key term is used within the literature and within which population groups." An evidence gap map will similarly address a closed question, for example, "is the term used in the following types of population group: children, adolescents, older people, and people with chronic conditions."

Scoping reviews can provide an approach that allows exploration and clarification of key concepts and definitions within the literature, as well as how research is undertaken. As this approach does not require predefined categories, it allows for more descriptive data extraction. Often the question will be narrower than in a mapping review, allowing a greater depth of exploration of the included studies.

These approaches enable a better understanding is gained of phenomena by seeing it within a wider context. Olson et al. 2021 [37] uses the allegory of the blind monks who examine the elephant, where close inspection of one part of the whole means that meaning is lost. A complete picture is needed to really understand what the elephant is. It is clear, when seeking to operationalize what is meant by a "broad" topic area that perspective matters. For a cell biologist, the cell nucleus might be a broad topic, which a single country might be too narrow a perspective for the geographer. Understanding this unique feature of "Big Picture" reviews is perhaps easier when seen in contrast to the approach used in a systematic review examining the effectiveness

Table 2 Examples of review aims

Type of approach	Aim
Scoping review	To report in detail the methodology employed to identify relevant theories and provide a list of agreed criteria for judging the quality of theories (Davis et al. 2015) [41] To document and describe the evidence base relating to stakeholder involvement in systematic reviews and to use this evidence to describe how stakeholders have been involved in systematic reviews (Pollock 2018) [42]
Mapping review	A mapping review of research on gambling harm in three regulatory environments (Baxter et al. 2019) [43]
EGMs	To identify what has been published on micronutrients and depression and identify gaps in the evidence and collections suitable for meta-analysis. (Campisi et al. 2020) To identify intergenerational interventions and the social and mental wellbeing outcomes that have been measured in their evaluation. (Thompson-Coon et al. 2022)

of a single intervention. A Big Picture review question will look at multiple interventions or exposures and multiple outcomes or effects, seeking not to synthesize but to describe (Table 2).

To provide a foundation for guiding future research priorities and decisions by identifying available evidence and gaps in research

Mapping reviews and EGMs incorporate a framework that is generated during development of the protocol—it is this framework which guides the development of the data extraction tool or coding tool. This framework becomes the "map" against which existing evidence is plotted.

Identifying research gaps is often a stated part of all types of research; indeed, implications for "research and practice" are an expected part of all health and social care-related research. Identifying research gaps is often a primary purpose of scoping, mapping, and mapping reviews with EGMs more than other types of review design. In particular, mapping reviews with or without evidence gap maps address this purpose with a transparency and rigor that is unique.

Evidence and gap maps aim to enable evidence to be located, both by showing what is there but also in demonstrating knowledge gaps. In order to identify knowledge gaps, an EGM begins by developing the framework against which the evidence is plotted. The development of the framework adheres to the following principles. Firstly, it may be constructed using an existing, widely accepted international typology for either interventions, exposures, or outcomes. Secondly, if no suitable framework is available then the research team may draw on a range of resources including consultation with stakeholders and relevant published theories to ensure the comprehensiveness of the framework. Without such a structure, the gaps are not identified in a systematic way, but rather inferred and chosen by the review authors (no doubt well informed) but nevertheless influenced by their own perspectives and bias. This may be particularly apparent where a review is undertaken to pave the way for further primary research by the same team. Review teams could be strongly invested in identifying their own planned research as the "research gap."

Evidence gap maps are a systematic approach to identifying the evidence and in particular—its gaps. No other review methodology has developed a systematic approach to identifying gaps in the evidence with this level of rigor and transparency. A limitation of the approach is that it only charts what is known and does not allow a more exploratory approach that may be employed in a scoping review.

Mapping and mapping reviews with EGMs aim to describe the state of evidence for a question or topic. The review questions may therefore be open framed and broad. However, the question can be close framed and narrow. Key elements of the question can be formulated by a framework such as PO (population, outcome). For an EGM, the objectives are formalized in the framework which defines the scope of the map [34].

To inform policy decisions, where an overview of an area may be more helpful than specific questions about specific types of interventions

Mapping (with or without an EGM) and scoping reviews often have pertinence for policy makers as they are able to cover the breadth of science often needed for policy-based questions; however, it needs to be remembered that the mapping approaches do not synthesize the findings and not include quality or risk of bias appraisal. These factors may limit their value to support some types of policy decisions. However, a mapping review with an accompanying EGM can take users to the research papers and facilitate the ready location of relevant evidence. An EGM can take users to the research papers and facilitate the ready location of relevant evidence. One example has been the use of a country evaluation map used by the Office of the Prime Minister of Uganda to identify studies

to inform policy work [38]. Similarly, scoping reviews can inform policy and further research through identifying the available literature pertaining to a particular topic, along with clarifying key concepts and definitions.

As a stepping stone to building the evidence architecture

Evidence mapping and EGMs may be used as a first step towards the generation of evidence-based decision-making products, such as guidance, checklists, and online decision-making tools [39]. Maps will identify the (i) existing reviews which are suitable to use a basis for guidance, etc., (ii) where there are clusters of primary studies but no review so reviews may be commissioned in priority areas to inform guidance, etc., and (iii) important policy areas in which evidence is missing. To serve this purpose, the map should be regularly updated (maintained).

Discussion

While the literature is inconsistent in its definitions of these types of reviews, and different reviews use different terminology to describe methods that appear very similar, many of these differences reflect the different research traditions and adoption of terms within organizations undertaking these types of syntheses. We argue that there is value in having these distinct terms to describe the different approaches within this family of broad review types. Scoping reviews allow a more inductive, in-depth approach with, including fewer included studies and a greater level of data extraction compared with mapping reviews. Mapping reviews and evidence gap maps address more closed questions, with pre-specified items defined and codeable when contrasted with scoping reviews. Evidence gap maps offer a visual, interactive output for users to locate evidence. The predefined framework offers a rigor to locating gaps in the existing literature and displaying these differences which is unique to these approaches.

This proposed new "Big Picture" review family within evidence synthesis contributes to the wide array of possible approaches to synthesizing literature. This multitude of choice presents challenges in selecting the correct evidence synthesis methodology. One tool that has been developed to assist in the appropriate selection of a method is the "right review" tool (https://whatreviewisrightforyou.knowledgetranslation.net/). The tool enables researchers to answer a series of simple questions regarding the type of research questions they are undertaking for their review and selects an appropriate type of review based on their answers to the questions. The tool currently includes 41 different types of evidence synthesis methods [40].

A recent development has been changes made to the SR Toobox (http://systematicreviewtools.com/index.php) to include searching for tools to support different review types, as well as for different stages of the review. The Big Picture review family is increasingly well supported by methodological guidance and automation tools to support the process of undertaking high quality systematic reviews.

The existing guidance for the conduct and reporting of scoping reviews also applies to mapping reviews (JBI). Further development is needed in the methods of preparing a coding framework, particularly when the mapping review will also include the development of an interactive EGM. Current models of good practice exist; however, current guidance and reporting standards are limited.

Conclusion

This commentary details and describes some of the broad approaches within the evidence synthesis toolkit, specifically scoping reviews, mapping reviews, and EGMs. We have identified similarities and differences, based on our expert experience, between these reviews. We propose grouping them as a family of evidence synthesis to address broad research question and objectives. In so doing, we advocate that adherence to the principles of rigor and transparency that give users of evidence synthesis confidence in the reliability of the results of the review.

Appendix

Useful resources: https://guides.temple.edu/c.php?g=78618&p=4156607 and https://wiki.joannabriggs.org/display/MANUAL/11.2+Development+of+a+scoping+review+protocol.

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Authors' contributions

FC and HW conceived the idea for the paper, and FC initiated the initial draft. HK, AT, HW, ZM, AS, and AS contributed to the comments and edits of subsequent drafts of the paper. HK and FC are co-chairs in the NAVIGATOR method group and took a lead in the preparation of this work. The final manuscript has been read, edited, and agreed by all the contributing authors.

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Declarations

Competing interests

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Publisher's Note

Paper B

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ORIGINAL ARTICLE

A scoping review found increasing examples of rapid qualitative evidence syntheses and no methodological guidance

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Abstract

Objectives: The objective of the study was to identify existing methodological guidance for the conduct of rapid qualitative evidence syntheses and examples of rapid qualitative evidence syntheses to describe the methods used.

Study Design and Setting: We conducted a systematic scoping review. We searched MEDLINE, CINAHL, gray literature, including PROSPERO, with no date limits and solicited examples through experts and researchers in the field.

Results: We found no methodological guidance to direct the conduct of rapid qualitative evidence synthesis and 15 examples including 13 completed reviews and two protocols. Diverse methods to abbreviate the review process were followed, which largely mirror methods developed for rapid reviews of clinical effects. Abbreviated search strategies, including date and language restrictions, were common, as was the use of a single reviewer for screening, data extraction, and quality appraisal. Descriptive approaches to synthesis, such as thematic synthesis, were more common than interpretive approaches, such as metaethnography.

Conclusion: There is a need to develop and explore methods for the synthesis of qualitative research that balance the need for rapidity with rigor. In the meantime, providing details on the methods used, shortcuts made, and the implications of such methodological choices, together with collective sharing of innovations, becomes more important under increased time constraints.

Keywords: Rapid reviews; Qualitative evidence synthesis; Review methods; Scoping review; Systematic review; Policy; Knowledge mobilization

1. Background

The past decade has witnessed the proliferation of methodological literature on, and examples of, Qualitative Systematic Reviews or Qualitative Evidence Synthesis (QES). QES is an umbrella term that refers to the methods used to search, select, and analyze findings from a set of primary qualitative research studies that relate to a specific topic or focus to arrive at new or enhanced understanding about the phenomenon under study [1].

Multiple factors have stimulated recent interest in the synthesis of qualitative studies. First, decision makers are recognizing the potential usefulness of, and distinctive contribution of, qualitative research. Qualitative evidence enables insights into the contexts that shape the use of, and therefore the effectiveness of complex interventions, and helps to understand the acceptability and feasibility of interventions, the value of outcomes to health service users, and the impact of interventions on equity and human rights [2,3]. Within health technology assessment (HTA), this interest in qualitative research reflects a policy imperative to ensure that the needs, preferences, and experiences of patients are central to decisions on technologies, treatments, or service redesign [4]. Furthermore, a QES can inform, enhance, extend, or supplement reviews addressing intervention effectiveness [1]. As the influence of the evidence-based practice agenda increases, so too comes the need for rigorous evidence synthesis of existing research, including qualitative research [5].

A key issue relates to the extent to which QES represents a recognizable variant of the systematic review, as opposed to being of its own kind. A systematic review of the literature (published and unpublished) follows explicit, transparent, and reproducible methods to address a clearly formulated, and traditionally clinically focused, research question [1]. Systematic reviews that follow rigorous and

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What is new?

Key findings

- We were unable to identify any published guidance specifically on methods for undertaking RQES.
- The RQES examples located in our search used many of the approaches seen in rapid effectiveness reviews such as single reviewer screening and data extraction and more restricted searching. The implications for rigor may differ for RQES and therefore guidance needs to be tailored to the different types of rapid evidence synthesis increasingly used.
- RQESs appear to diverge from full qualitative evidence syntheses in the methods of synthesis used. RQES more commonly adopted descriptive aggregative approaches rather than interpretive approaches that develop conceptual understanding.

What this adds to what was known?

- This is the first scoping review undertaken exploring the methods of an emerging approach to evidence synthesis: rapid qualitative evidence synthesis (RQES).
- Current guidance on rapid review methods, although increasingly plentiful, either focuses exclusively on rapid effectiveness reviews or offers generic guidance that does not explicitly specify the type of review.

What is the implication and what should change now?

- The use of timely synthesis in policy-making requires both rigor and transparency. This work lays foundations for the development of guidance to support this emerging methodology, which is increasingly being used to support patient-oriented decision-making.

transparent methods and include high-quality primary studies are regarded as optimal sources of research evidence to address clinical and health policy questions. Accordingly, systematic review methods are increasingly applied or modified to answer questions using other types of evidence, including qualitative research. Although many synthesis methods are common across different types of evidence, fundamentally different aims and assumptions underpin synthesis of qualitative research, quantitative research, or mixed research types.

The rigor of systematic review standards requires that they typically take between 6 months and 2 years or more to complete [6]. To address this challenge, methods to expedite the process are increasing [7] and rapid reviews are increasingly common, recognizing that policy makers cannot always afford to wait for findings from a systematic review [8]. Although estimates vary, rapid reviews may be conducted within as little as 8 weeks, potentially saving about 75% of the time from a typical systematic review timeline [9]. Notwithstanding substantial time savings, this shorter timeline requires either extensive resource use or, more commonly, limitations in scope and/or compromises in rigor.

Methods for the development of rapid reviews are evolving to address risks of bias, reporting guidelines, and decisions about appropriate rapid review processes. Given fundamental differences between clinical studies and qualitative studies, it is unclear whether methods used to rapidly synthesize results from the former apply equally to the syntheses of the latter. With nineteen documented approaches to synthesizing qualitative research [10], it is challenging to identify where best to target abbreviated or accelerated qualitative synthesis processes [11]. For example, risk of bias, or quality, is conceived differently and "shortcuts" may present different threats to rapid QES. Similarly, a rapid QES may require demonstrably different processes of synthesis. We have identified a need to understand the extent to which generic rapid review methods translate to rapid QES, and any consequences for rigor. A prerequisite step is to map existing guidance and how reviewers adapt methods to acknowledge the twin needs associated with rapid evidence synthesis of qualitative evidence.

2. Objectives

Our objectives were to

1. Identify existing methodological guidance for the conduct of rapid QES and
2. Identify examples of rapid QES and describe the methods used.

3. Methods

A systematic scoping review approach was chosen to collate, catalog, and describe the state of knowledge for rapid QES methodology [12]. The review protocol lies outside the scope of PROSPERO registration, as it does not address health outcomes, and is available from the authors on request.

3.1. Criteria for considering studies and methodological papers for review

To address our first objective, we sought to identify and include articles that describe methods, or offer guidance, for the conduct of rapid QES. This includes articles describing rapid review approaches to synthesis of any type of qualitative study, or of studies of an unspecified type, but

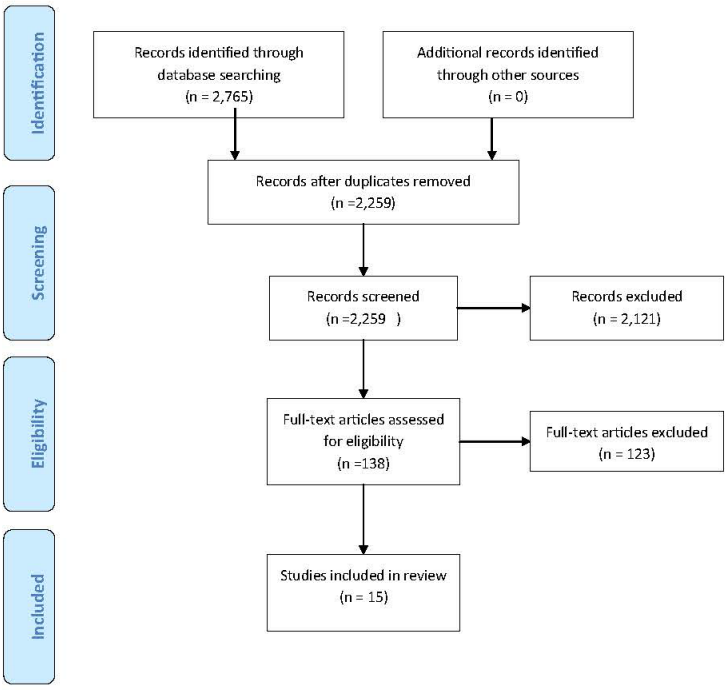


Fig. 1. Flow of studies through the scoping review.

excludes articles explicitly describing synthesis of quantitative or mixed method study designs. To address our second objective, we sought to identify examples of QES that incorporate abbreviated or accelerated approaches within restricted timelines. We focused on syntheses that address a health care intervention or health condition, and we excluded evidence for non–health care topics. During citation screening, we further refined our prespecified inclusion criteria such that examples of rapid QES were required to fulfill three criteria for inclusion. First, authors should explicitly identify their study as "rapid" or otherwise provide evidence that the review was expedited using "shortcuts." For example, reports where the systematic review process was modified to facilitate efficiency or time savings, such as use of text mining, were eligible even where a review was not described as "rapid." Second, evidence of systematization (e.g., in identifying data sources or assessing study quality) was required. Finally, a review should positively indicate that it only included qualitative primary studies (i.e., excluding mixed methods reviews).

3.2. Identification of articles

A peer-reviewed literature search was conducted using MEDLINE via Ovid and CINAHL via EBSCO. The search strategy was developed using Medical Subject Headings and keywords related to "rapid reviews" and "qualitative" research. The search strategy for MEDLINE is provided in Appendix A. The MEDLINE strategy was run on March 31, 2017, and was adapted for use in CINAHL on April 11, 2017. Update searches of both databases were run on March 13, 2018. Gray literature was identified from relevant databases or websites of HTA agencies listed in the Gray Matters checklist (https://www.cadth.ca/grey-matters). A focused Google Scholar search was performed using Publish or Perish software [13] for permutations of rapid with qualitative synthesis or review. The PROSPERO international prospective register of systematic reviews (https://www.crd.york.ac.uk/prospero/) was searched using broad keywords (e.g., qualitative and rapid, abbreviated or brief) to identify relevant protocols. Follow-up searching for publications relating to PROSPERO protocols was undertaken in PubMed, Google, and Google Scholar. Studies from the gray literature were identified through contact and consultation with experts. The reference list of all eligible studies was examined to identify potentially relevant guidance for the conduct of rapid QES, or examples of rapid QES.

3.3. Selection of articles

Titles and abstracts of all records obtained from the search were independently double screened. Four reviewers

Resources • Personnel • Databases • Data extraction (blind dual data extraction or single reviewer) • QA tool	Expertise	Audience and purpose	Type of data
• 2 authors including IS • 6 DBs • Single reviewer • CASP	Qualitative plus IS	Commissioned for HTA agency	5 studies; Qualitative descriptive design [1], phenomenologies [2], grounded theory [1], consensus design [1].
• 2 authors including IS • 6 DBs • Single reviewer • CASP	Qualitative plus IS	Commissioned for HTA agency	14 publications, 9 studies; hermeneutic phenomenology (2S, 4P), content analysis [1], mixed method study with content analysis [1], grounded theory (1S, 4P), thematic analysis [2], psychological interview study [1], highly theorized personal narrative [1].
• 3 authors, including 1 advisor and 1 IS • 3 DBs • Single reviewer • CASP	Qualitative plus IS	Commissioned for HTA agency	26 studies, 2 questions; qualitative description [7], mixed methods [3], grounded theory [5], focus group methodology [1], video-reflexive ethnography [1], framework analysis [2], content analysis [3], phenomenology [1], interpretive description [1], and action research [2]
• 3 authors, including IS • 6 DBs • Two independent reviewers • CASP (single reviewer)	Qualitative plus IS	Commissioned for HTA agency	23 publications; Qualitative descriptive designs [6] phenomenologies [7], grounded theory [4], systematic review [1], and mixed methods [5]
• 3 authors including 1 advisor and 1 IS • 5 DBs • Single reviewer • CASP	Qualitative plus IS	Commissioned for HTA agency	18 articles, 15 studies; thematic analysis [8], generic qualitative design [3], phenomenologies [3], content analysis [1]
• 2 authors including IS • 5 DBs • Single reviewer • NR	Qualitative plus IS	Commissioned for HTA agency	12 studies; qualitative descriptive designs [8], phenomenologies [2], grounded theory [1], and cross-sectional design [1]
• 4 authors • 3 DBs • Single reviewer with verification • Framework used	Not stated	Not stated	20 papers; not stated
• 1 author • >7 DBs • NR • Not done	Not stated	Not stated	5 articles (study types not stated)
• 2 authors • 3 DBs • Single reviewer • NR	Staff candidate and supervisors	Chief Scientist's Office Doctoral fellowship	Protocol
• 7 authors • 4 DBs • Single reviewer • Not done	Not stated	Commissioned from RAND Corporation	79 articles
• 3 authors • >7 DBs • Two independent reviewers • NR	Not stated	Commissioned for not-for-profit institute	39 studies; thematic analysis [22], discourse analysis [4], grounded theory [4], phenomenology [2], "Taxonomic" [1], framework analysis [1], not stated [1]
• 3 authors including IS • 6 DBs • Single reviewer • CASP	Qualitative plus IS	Commissioned for HTA agency	7 studies; systematic review [1], phenomenology [2], not reported but descriptive without theoretical orientation [4]
• 3 authors (plus IS) • 3 DBs • Two independent reviewers • CASP single reviewer with verification	Qualitative	UK NIHR funded	20 studies (2 mixed methods, 18 not reported)
• 3 authors (4 researchers) • >7 DBs • NR • Not done	Not stated	Not stated	17 studies (study types not stated)
• 3 authors • 4 DBs • Single reviewer • CASP single reviewer with verification	Student and supervisors	PhD Scholarship	Protocol

undergoing particular interventions or diagnostic tests. Most of the included reviews sought to understand expectations around the outcomes of interventions and which outcomes mattered most to patients. Reviews also sought to explore patient perspectives on the acceptability of interventions and barriers and facilitators to uptake. One review [22] explored help-seeking behavior, and another examined methods to engage patients online in guideline development [24].

4.4. Rapid methods to search for and identify eligible studies

Strategies to search for relevant literature varied across included reviews. Reviewers searched or planned to search three (n = 4 rapid reviews) [17,21,23,27], four (n = 2 rapid reviews) [24,29], five (n = 2) [19,20], six (n = 4) [15,16,18,26], or seven and more (n = 3) [22,23,25] electronic databases, with 21 unique databases being searched across included reviews. Searched databases included MEDLINE (n = 13), PsycINFO (n = 9), CINAHL (n = 9), PubMed (n = 8), The Cochrane Library (n = 5), University of York, Centre for Reviews and Dissemination databases (n = 5), Embase (n = 4), Web of Science (n = 3), ASSIA (n = 2), Scopus (n = 2), Medline (n = 1), TRIP database (n = 1), Science Direct (n = 1), EBSCO (n = 1), SwetsWise (n = 1), JSTOR (n = 1), InformIT (n = 1), JBI Database of Systematic Reviews and Implementation Reports (n = 1), Ovid Nursing Full Text Plus (n = 1), Social Policy and Practice (n = 1), and Web of Knowledge (n = 1). Two reviews [19,27] only undertook electronic database searching, whereas the remainder also searched gray literature (n = 9) [15–18,20–22,26,27], hand-searched specific journals (n = 2) [22,27,30], searched reference lists (n = 3) [21,23,29], and contacted experts (n = 1) [24]. Five reviews limited the number of returned citations: four limited the number of years searched and used language limits [15,16,18,20], and one review limited by country [22]. Ten reviews mentioned involvement of an information specialist or medical librarian to develop and execute the search strategy [15–20,22–24,26], whereas the remainder did not.

Of the thirteen completed reviews, three included less than ten studies [15,22,26], seven included between 11 and 20 studies [16,17,19–21,27,29], one included between 21 and 30 studies [18], one included between 31 and 40 [25] studies, and one included more than 40 studies [24].

4.5. Rapid methods for screening, data extraction, and quality appraisal

Most reviews used a single reviewer for title and abstract screening (n = 10) [15–17,19,20,23,24,26,27,29] and/or full-text screening (n = 9) [15–17,19,20,23,24,26,29]. Two reviews used two independent reviewers for both title and abstract screening and full-text screening [18,25], whereas one review used a single reviewer for title and abstract screening and two independent reviewers for full-text screening [27]. A further review used partial verification, with a primary reviewer screening titles, abstracts, and full text, involving a second reviewer to clarify uncertainties [21]. Two reports omitted details and we were unable to verify the title and abstract or full-text screening processes with review authors [22,28]. See Table 2 for further details.

Similarly, a single reviewer was most commonly used for data extraction (n = 11) [15–21,23,24,26,28] and quality appraisal (n = 8) [15–21,26] with one using two independent reviewers [24]. In two reviews (n = 2) [27,29], a single reviewer extracted data and conducted quality appraisal, whereas a second reviewer verified either all or a random sample of extractions and assessments. Three reviews [22,24,28] did not use quality appraisal. One report did not include any information on data extraction [22], and three [20,23,25] did not include any information on how, or whether, quality appraisal was completed. Eleven of the 12 reviews [15–21,23,25–27,29] for which quality appraisal was undertaken used the Critical Appraisal Skills Programme Qualitative Checklist tool [31,32], and one review [21] used a published framework to guide quality appraisal. Although methodological literature was cited within all 13 reviews (and none of the protocols), none made reference to any methodological guidance or framework specific to the conduct of rapid QES. Instead, references to methodological literature specific to standard QES or the conduct of rapid reviews in general were used, for example, Sandelowski and Barroso (2003) [33], Melia (2010) [34], Petticrew and Roberts (2006) [35].

4.6. Rapid methods for data synthesis

Methods of synthesis included narrative summary (n = 8) [15–18,20–23] with themes from the included articles aggregated within the rapid QES. Four reviews [19,24,26,27] used thematic analysis, two reported using framework synthesis [28,29], and one [25,30] described a metanarrative approach. Where the synthesis approach was not reported, we assessed the output from the synthesis to make a determination [15–18,20,22]. Five reviews [15,22,23,25,27] did not describe any methods used to improve rigor. In four [21,24,26,28], team members examined preliminary results to validate the interpretation of study findings. In another five [16–20], memos and annotation were used to enhance rigor in the coding process. Methodological literature, where cited, referred to the methods for the particular synthesis approach used, with no reference to how it might be applied in a rapid context.

5. Discussion

Through this scoping review, we aimed to identify and describe methodological guidance for the rapid conduct

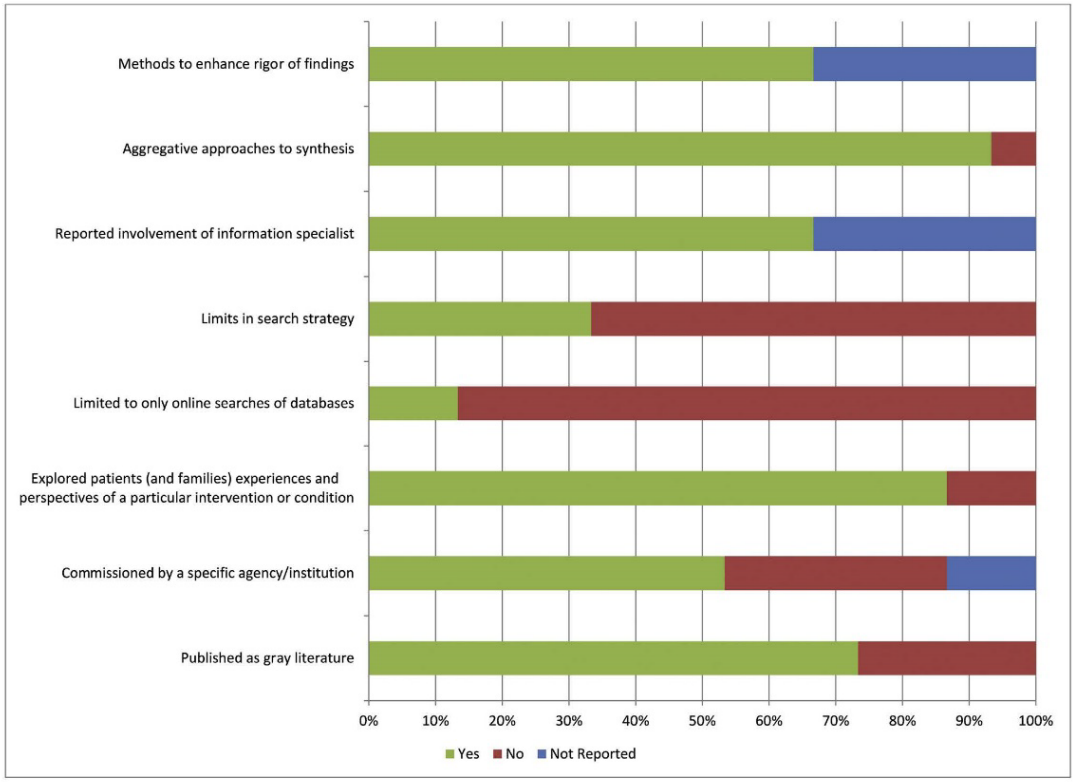


Fig. 2. Characteristics of example rapid qualitative evidence syntheses.

of QES, as well as the methods used, or planned, within published examples. We sought to examine the extent to which current guidance and practice offers a methodological evidence base to facilitate common expectations and methods for a rapid QES process.

We did not identify any guidance specific to rapid QES, although we identified 15 examples of rapid QES, including 13 reports of completed reviews [15–22,24–28] and two protocols [23,29]. Examples were planned or undertaken and published in the past 8 years, many being published as gray literature in online databases and websites. Over half were undertaken as specific commissions to guide decision-making in health policy [15–20,25,26].

Our team initially assumed that published rapid QES was more prevalent than established by this scoping review. Published reviews retrieved through our literature search commonly used both quantitative and qualitative studies within a rapid "mixed method review," which were not eligible for this review. It is possible that these reviews sought to address multiple aspects of decision support rather than a single issue or perspective [36]. Similarly, when asked for published examples, experts did not explicitly identify or report the use of methods to abbreviate or accelerate the review process in the examples they identified. Markers of rapidity may have been withheld to increase the likelihood of publication, or a rapid QES may have subsequently been upgraded to a full synthesis before publication. As a consequence, the actual prevalence of published rapid QES remains unclear, with the sample identified for this review likely underrepresenting actual numbers. Given what appears to be an increase in the incidence of rapid QES in recent years, and an associated demand for such evidence to support decision making, there is a clear need to develop methodological and reporting guidance that reflects the nature of qualitative inquiry and preserves its iterative, inductive, and interpretive qualities.

Where reported, the included reviews were conducted within less than 6 months. To meet these short time frames, reviewers used methods to increase the speed and efficiency of the review process. Our results suggest that QES review teams are largely borrowing rapid methods from the wider rapid review community; for example, by imposing date limitations on the search and including only articles published in English. Although gray literature was commonly searched, search methods did not typically extend to more time-consuming activities such as hand-searching journals or approaching experts. Interestingly, the number of databases searched seems to suggest that searches were not

typically limited to one or few, databases. It is possible that qualitative search results may be less plentiful, hence requiring more extensive searching to locate, or experienced teams of reviewers may include information specialists familiar with, and with access to, multiple databases. It is established that qualitative research is less readily located within biomedical journal databases [37] and so compromises in the number of databases may risk missing relevant studies. Perhaps accordingly, researchers engaged in QES seem to share with clinical reviewers a concern with being comprehensive and not "missing" eligible studies.

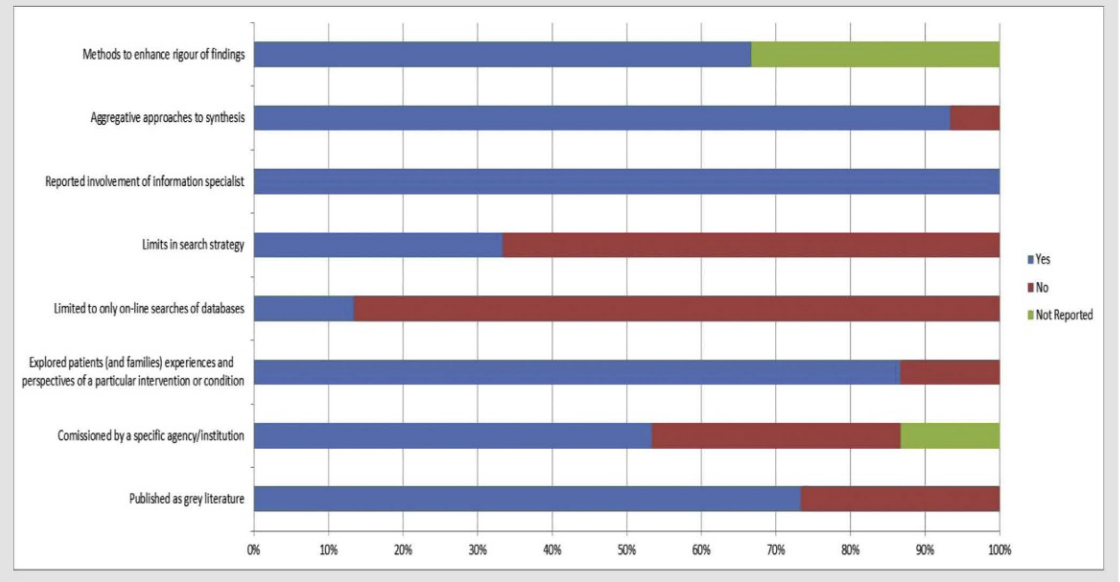
Quality appraisal was omitted or not reported in approximately one-third of identified examples, mirroring a review of classic rapid review methods which suggests that quality appraisal was omitted or not reported in 24% of examples [38]. Given the debates within the broader QES community regarding methods for, and the importance of, quality appraisal [34], it is surprising that this step was not more commonly omitted.

Although mirroring methods used for the rapid review of evidence of intervention effects may offer a reasonable starting point for a QES, it remains unclear which shortcuts, if any, are appropriate and which require further examination. For example, does the increased error rate of single reviewer citation screening and data extraction [39] translate to qualitative data and, if so, what are the implications of errors in qualitative data extraction? Typically, in qualitative analysis, data collection, coding, and interpretation concentrate on reflection and discussion within the team and engaging with other literature, as opposed to identifying, counting, and correcting errors. The effects of having a limited opportunity for reflection and discussion cannot be quantified in a QES. However, we can hypothesize that limited reflection and discussion will lead to a superficial analysis, with the potential loss of additional insights and interpretations.

Booth et al. (2018) [10] itemize how nineteen QES methodologies divide into aggregative approaches that aim to describe the findings of the primary studies or interpretive approaches that aim to develop a new conceptual understanding or "theory" [40]. Similarly, Thomas et al. (2017) [41] suggest that methods of synthesis lie on a continuum from mostly unchanged (aggregating categories or findings within "thematic summaries") to mostly emergent (de novo analysis and conceptual ordering as for "metaethnography"). The predominance of aggregative synthesis within rapid QES, as identified through this scoping review, contrasts with extensive use of interpretative approaches within full QES, where metaethnography is at least as common as thematic approaches [42]. Aggregative approaches are similarly more common in rapid quantitative reviews with 78% of rapid reviews using narrative or descriptive summary and meta-analysis occurring less commonly [43].

Incomplete description of individual methods or approaches of synthesis and gaps in the empirical base that underpins them have been targeted for a potential research agenda [10], and this extends to rapid QES. We identified several examples where authors omitted methodological details (e.g., the number of reviewers used or the approach to critical appraisal). Although authors were forthcoming about their methodological approach once we contacted them for further details, it is unclear if this omission sought to mask the rapid nature of the review to optimize subsequent

Table 2. Summary of findings



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Interventions for the prevention of spontaneous preterm birth: a scoping review of systematic reviews

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ABSTRACT

Background Globally, 11% of babies are born preterm each year. Preterm birth (PTB) is a leading cause of neonatal death and under-five mortality and morbidity, with lifelong sequelae in those who survive. PTB disproportionately impacts low/middle-income countries (LMICs) where the burden is highest.

Objectives This scoping review sought to the evidence for interventions that reduce the risk of PTB, focusing on the evidence from LMICs and describing how context is considered in evidence synthesis.

Design We conducted a scoping review, to describe this wide topic area. We searched five electronic databases (2009–2020) and contacted experts to identify relevant systematic reviews of interventions to reduce the risk of PTB. We included published systematic reviews that examined the effectiveness of interventions and their effect on reducing the risk of PTB. Data were extracted and is described narratively.

Results 139 published systematic reviews were included in the review. Interventions were categorised as primary or secondary. The interventions where the results showed a greater effect size and consistency across review findings included treatment of syphilis and vaginal candidiasis, vitamin D supplementation and cervical cerclage. Included in the 139 reviews were 1372 unique primary source studies. 28% primary studies were undertaken in LMIC contexts and only 4.5% undertaken in a low-income country (LIC) Only 10.8% of the reviews sought to explore the impact of context on findings, and 19.4% reviews did not report the settings or the primary studies.

Conclusion This scoping review highlights the lack of research evidence derived from contexts where the burden of PTB globally is greatest. The lack of rigour in addressing contextual applicability within systematic review methods is also highlighted. This presents a risk of inappropriate and unsafe recommendations for practice within these contexts. It also highlights a need for primary research, developing and testing interventions in LIC settings.

Strengths and limitations of this study

⇒ Scoping review methodology enabled us to look at a broad topic area and analyse how context is taken into account in the included systematic reviews. Primary studies not reported in systematic reviews will therefore have not been included in our analysis.
⇒ We were not able to identify the setting of all primary studies where this was not reported and there is a risk that some studies, which have multiple publications may have been double counted.
⇒ We only included systematic reviews published in English.

BACKGROUND

Preterm birth (PTB) is a global and public health priority. It is defined by the WHO as delivery before 37 completed weeks of gestation, with extremely preterm delivery defined as occurring at less than 28 weeks, very preterm delivery occurring between 28 and 32 weeks, and moderate to late preterm delivery occurring from 32 through 36 weeks.[1] It is one of the leading causes of neonatal death and under-five mortality and morbidity, with lifelong sequelae.[2] Children born prematurely have increased risks of cognitive problems, such as academic underachievement, behavioural problems and cerebral palsy than those born at full term.[3] They are more likely to experience hospital admission due to infection, particularly during infancy.[4] For parents, the financial, social and emotional effects are devastating.[3]

The global burden of preterm birth (PTB) is falling more heavily on countries with fewer resources to manage the medical, social and economic complexities of caring for premature infants. Globally, there are approximately 15 million live PTBs each year, which is estimated to be about 11% of all deliveries each year, ranging from about 8.7% in northern Europe to 13.4% in North Africa.[5 6] The majority of PTBs occur in low/middle-income countries (LMICs).[6] The highest PTB rates in 2014 occurred in South-east Asia, South Asia and sub-Saharan Africa.

Nine of the 11 countries with the highest rates were in Africa. Furthermore, 60% of all PTBs were estimated to have occurred in sub-Saharan Africa and South Asia accounting for just over 9 million of the almost 15 million PTBs that occurred worldwide in 2010 resulting in a PTB rate of 12.8% in those settings.

Patterns of PTB differ between high-income countries (HICs) and LMICs. However, the differences in these patterns, causes and distribution of PTB is unclear and have not been fully explored. PTB is multifactorial in its aetiology and has distinct biological pathways. The aetiologies differ according to gestational age, ethnicity and characteristics unique to each population. In order to redress the burden of PTB in LMICs, additional insight into the causative and associated factors in these settings is required.

While a number of reviews and overviews of reviews of interventions to reduce the risk of PTB have been undertaken,[7–10] none have explored how many of the primary studies included in these reviews were undertaken in LMIC contexts. It is clear that some interventions that are effective in HIC contexts but may be harmful in LMIC settings, such as the use of antenatal corticosteroids[11] and cerclage.[12] It is also possible that treatments effective in HIC contexts may be even more beneficial or appropriate in LMIC contexts, such as nutritional supplements, interventions to increase birth spacing or interventions to improve the accuracy of measuring gestational age.

We have undertaken a broad scoping review of systematic reviews on interventions to reduce the risk of PTB identifying primary studies undertaken in LMICs. This will allow us to identify potential areas for further synthesis of the evidence and also to identify gaps in the research in order to direct future primary research.

Review objectives

1. To identify systematic reviews that have sought to explore the effectiveness, safety and acceptability of interventions to prevent PTB.
2. To map research evidence to global settings to identify the geographical and economic contexts in which evidence is derived.
3. To identify where gaps in the research base exist (for real world, effectiveness, pragmatic studies) in LMIC contexts to inform future research and to generate research priorities.
4. To describe the methods used in meta-analysis to take into account geographical and regional differences in PTB.

METHODS

We used a scoping review methodology[13] to describe the existing evidence (systematic reviews) available across primary and secondary interventions to prevent PTB, published between 2009 and 2020. Systematic scoping draws on methods described by Arksey and O'Malley[14] for scoping reviews: '[…] a form of knowledge synthesis that addresses an exploratory research question aimed at scoping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge'.[14] The approach enabled us to highlight the evidence gap and to assist with simultaneously undertaking a research prioritisation exercise and guideline development, as well as to inform a broader programme of research that aimed to develop effective postnatal interventions to mitigate PTB in LMIC settings. It also enabled us to generate a mega-map, an interactive table supported on our project website and designed as a visual tool to identify research gaps and facilitate ready access to relevant evidence (https://www.primeglobalhealth.co.uk/evidence-map-2-7-2020.html).

Identifying relevant studies

Relevant systematic reviews were identified by systematic searches in the following electronic databases: Ovid MEDLINE, Cochrane Database of Systematic Reviews, PsycINFO via Ovid, EMBASE via Ovid and CINAHL via EBSCO. Each database was searched using the database thesaurus and the key word/free text method with terms relating to PTB combined with a systematic reviews filter. The search strategy incorporated the following limitations: articles written in English, and Human studies only from April 2009 to July 2020. Relevant systematic reviews were identified by systematic searches in the following electronic databases: MEDLINE, The Cochrane Library, PsycINFO, EMBASE and CINAHL. Each database was searched using the database thesaurus and the key word/free text method. The search strategy incorporated the following limitations: articles written in English, and Human studies only from April 2009 to July 2020. The date limit was selected due to the existence of a previous review for which the studies were conducted in April 2009.[15] Full search strategies have been described and published.[16]

We began with a framework of interventions identified by two existing reviews[7 8] as these were broad in their focus and encompassed a range of interventions. Any new intervention types identified during the screening process were then added to the map.

The process of study selection was based on inclusion and exclusion criteria as described in box 1. After removal of duplicates and irrelevant studies, based on the titles and abstracts, all potentially relevant reviews were read in full. Citations were screened by two reviewers (FC and one of the following team members SS, SMJ, EA, JB, BMG, BN, KP) independently and differences were resolved by discussion.

Data extraction and coding

Data were extracted using an agreed and piloted template and coded in Excel by two reviewers working independently (FC and one of the following team members SS, SMJ, EA, JB, BMG, BN, KP) differences were resolved by discussion. The following data categories were extracted:

Box 1 Inclusion/exclusion criteria based on PICOS

Population
⇒ Pregnant women at less than 37 completed weeks gestation without signs of threatened preterm labour or premature rupture of membranes.
⇒ Excluded reviews where the study population was defined by comorbidities.

Intervention
⇒ All interventions deliverable during pregnancy to prevent spontaneous preterm birth (PTB) (these included clinical, behavioural and nutritional interventions and health systems and policy interventions).
⇒ All interventions assessed the risk of PTB.
⇒ Excluded interventions given to pregnant women to improve neonatal outcomes.

Comparators
⇒ We included any comparator, including placebo or alternative treatments.

Outcomes
⇒ We included reviews which focused on PTB as an outcome.
⇒ Where it is reported, we state how many of the primary studies measured PTB as an outcome and the resulting data used in the synthesis.

Study design
⇒ Systematic reviews published between April 2009 and July 2020, of studies that have evaluated interventions to prevent PTB, or that measured PTB as a relevant outcome.

Outcomes
⇒ PTB (<28, <34, <37 weeks gestation) .
⇒ We recorded neonatal outcomes and adverse outcomes if reported within the review.

number of included studies, review PICO, setting of primary studies and any analysis that took into account study setting or population characteristics, PTB outcomes, assessment of adverse effects and recommendations for practice and research. PTB rates in low-income countries (LICs), lower middle-income countries (LMCs), upper middle-income countries (UMCs) and HICs settings were drawn from data published in a rigorous review of national civil registration and vital statistics to determine global, regional and national estimates of levels of PTB.[6]

Where reported information allowed, we used the World Bank categories to identify the categories of all country settings identified in the reviews.[17]

The population, interventions, comparators, outcomes and reviewer conclusions for future research were tabulated and described narratively. The country or countries of the included primary studies were noted, and the methods used in the review for analyses of data from different settings was also recorded and described. We did not contact review authors for missing data.

Patient and public involvement

This review was undertaken as part of a larger programme of research in PTB (NIHR Global Health under grant (17/63/26)). The programme iPatient and public involvements informed by key stakeholders and a patient and public involvement (PPI) advisory group comprising representatives from Sheffield, Bangladesh, and South Africa. The design and questions for the review were informed by consultation with these groups.

RESULTS

Our search identified 3133 citations which were screened by two reviewers. A third reviewer was also involved where there was a lack of consensus or uncertainty regarding inclusion. Following screening, 424 full text papers were retrieved for data extraction. At data extraction a further 285 were excluded. The process of identifying the included reviews is summarised in figure 1.

We included 139 reviews which addressed a range of primary and secondary interventions and measured the effectiveness of the intervention in reducing the risk of PTB. These are summarised in table 1. There was a considerable variation in the number of included studies in the reviews for each intervention, reflecting differing research questions objectives (therefore different PICOs) and search strategies.

Context of primary studies

A total of 1372 primary studies were included across all of the 139 reviews Not all of these studies will have been measuring PTB as an outcome but were included within the review which may have been measuring a range of maternal outcomes including PTB. The largest number of primary studies were those evaluating micronutrient supplements (n=481) and tocolytics (n=167). A total of 113 of the reviews described the country in which the primary studies were undertaken and so these data were known for 1288 (93.9%) of 1372 included primary studies. Of these, 390 (30.3%) were undertaken in LMICs, 15 primary studies were multicentre and included data gathered from LMIC and HIC settings, though only 3 of these studies included LICs. Of the studies undertaken in LMICs, a majority (n=255) examined the effects of nutritional supplements. Excluding nutritional intervention studies, the proportion of LMIC-based primary studies of interventions to reduce PTB accounts for only (n=135) 10.5% of the included studies where settings are known.

Of the total number of primary studies undertaken in LMIC contexts, those studies undertaken in LIC settings represented a very small proportion of included studies. Participants from LICs were represented in only 4.5% (n=58) of the total number of studies, and if the nutritional intervention studies are excluded, they account for only 2.5% (n=32) of the studies evaluating interventions. Of those primary studies that were undertaken in LMIC settings the numbers within each country category differed significantly. The proportion of the studies that are undertaken in LIC, LMC and UMC were 14.9% (n=58), 34.8% (n=136) and 50.2% (n=196), respectively. There are only single trials that have evaluated the impact

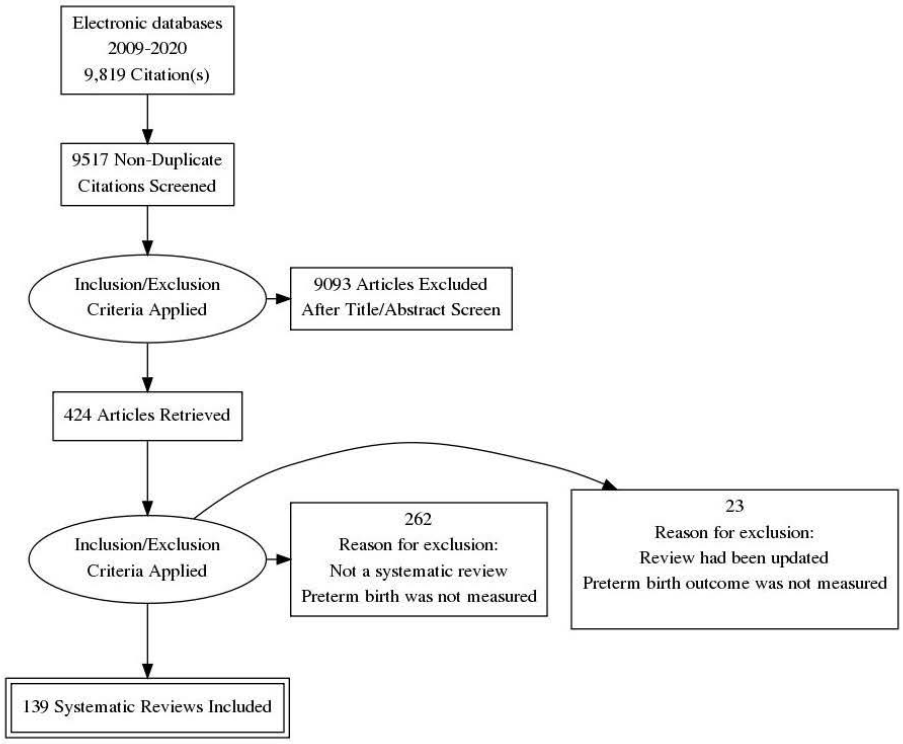

Figure 1 Flow of studies through review process.

of progesterone, tocolytics and interventions to increase calorie intake in LIC settings. There are no trials that have evaluated smoking cessation, preventing excessive weight gain, prevention and treatment of periodontal disease, influenza vaccine and cervical pessaries. The number of trials in each of the country categories within each intervention type are shown in table 1.

When these data are compared alongside data that shows the prevalence of PTB globally it is clear that there is an inverse pattern in the distribution of the data (figure 2).

Effectiveness of interventions

The effectiveness of interventions in reducing the risk of PTB was variable with no intervention showing consistent effectiveness across the included reviews. Although interpretation of these data is limited by the lack of quality appraisal of the included reviews, and therefore should be viewed with caution. Overall, the scoping review demonstrates considerable inconsistency of results of interventions. Of the 139 reviews, 28 reported a reduction in PTB in intervention versus control, 80% (n=111) of the reviews found that the intervention had no impact in reducing the risk of PTB. The summary result (relative risk (RR) and OR are shown in figure 3). The results show the reduction in PTB less than 37 weeks gestation. In three reviews the intervention was not statistically significant at 37 weeks but was reported as statistically significant at 34 weeks,[18] 35 weeks[19] and 36 weeks[20]. Two reviews reported a positive effect of the intervention in reducing risk of PTB but reported the outcome on a continuous measure. These included the effectiveness of macronutrient supplements[21] (SMD −0.19 (95% CI −0.34 to −0.04)) and cerclage (mean difference 95% CI 33.98 days (17.88 to 50.08)).[22] The interventions reporting binary outcomes which appear to have the greatest effect (RR=0.2–0.4) in reducing PTB are: antibiotics for asymptomatic bacteriuria[23] (RR=0.34 (95% CI 0.11 to 0.62), the screening and treatment of syphilis[24] (RR=0.36 (95% CI 0.27 to 0.47), and treatment of vaginal candidiasis[25] (RR=0.36, (95% CI 0.17 to 0.75). Interventions with moderate effects (RR=0.4–0.6) included treating lower genital tract infection[26] and vitamin D supplements.[27] Four of the reviews (figure 2) with a positive effect of the intervention considered that the strength of evidence supporting the finding could be considered high and the finding reliable. None of these reviews included studies conducted in LIC settings, and only one included one study in an LMIC.

Dealing with context and generalisability within evidence synthesis

The authors of the included reviews used different approaches to dealing with the contextual variation when

Table 1 Summary of included systematic reviews and settings of primary studies included in the review

Interventions	Number of reviews	Number of primary studies	Country NR	Country of primary study					Studies where setting NK
				LI	LM	UM	HI	Mixed	
Primary prevention interventions									
Health systems									
Models of antenatal care delivery (group/ specialised)[61–71]	11	68	2	0	2	2	64	0	0
Midwifery led care[72]	1	15	0	0	0	0	15	0	0
Improving ANC coverage[28]	1	34	0	10	15	5	0	0	0
Health behaviours									
Smoking cessation[35 73]	2	111	0	0	0	1	110	0	0
Weight management[21 74–78]	6	70	1	0	2	8	60	0	0
Nutritional interventions									
Macronutrient supplements[29 30]	2	34	0	3	9	10	8	4	0
Micronutrient supplements[21 27–31 35–40 67–86]	33	481	2	29	82	122	214	6	9
Vitamin D[27 31 36 79–81]	6	75							
Vitamin A[37 82]	2	24							
Vitamin E, C, E and C[38 39 83]	3	67							
Iron, folic acid, iron and folic acid[40 84–90]	8	182							
Fish oil[91–95]	5	38							
Zinc[32 96]	2	25							
Calcium[41 42]	2	27							
Iodine[97]	2	14							
Multiple micronutrients[43 44 98]	3	29							
Screening and treatment of periodontal disease[99–110]	12	46	0	0	3	7	36	0	0
Screening and prevention/ treatment of infection	14	91	2	2	2	6	79	0	2
Asymptomatic bacteriuria[23 111–113]	4								
Screening and antibiotics for syphilis[24]	1								
Influenza vaccine[114 115]	2								
Lower genital tract infection[26]	1								
UTI[116 117]	2								
Vaginal candidiasis[25]	1								
Non-specific infection[118 119]	2								
Malaria[33 120 121]	3	17	0	8	7	2	2	0	0
Secondary prevention interventions									
Cerclage[18 22 45 122–136]	18	123	10	0	7	11	42		51
Bed rest[137–139]	3	40	1	4	0	0	36	0	0
Cervical pessary[140–145]	6	16	0	0	0	1	14	1	0
Progesterone[19 20 146–159]	16	59	5	1	7	8	28	4	11
Tocolytics[160–172]	11	167	3	1	0	13	68	0	84

ANC, antenatal care; HI, high income; LI, low income; LM, low middle; NK, not known; NR, not reported; UM, upper middle; UTI, urinary tract infection.

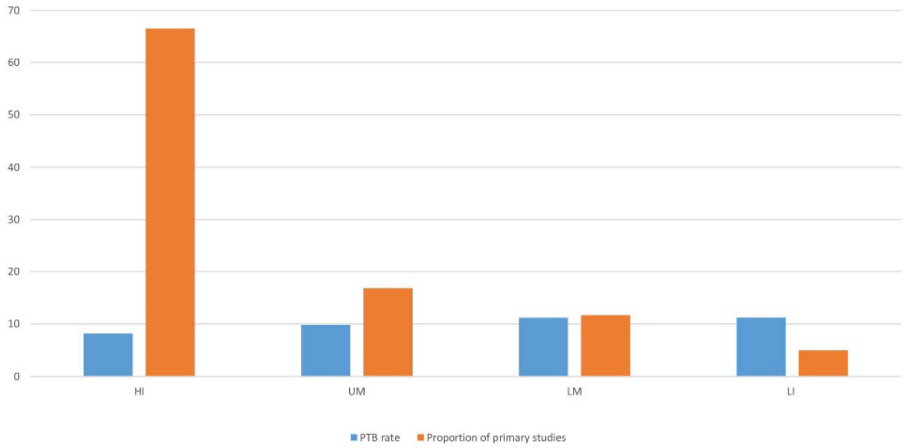

Figure 2 Rates of PTB and proportion of primary studies undertaken in each setting. HI, high income; LI, low income; LM, low middle; PTB, preterm birth; UM, upper middle.

pooling data from primary studies, which was either to ignore, document, explore or control for differences. Twenty-seven reviews (23.8%) did not describe the setting of the primary study, ignoring variation in outcomes that may arise as a result of these differences. This occurred most frequently in reviews of cervical cerclage (see table 1). The majority of the included reviews 86 (76.1%) documented the country in which the primary study was carried out either within the text, tables of study characteristics or in accompanying appendices, but this was not considered further in terms of its implications for the findings, or application for future practice or research.

Eight reviews[27–34] sought to explore the impact of geographical and economic context by undertaking a subgroup analysis comparing trials conducted in low income settings with those in high income settings or regression analysis with geographical regions as covariates (Africa, Americas, Southeast Asia, Europe, Eastern Mediterranean, Western Pacific). In addition, one study[34] listed the country instead of the author name on the forest plot allowing ready visualisation of differences across settings. Nine reviews[35–43] undertook subgroup analysis based on features of the population that might vary across settings and influence the effectiveness of the

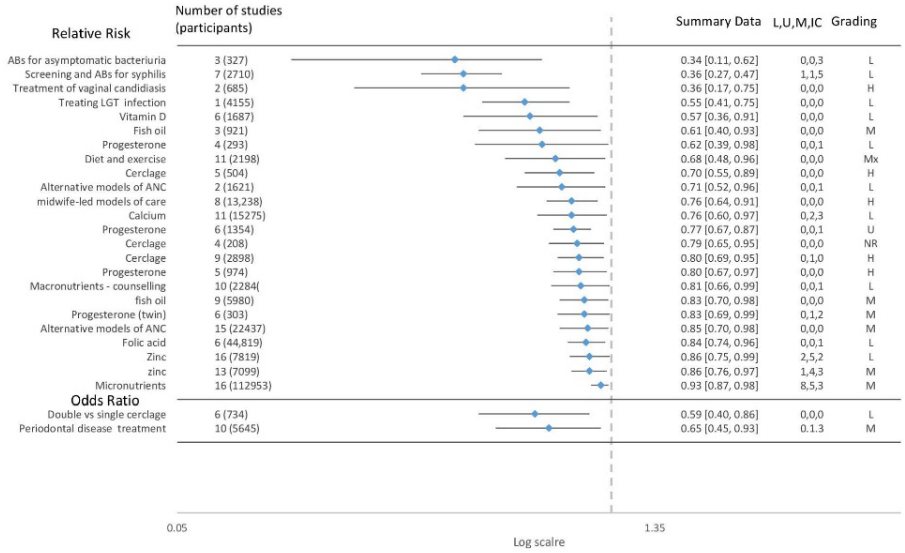

Figure 3 Summary results of systematic reviews of interventions showing reduction in risk of preterm birth. ANC, antenatal care; L, U, M, IC, low, low middle, upper middle-income countries; LGT, lower genital tract; RR, relative risk.

intervention, such as baseline nutritional status of the mother. One review[44] exploring multiple micronutrient supplementation controlled for settings by limiting the review to include only those studies undertaken in LMIC contexts. Four reviews[19 20 44 45] undertook an IPD (individual patient data) analysis, allowing subgroup analyses about differences in effect more easily than with aggregate data. This approach allowed comparison between effects for women recruited and receiving the intervention in different settings, effect sizes in each country could also be shown in the analyses.

DISCUSSION

This scoping review has revealed an inverse pattern of research, with only 30.3% of published research included in systematic reviews of interventions reporting PTB outcomes carried out in LMIC settings, and only 4.5% was conducted in the poorest countries in the world where the burden of PTB is greatest. The distribution of types of intervention tested and evaluated in these settings is not even across interventions, but is largely focused on very context specific interventions (prevention of malarial infection) and nutritional supplementation. Similar patterns of a mismatch between research effort and health needs in non-high income regions have been identified across a broad range of diseases.[46 47] It has also been previously reported that primary research often fails to capture those with the greatest healthcare needs such as vulnerable populations.[48 49]

This review has also revealed a limited approach in evidence synthesis to explore the applicability of findings across geographical settings and to draw attention to these gaps with a resultant risk that interventions shown to be effective in HI settings may not translate to LIC settings and may indeed have adverse effects when applied to LIC settings. Likewise, the focus of research in HIC settings means that interventions that may have greater benefit in LIC settings—where the problem is greatest—remain untested or replicated with larger numbers of participants. Adolescent pregnancy and short inter pregnancy intervals, both of which are more common in LMICs, have been highlighted as important risk factors for PTB[50] yet there is a lack of data on interventions to address these and their effectiveness in reducing the risk of PTB.

The lack of robust evidence to inform both the primary and secondary prevention of PTB in LIC settings, where the prevalence of PTB is highest presents challenges for developing appropriate and contextually relevant clinical guidance. The factors that mean findings cannot be generalised from high resource settings to low and middle resource settings are multiple and will differ across interventions. Ethnicity, poverty, gender dynamics, pollution, temperature, climate, diet, access to healthcare, educational status, employment conditions are all examples of factors that might play a role in these differences. Improved understanding of the aetiopathogenesis of PTB is also necessary for defining an accurate model of risk prediction and would help in understanding what factors in local settings increase risk and facilitate the development of an accurate model of risk prediction.[51]

Two recent overviews of reviews[9 10] also found that few interventions are effective in PTB prevention. The following interventions were identified in these reviews as showing positive or possible benefit: lifestyle and behavioural changes (including diet and exercise); nutritional supplements (including calcium, zinc and vitamin D supplementation); nutritional education; and screening for lower genital tract infections. Positive effects of secondary interventions were found for low dose aspirin among women at risk of pre-eclampsia; clindamycin for treatment of bacterial vaginosis; treatment of vaginal candidiasis; progesterone in women with prior spontaneous PTB and in those with short mid-trimester cervical length; L-arginine in women at risk for pre-eclampsia; levothyroxine among women with thyroid disease; calcium supplementation in women at risk of hypertensive disorders; smoking cessation; cervical length screening in women with history of PTB with placement of cerclage in those with short cervix; cervical pessary in singleton gestations with short cervix; and treatment of periodontal disease. Our review findings were in concordance, although, in addition, we identified screening and antibiotic treatment for syphilis, and positive effects of fish oil supplements. In most instances the trials were small and authors recommended larger well-designed randomised controlled trials (RCTs). The lack of consistency across review findings for interventions also merits more exploration. Compromised methodological rigour can inflate trial findings by 30%–50%.[52 53] Some of the differences in our review findings reflect some differences in the included reviews.

The interventions identified in this review, and those of Matei *et al*[9] and Medley *et al*[10] informing guideline development, clinical practice and policy decision making have been little tested in LMIC settings. In those interventions where there is more consistency in review findings such as cervical cerclage, there are no studies that have been conducted in low-income settings and over half of the reviews did not report or consider settings in their analyses.

This scoping review has shown that many authors of systematic reviews fail to use design and statistical approaches that adequately address contextual variations between the included source studies and imperfectly represent 'real world' conditions within the target context. While those reviews that sought to take into account LMIC contexts were unable to conduct the analyses due to a lack of data, they nonetheless were able to highlight the gaps in research, for example the lack of studies in vitamin D undertaken in Africa.[31]

The Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) reporting standards reference 'context' in terms of the circumstances requiring the review itself, rather than referencing the contexts of studies included in the review.[54] The PRISMA extension

for Complex Interventions includes the elements of 'time' and 'setting'.[55] However, grouping LMIC data, or even LI data may still be too broad. Even within the categories of LIC there is considerable diversity that may impact on how an intervention works and within countries there may also be considerable diversity between the wealthiest and poorest groups. For example, the time taken to reach comprehensive emergency obstetric care facilities in low resource settings is often underestimated and for most women is likely to be 120 min of travel time.[56] Context cannot be standardised, it will vary from review to review, as different interventions and different populations are considered. 'Context' and the factors that might influence the efficacy, uptake, acceptability, appropriateness, accessibility and availability of an intervention requires a good understanding of the aetiology and mechanisms by which risk factors interact with environmental, microbial, socio-political and health system variations across settings.[57]

It must be acknowledged that there are significant barriers to undertaking research in many settings across the globe. These include very practical challenges such as a lack of access to high-quality data and the challenges of estimating gestational age.[58] Recent changes to global health funding arena include a very large proportion being spent on the pandemic as well as government reductions, for example, in the UK.[59] These reductions in funding will undermine what has been a growth in research in LMIC settings and will impede efforts to address the imbalances highlighted in this scoping review.

A number of limitations exist in this scoping review. We have not sought to identify the setting of primary studies where this is not reported in the systematic review. We have also not limited our analysis to studies within the reviews that only contributed findings to the risk of PTB. Most reviews explored several maternal and infant outcomes. Therefore, in this scoping review, included primary studies may not have contained PTB outcome data. We limited our scoping review to exploring evidence within systematic reviews as these are key sources of evidence to inform guideline development and policy decision making. It is possible that further primary studies have been published but are not included in this analysis. Nevertheless, it gives an indication of the distribution of research being undertaken in the poorest regions of the world that address PTB.

CONCLUSION

Only 4.5% of primary research to examine the effectiveness of interventions to reduce the risk of PTB is carried out in settings where the burden is greatest. No interventions which reduce the risk of PTB, judged to be supported by strong evidence, include studies undertaken in low resource settings. In the synthesis of studies, current methods often fail to address the contextual variation and consider the applicability of findings in low resource, high burden settings. This has implications for supporting policy making, and development of contextually relevant clinical guidelines. While methods can be undertaken to improve approaches to evidence synthesis, they cannot compensate for the lack of primary research in low resource settings. This is critical if global health inequalities are to be addressed and millennium development goals[60] to reduce under-five mortality are to be achieved. Funding and supporting research in LMICs would have a threefold benefit; first, if the prevalence of the disease is higher it is easier to reach statistical significance for efficacy or inefficacy of each tested intervention. Second, it would address the knowledge gap highlighted in this review and finally—and most importantly—the implementation of effective interventions would have the potential for greater public health impact where the risks are greater, more prevalent and outcomes more severe.

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Paper D

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RESEARCH ARTICLE

Research Synthesis Methods WILEY

The ScHARR LMIC filter: Adapting a low- and middle-income countries geographic search filter to identify studies on preterm birth prevention and management

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Abstract

Search filters are used to find evidence on specific subjects. Performance of filters can be varied and may need adapting to meet the needs of research topics. There are limited geographic search filters available, and only one pertaining to low- and middle-income countries (LMICs). When searching for literature on preterm birth prevention and management in LMICs for a research project at the School of Health and Related Research (ScHARR), we made use of the Cochrane Effective Practice and Organisation of Care (EPOC) LMIC geographic search filter for the databases; Ovid MEDLINE, Ovid Embase, Cochrane Library. During screening following a broad scoping search in Ovid MEDLINE, it was found that the EPOC LMIC filter did not identify a relevant study. Adaptations were made to the LMIC geographic search filter to maximise retrieval and identify the missing study. Institution was included as a search field, and the search terms high burden or countdown countries were added. The filter was translated for the databases; Ovid Embase, Cochrane Library, Ovid PsycINFO, and CINAHL via EBSCO. The adapted ScHARR LMIC filter is a non-validated 1st generation filter which increases the sensitivity of the EPOC LMIC search filter. Validating the filter would confirm its retrieval performance and benefit information professionals, researchers, and health professionals. We recommend that the ScHARR LMIC filter is used to improve sensitivity of the Cochrane EPOC LMIC filter and reduce the risk of missing relevant studies.

KEYWORDS

evidence synthesis, information retrieval, literature searching, low-and middle-income countries, search filters, systematic reviews

Highlights

What is already known

- The Cochrane EPOC LMIC geographic search filter is currently the only pre-made search filter to retrieve literature from low- and middle-income countries. The filter has not been validated.

What is new

- We present a useful adaptation of the Cochrane EPOC LMIC geographic search filter (the ScHARR LMIC filter), which may identify further studies and therefore be more comprehensive. The ScHARR LMIC filter can be used in Ovid MEDLINE, Ovid Embase, Ovid PsycINFO, Cochrane Library, and CINAHL via EBSCO.

Potential impact for research synthesis methods readers outside the authors' field

- Information specialists, researchers, and healthcare professionals' wishing to locate evidence in LMICs may find our revised geographic search filter useful.

1 | INTRODUCTION

1.1 | Search filters

Evidence synthesis requires the design of effective search strategies to identify the relevant evidence of interest.[1] Information specialists usually perform this task and use various tools and techniques to ensure that search strategies meet the requirements for the scope of the review. One method is to combine existing search filters with the search terms developed according to the review question.

Search filters are pre-made search strategies designed to retrieve particular types of study designs or topics, from bibliographic databases.[2] There are three search filters categories (see Table 1).

It is important for systematic searches to strike a balance between sensitivity (minimising the risk of failing to retrieve relevant records) and specificity (minimising the number of irrelevant records in order to manage "screening burden").[4] Appropriate use of search filters can assist with this challenge. Search filters are often developed for use in a particular context, and their effectiveness may be informally evaluated by comparing their results against a list of known studies to see what proportion are retrieved (1st generation search filter). Ideally, search filters are "externally validated", that is, tested against a "gold standard" reference set of results from outside the context for which they were developed (2nd generation search filter).[5] This gives a more rigorous assessment of how the filter performs overall, including in contexts beyond the one in which it was developed. Performance of search filters are usually measured in terms of sensitivity, specificity, and precision. See Table 2 for definitions of search filter performance measures.

Search filters are often adapted to suit the needs of research topics,[4,6] particularly if there are concerns that a pre-made filter will not retrieve some of the relevant studies. Cooper et al.[7] conducted a case study, which found that two established and widely used randomised controlled trial (RCT) filters missed studies, and therefore recommended incorporating an additional search filter they developed to improve sensitivity. Other studies have reported low precision of filters.[8] Search filters are often developed to find evidence in the context of a specific health speciality, for example, the Africa geographic filter was developed to find RCTs on HIV/AIDS[9] and therefore may not be as effective for other areas.[10] The choice of filter will be influenced by the type of the review, the aim, and the time and resources available. The most appropriate filter should be selected to meet to needs of the scope of the research topic. For example, a rapid review may favour high precision over sensitivity, and there may be some types of reviews, where use of filters are not recommended.[11] Formal appraisal of existing filters[3] will assist information specialists making decisions regarding choice of filter and potential adaptation. Both the Canadian Agency for Drugs and Technologies in Health and the InterTASC Information Specialists' Sub-Group (ISSG) have developed checklists to use to appraise search filters.[12,13] However, due to the potential time constraints of reviews, particularly those of a rapid nature, published appraisals are of benefit, and the ISSG collating and linking to these is an incredibly valuable resource.[14]

TABLE 1 Search filter categories[3]

1st generation: non-validated	Designed subjectively and not tested for effectiveness against an independent set of relevant records (validation)
2nd generation: validated (subjective methods)	Developed as 1st generation, but effectiveness is tested against an independent set of relevant records (known as a "gold standard"), resulting in a validated filter
3rd generation: validated (objective methods)	Designed more objectively through various established methods, such as frequency analysis and logistic regression. The designed filter is then validated as in 2nd generation filters

TABLE 2 Search filter performance measures[5]

Measure	Definition
Sensitivity (recall)	Number of records retrieved by the filter as a proportion of the gold standard set of references
Precision	Number of relevant records retrieved as a proportion of the total records retrieved
Specificity	Number of irrelevant records correctly not retrieved, as a proportion (usually expressed as a percentage) of all the irrelevant records in the database searched

The most common type of search filter is methodological,[14] and therefore retrieves specific study types, such as systematic reviews or RCTs. Methodological search filters are well-established and used, and there are various studies evaluating their performance, including a review published in 2004.[3] Topic search filters are less common, but various ones have been developed covering areas, such as geographical settings, age groups, or specific conditions.[15]

1.2 | Geographic search filters

The ISSG Search Filters Resources is a comprehensive catalogue of filters, including citing papers that review search filter performance.[14] The ISSG Search Filters Resource categorises filters into study design filters and "other" filters. Geographic search filters are one of the types of filter categorised as "other". Geographic search filters are designed to retrieve research from specific locations, usually individual countries, or groups of countries. There are currently 15 search filters covering 12 different geographic areas or populations listed on the ISSG Search Filters Resource (Africa, OECD Countries, and the USA all have two filters each), with nine of these filters designed for Ovid MEDLINE[14] (see Table 3). However, only one filter (which has versions for Ovid MEDLINE, PubMed, Ovid Embase, and CENTRAL via Cochrane Library, Wiley) aims to cover all low- and middle-income countries (LMICs).[16] The Cochrane Effective Practice and Organisation of Care (EPOC) LMIC geographic search filter has not been validated.[16] However, an evaluation was published in June 2020, that found whilst overall the filter does a good job of including the relevant MeSH and keyword terms, there were some suggestions for improvements, including expanding the regional terms and including terms for nationalities.[17]

1.3 | Scoping the literature on pre-term birth in low- and middle-income countries

In 2018, an information specialist (AS) and a systematic reviewer (FC) at the School of Health and Related Research (ScHARR), University of Sheffield conducted a scoping exercise to inform a research application to Global Challenge Research Fund (UK Research and Innovation) on preterm birth (PTB). Subsequently, this scoping exercise was used to inform a mapping review of interventions delivered in low- and middle-income country settings to prevent spontaneous PTB, commissioned by the National Institute for Health Research (NIHR) funded Global Health Research Group on Preterm birth prevention and management (PRIME). PRIME is a program bringing together a group of interdisciplinary researchers from the United Kingdom, South Africa, and Bangladesh to address the challenges of PTB in low-middle income countries (LMICs), where its prevalence is highest.[34] The mapping review aimed to identify and describe the quantity and quality of systematic reviews that have sought to explore the effectiveness, safety and, acceptability of interventions to prevent PTB. Based on the existing evidence, the review would identify research gaps in LMIC contexts to inform future research and identify areas for potential further research synthesis.

For the mapping review, it was particularly important to identify literature relating to LMICs in this context, as some interventions that have been used in developed world contexts may be harmful in LMIC settings, and conversely there may be interventions that are even more effective in LMICs than in other settings,[35,36] the applicability of the intervention was key in this review. Therefore, we sought to use an LMIC geographic search filter to ensure the results of our search were relevant to the LMIC setting. The Cochrane Effective Practice and Organisational Care Group's LMIC geographic search filter[16] was selected. The filter has several versions available

TABLE 3 Geographic search filters currently available

Geographic locations	Database filters available	Validated?
Africa[9]	Ovid Embase Ovid MEDLINE PubMed	Yes
Africa (a set of filters to cover, Northern, Eastern, Western, Southern, and Middle Africa) – note that some of the filters have been developed for more databases than others[18]	Ovid Embase Ovid MEDLINE EBSCO CINAHL Ovid Global Health Ovid ERIC	No
Canada (a set of filters to cover various provinces) – note that some of the filters have been developed for more databases than others[19]	Ovid Embase Ovid MEDLINE MEDLINE via EBSCO Ovid Global Health Ovid ERIC	No
Canadian Indigenous Peoples[20]	Ovid MEDLINE	No
Eastern Mediterranean Region[21]	PubMed	No
"New" European Union Member States (as in 2010)[22]	PubMed	No
German-speaking Countries[23]	Web of Science	No
Low- and middle-income countries[16]	CENTRAL via The Cochrane Library Ovid Embase Ovid MEDLINE PubMed	No
OECD Countries (NICE)[24]	Ovid MEDLINE Ovid Embase	Yes[25]
OECD Countries (Canadian Health Libraries Association)[26]	Ovid Databases	No
Spain[27]	PubMed	Yes
Sub-Saharan Africa[28]	Ovid Embase Ovid MEDLINE	No
UK[29,30]	Ovid MEDLINE Ovid Embase	Yes[31]
USA (Popoff et al.[32])	Ovid MEDLINE	No (validation in process)
USA (University of Texas, School of Public Health)[33]	Ovid MEDLINE	No

Note: For full details of the filters, visit the ISSG Search Filters Resource.[14]

for different databases: Ovid MEDLINE, PubMed, Ovid Embase, and CENTRAL.

1.4 | Requirement for adapting the Cochrane EPOC LMIC geographic filter

During the scoping search, we found that the Cochrane EPOC LMIC geographic search filter did not retrieve a known study of interest, therefore, we investigated possible adaptations to the filter to ensure this study was retrieved, and potentially further relevant studies.

1.5 | Aims and objectives

This filter development study aimed to develop a new 1st generation (non-validated) geographic search filter based on the Cochrane EPOC LIMIC geographic search filter, with some adaptations to improve retrieval of LMIC studies relating to pre-term birth prevention and management.

2 | METHODS

The search strategy we developed aimed to identify systematic reviews and primary studies about PTB in

TABLE 4 Adding the institution search field (Ovid MEDLINE)

Line of existing filter (2012 version)	Changes highlighted in bold *note that quotation marks were added to "Cote d'Ivoire" as the apostrophe is an unsupported character in Ovid MEDLINE
2.	(Africa or Asia or Caribbean or West Indies or South America or Latin America or Central America).hw,kf,ti,ab,cp,**in**.
3.	(Afghanistan or Angola or Armenia or Armenian or Bangladesh or Benin or Bhutan or Bolivia or Burkina Faso or Burkina Fasso or Burundi or Cambodia or Central African Republic or Chad or Comoros or Congo or "Cote d'Ivoire" or Ivory Coast or Djibouti or Egypt or El Salvador or Eritrea or Ethiopia or Gambia or Gaza or Georgia or Ghana or Guatemala or Guinea or Guam or Haiti or Honduras or India or Indonesia or Kenya or Kiribati or Korea or Kosovo or Kyrgyzstan or Lao PDR or Lesotho or Liberia or Madagascar or Malawi or Mali or Mauritania or Moldova or Mongolia or Morocco or Mozambique or Myanmar or Myanma or Nepal or Nicaragua or Niger or Nigeria or Pakistan or Paraguay or Philippines or Philipines or Phillipines or Phillippines or Rwanda or Ruanda or Sao Tome or Senegal or Sri Lanka or Solomon Islands or Somalia or Sudan or Swaziland or Tajikistan or Tanzania or Timor-Leste or Tokelau or Togo or Tuvalu or Uganda or Ukraine or Uzbekistan or Vanuatu or Vietnam or Viet Nam or West Bank or Yemen or Zambia or Zimbabwe).hw,kf,ti,ab,cp,**in**.
Existing filter (2020 LMIC COUNTRY NAMES AND GENERAL LMIC TERMS Version)	Changes highlighted in bold Recommend adding an additional line to the filter to search for institution as it does not work by adding .in to the existing search fields (ti,ab,sh,kf.)
Note: added .in to the country names and regions only	(afghanistan OR albania OR algeria OR american samoa OR angola OR "antigua and barbuda" OR antigua OR barbuda OR argentina OR armenia OR armenian OR aruba OR azerbaijan OR bahrain OR bangladesh OR barbados OR republic of belarus OR belarus OR byelarus OR belorussia OR byelorussian OR belize OR british honduras OR benin OR dahomey OR bhutan OR bolivia OR "bosnia and herzegovina" OR bosnia OR herzegovina OR botswana OR bechuanaland OR brazil OR brasil OR bulgaria OR burkina faso OR burkina fasso OR upper volta OR burundi OR urundi OR cabo verde OR cape verde OR cambodia OR kampuchea OR khmer republic OR cameroon OR cameron OR cameroun OR central african republic OR ubangi shari OR chad OR chile OR china OR colombia OR comoros OR comoro islands OR iles comores OR mayotte OR democratic republic of the congo OR democratic republic congo OR congo OR zaire OR costa rica OR "cote d'ivoire" OR "cote d' ivoire" OR cote divoire OR cote d ivoire OR ivory coast OR croatia OR cuba OR cyprus OR czech republic OR czechoslovakia OR djibouti OR french somaliland OR dominica OR dominican republic OR ecuador OR egypt OR united arab republic OR el salvador OR equatorial guinea OR spanish guinea OR eritrea OR estonia OR eswatini OR swaziland OR ethiopia OR fiji OR gabon OR gabonese republic OR gambia OR "georgia (republic)" OR georgian OR ghana OR gold coast OR gibraltar OR greece OR grenada OR guam OR guatemala OR guinea OR guinea bissau OR guyana OR british guiana OR haiti OR hispaniola OR honduras OR hungary OR india OR indonesia OR timor OR iran OR iraq OR isle of man OR jamaica OR jordan OR kazakhstan OR kazakh OR kenya OR "democratic people's republic of korea" OR republic of korea OR north korea OR south korea OR korea OR kosovo OR kyrgyzstan OR kirghizia OR kirgizstan OR kyrgyz republic OR kirghiz OR laos OR lao pdr OR "lao people's democratic republic" OR latvia OR lebanon OR lebanese republic OR lesotho OR basutoland OR liberia OR libya OR libyan arab jamahiriya OR lithuania OR macau OR macao OR republic of north macedonia OR macedonia OR madagascar OR malagasy republic OR malawi OR nyasaland OR malaysia OR malay federation OR malaya federation OR maldives OR indian ocean islands OR indian ocean OR mali OR malta OR micronesia OR federated states of micronesia OR kiribati OR marshall islands OR nauru OR northern mariana islands OR palau OR tuvalu OR mauritania OR mauritius OR mexico OR moldova OR moldovian OR mongolia OR montenegro OR morocco OR ifni OR mozambique OR portuguese east africa OR myanmar OR burma OR namibia OR nepal OR netherlands antilles OR nicaragua OR niger OR nigeria OR oman OR muscat OR pakistan OR panama OR papua new guinea OR new guinea OR paraguay OR peru OR philippines OR philipines OR phillipines OR phillippines OR poland OR "polish people's republic" OR portugal OR portuguese republic OR puerto rico OR romania OR russia OR russian federation OR ussr OR soviet union OR union of soviet socialist republics OR rwanda OR ruanda OR samoa OR pacific

(Continues)

TABLE 4 (Continued)

	islands OR polynesia OR samoan islands OR navigator island OR navigator islands OR "sao tome and principe" OR saudi arabia OR senegal OR serbia OR seychelles OR sierra leone OR slovakia OR slovak republic OR slovenia OR melanesia OR solomon island OR solomon islands OR norfolk island OR norfolk islands OR somalia OR south africa OR south sudan OR sri lanka OR ceylon OR "saint kitts and nevis" OR "st. kitts and nevis" OR saint lucia OR "st. lucia" OR "saint vincent and the grenadines" OR saint vincent OR "st. vincent" OR grenadines OR sudan OR suriname OR surinam OR dutch guiana OR netherlands guiana OR syria OR syrian arab republic OR tajikistan OR tadjikistan OR tadzhikistan OR tadzhik OR tanzania OR tanganyika OR thailand OR siam OR timor leste OR east timor OR togo OR togolese republic OR tonga OR "trinidad and tobago" OR trinidad OR tobago OR tunisia OR turkey OR turkmenistan OR turkmen OR uganda OR ukraine OR uruguay OR uzbekistan OR uzbek OR vanuatu OR new hebrides OR venezuela OR vietnam OR viet nam OR middle east OR west bank OR gaza OR palestine OR yemen OR yugoslavia OR zambia OR zimbabwe OR northern rhodesia OR global south OR africa south of the sahara OR sub-saharan africa OR subsaharan africa OR africa, central OR central africa OR africa, northern OR north africa OR northern africa OR magreb OR maghrib OR sahara OR africa, southern OR southern africa OR africa, eastern OR east africa OR eastern africa OR africa, western OR west africa OR western africa OR west indies OR indian ocean islands OR caribbean OR central america OR latin america OR "south and central america" OR south america OR asia, central OR central asia OR asia, northern OR north asia OR northern asia OR asia, southeastern OR southeastern asia OR south eastern asia OR southeast asia OR south east asia OR asia, western OR western asia OR europe, eastern OR east europe OR eastern europe).**in.**

LMICs. The search was limited to English Language and Human studies only, added to MEDLINE since 1st April 2009. The date limit was used due to a previous mapping review being identified, which covered studies pre-April 2009.[37] For the scoping search, the Cochrane EPOC LMIC geographic search filter for Ovid MEDLINE was used.[16] The searches were conducted in July 2018, and the version of the Cochrane EPOC LMIC Filter at that time dated from 2012.[38] The filter has since been updated in 2020,[16] however neither versions of the filter have been validated. The main changes to the 2020 version of the filter relate to the list of countries, updated by the World Bank in 2019,[39] and including subject heading as a search field. There is also now a LMIC Demonyms version of the filter,[16] but our focus is on the LMIC Countries and General LMIC terms version, as this is the equivalent of the 2012 filter.[38]

The results of the Ovid MEDLINE scoping search were screened by the systematic reviewer (FC). Following the screening of the scoping search results, a relevant study was identified, which had not been retrieved by the search.[40] The study was looked up on Ovid MEDLINE to check it was indexed on the database and the record was examined for potential additions to the search strategy (search terms or syntax) to ensure that the Bhutta et al.[40] study was retrieved, along with any other potentially missed studies.

Following the examination of all the search fields of the Bhutta et al.[40] study, including the MeSH headings, two edits were made to the LMIC geographic search filter[16] as described below and depicted in Tables 4 and 5 for the Ovid MEDLINE version. We present the changes we made to the 2012 filter,[38] but also present the same changes as they would apply to the updated filter.[16]

2.1 | Named countries: searching in the institution field (in)

Despite the Bhutta et al.[40] study covering LMICs, its record on MEDLINE contained no MeSH headings relating to LMICs, neither the generic "Developing Countries" MeSH heading nor any specifically named countries. India is mentioned, but in the address (institution) of one of the authors. In the Cochrane EPOC LMIC geographic search filter used, the countries are searched for in title, abstract, country of publication, headings, or author keywords, so we added institution (in) for our version of the filter (see Table 4).

2.2 | Generic LIMC terms: high-burden or countdown countries

The Bhutta et al.[40] study did not include any of the generic terms for LMICs included in the Cochrane EPOC LMIC geographic search filter.[38] However, it did mention "high burden Countdown countries". The concept of

TABLE 5 Adding high-burden/countdown countries as a free-text search (Ovid MEDLINE)

Add this line at the end of the filter combining with OR (2012 & 2020 versions)	((high burden or high-burden or countdown) adj countr*).ti,ab.

high burden countries is most commonly used in the context of tuberculosis,[41] but it was used as a more general term in Bhutta et al.[40] It is stated in the abstract that they *"modelled the effect and cost of scale-up in the 75 high-burden Countdown countries"*.[40] Countdown countries relates to an initiative in the use of data to foster accountability for women's and children's health.[42–44]

To retrieve the Bhutta et al.[40] study, a free-text title or abstract search for high burden or countdown countries was added to the LIMC geographic search filter (see Table 5).[16,38]

The adapted search filter was developed in Ovid MEDLINE, and then translated for use in Ovid Embase, Cochrane Library, Ovid PsycINFO, and CINAHL via EBSCO.

3 | RESULTS

The ScHARR LMIC filter we developed is a non-validated 1st generation geographic search filter adapted from the Cochrane EPOC LMIC filter. We adapted the ScHARR LMIC geographic search filter to be run in Ovid Embase, Cochrane Library, Ovid PsycINFO, and CINAHL via EBSCO. For PyscINFO and CINAHL, there was not an existing filter, so these were developed from scratch, based on the Ovid MEDLINE filter. Full details of our adapted geographic search filter (ScHARR LMIC) for each database can be found in Appendix S1.

We developed the filter during scoping searches to inform a mapping review of interventions delivered in low- and middle-income country settings to prevent spontaneous PTB. The ScHARR LMIC filter is more sensitive than the Cochrane EPOC LMIC filter for this particular topic. The scoping searches (conducted on Ovid MEDLINE) were complex and went through several iterations, trialling combinations of terms, date limits and with or without a geographical filter applied to retrieve reviews about LMICs, or primary studies conducted in LMIC countries. In addition, the adaptation of the Cochrane EPOC LMIC geographic search filter was not designed as methods work, it was a pragmatic decision made during the scoping of the literature to inform a future project, which was a time-limited exercise due to internal deadlines. As such the detail on search results from the scoping search and the impact of adapting the Cochrane EPOC LMIC geographic search filter is limited. An initial scoping search to retrieve reviews found 251 references, with 131 of these being conducted from 2009 onwards. Following the inclusion of more terms related to pre-term birth and adapting the Cochrane EPOC LMIC geographic search filter, the number of increased to 1465, with 842 being published since 2009. Approximately 400 of the total increase were due to the adapted geographic search filter. The final iteration of the scoping search retrieved the Bhutta et al.[40] study, which went on to become an included study for the mapping review.[35,36] For full copies of the adapted geographic search filters used, see Appendix S1.

Following the scoping searches, it was decided that the search for systematic reviews for the mapping review would not be limited to setting due to the paucity of evidence, therefore the LMIC filter was removed from this part of the search. Due to this, we did not map the included studies onto the MEDLINE scoping search as it was a reference set with different search parameters. As such we do not have an exact calculation of the "number needed to read"[5] versus the yield. The scoping searches were done in full consultation with the reviewer, who was satisfied with the increase in search result volume to retrieve the Bhutta et al[40] study. The ScHARR LMIC geographic search filter was used for the primary study searches in the mapping review.

4 | DISCUSSION

The ScHARR LMIC filter is an adapted version of the Cochrane EPOC LMIC geographic search filter. From our brief investigation, we found that studies about LMICs may not be indexed accordingly or have no information in the abstract about the geographical setting. Comparing our findings with other geographic search filters, only one makes use of the institution field, which is the National Institute for Health and Care Excellence (NICE) UK filter developed for both Ovid MEDLINE and Embase.[29,30] The NICE UK filter had not been published at the time of our investigation, but our study findings that including institution as a search field is beneficial matches the findings from these more recent studies.

If using our adapted filter, information specialists and researchers should note that the ScHARR LMIC geographic search filter has only been used in scoping searches and the subsequent PRIME mapping review[35,36] so far. Beyond our initial development, it has not been tested for sensitivity and precision, nor undergone any validation. Future developments for the filter would

benefit from such testing and validation, similar to that conducted by the developers of other geographic search filters.[9,24,25,27,29–31]

Future research on the use and efficiency of geographic search filters would be of benefit to information specialists and the evidence synthesis community. Ayiku et al.[45] provide a useful guide to developing and validating geographic search filters and encourage more to be developed. There are perhaps enough geographic search filters now to warrant a review, similar to those about methodological and topic search filters.[3,46] Damarell et al.[46] included three geographic search filters in their review of topic search filters, but their searches were conducted in 2016–2017, therefore there are new filters published since this review. When conducting reviews of search filters, it is important to include grey literature searching as findings may first be presented at conferences prior to publication, or never be formally published as a journal article. It is also recommended to consult with experts (e.g., information specialists) via existing networks, such as mailing lists, social media, and personal contacts of colleagues and peers. Damarell et al.[46] found 28 studies to screen from their supplementary search methods.

4.1 | Limitations

The ScHARR LMIC geographic search filter has several limitations. First, it has not been validated. Second, it was designed for finding evidence to identify studies on pre-term birth, so it's applicability for other topics is unknown. Final, we know that our filter identified one further relevant study, however we do not know, whether it retrieved further relevant studies as this investigation was beyond the resources that were allocated to the project. Using the ScHARR LMIC geographic search filter increased the search result volume, but we do not know if this increase in screening burden was worth the effort to retrieve potentially only one additional relevant study, that was already known to the research team.

5 | CONCLUSION

When searching for literature from LMICs, there is currently limited choice regarding search filters to use. We recommend using the ScHARR LMIC filter if you wish to improve sensitivity and reduce the risk of missing relevant studies, when searching for literature about LMICs. Versions of the ScHARR LMIC filter exist for Ovid MEDLINE, Ovid Embase, Ovid PsycINFO, Cochrane Library, and CINAHL via EBSCO. The ScHARR LMIC filter has not yet been validated therefore any recommendations are based on this initial case study. Future research on validating our adaptations and the Cochrane EPOC LMIC geographic search filter in general would be beneficial.

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CONFLICT OF INTEREST

The authors declare no conflicts of interest.

AUTHOR CONTRIBUTIONS

Anthea Sutton carried out the searches and adapted the search filter, in discussion with Fiona Campbell. Anthea Sutton drafted the manuscript with contributions from Fiona Campbell. Anthea Sutton and Fiona Campbell made revisions, with final approval of the manuscript from both authors.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

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SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

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Review Paper

A scoping review of the experience of implementing population testing for SARS-CoV-2

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ABSTRACT

Objectives: The severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) – also known as the coronavirus disease (COVID-19) – pandemic has led to the swift introduction of population testing programmes in many countries across the world, using testing modalities such as drive-through, walk-through, mobile and home visiting programmes. Here, we provide an overview of the literature describing the experience of implementing population testing for severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2).

Study design: Scoping review.

Methods: We conducted a scoping review using Embase, Medline and the Cochrane library in addition to a grey literature search. We identified indicators relevant to process, quality and resource outcomes related to each testing modality.

Results: In total, 2999 titles were identified from the academic literature and the grey literature search, of which 22 were relevant. Most studies were from the USA and the Republic of Korea. Drive-through testing centres were the most common testing modality evaluated and these provided a rapid method of testing whilst minimising resource use.

Conclusions: The evidence base for population testing lacks high quality studies, however, the literature provides evaluations of the advantages and limitations of different testing modalities. There is a need for robust evidence in this area to ensure that testing is deployed in a safe and effective manner in response to the COVID-19 pandemic.

Introduction

In response to the severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) pandemic, many countries implemented population testing programmes as part of countermeasures to contain the spread of infection and mitigate its health and economic impacts. Population testing provides disease surveillance required to inform broader policy decisions, target resource utilisation and, when twinned with timely case isolation and contact tracing, more effective containment of the virus.[1] Worldwide, population testing programmes are diverse, depending on the population eligible for testing, the technology used to sample and analyse specimens, as well as the timing and frequency of testing. In this article, we have defined population testing as any testing programme which uses an antigen or antibody test to identify coronavirus disease 2019 (COVID-19) in a group of symptomatic and/or asymptomatic individuals. In the UK, there are two distinct testing programmes. The UK National Health Service (NHS) Test and Trace system [NHSTT] tests self-reported, symptomatic individuals using quantitative polymerase chain reaction (Q-PCR) assays. More recently, mass testing of asymptomatic individuals using cheaper, faster lateral flow devices has also been introduced.

Population testing seeks to identify people infected with SARS-CoV-2 in a predefined group such as healthcare workers. By identifying cases of infection through testing, action can be taken to limit infection spread by isolating infected individuals and their contacts during their infectious period. As SARS-CoV-2 may be spread by asymptomatic individuals, including these individuals in testing programmes could help reduce viral transmission. Cumulatively, these actions help control the spread of infection and create conditions that would enable the relatively normal functioning of society.

Modalities used for COVID-19 population testing include drive-in, walk-in, mobile sites, postal testing and home visits. The UK, for example, adopted a testing strategy with five pillars, each pillar pertaining to a population subgroup and focussing on either

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diagnosis, detecting past infection or for surveillance purposes to estimate population prevalence. Testing is co-ordinated centrally and delivered from satellite centres and via postal testing.[2] South Korea adopted walk-in and drive-through testing,[3,4] whilst some areas in Scotland have instituted home-testing to reach more vulnerable groups who cannot access test facilities easily.[5,6] There is also increasing political and societal concern of the socio-economic impact of blunt strategies such as national 'lockdowns' to combat COVID-19,[7] and their effect on health inequalities. There may therefore be value in studying the different approaches used worldwide and to learn from successful programmes from other countries.

What is currently unclear is whether any modality of population testing is more robust and efficacious for containing the virus. There is a need to identify population testing programmes that are more accessible and effective at containing the spread of infection, together with the determinants of success. This can help inform national testing policies as part of the pandemic response efforts to minimise the health, social and economic harms. We conducted a scoping review to describe the volume and type of evidence reporting on the experience of implementing population testing for SARS-CoV-2 in high and upper-middle income countries during the pandemic.

Methods

Scoping review methodology

Scoping reviews aim to rapidly map the key concepts underpinning a research area, by comprehensively summarising evidence to inform practice and policy and provide direction for future research.[8,9] Scoping reviews use rigorous and transparent literature searching methods but differ from systematic reviews as the quality of included studies are not routinely assessed, nor do they provide a synthesised answer to a particular research question.[10]

This scoping review followed the framework proposed by Arksey and O'Malley[8] and refined by Levac et al.[11] briefly comprising: identification of the research question, identification of studies, selection of studies, charting of data and collation of results. This review was commissioned by Public Health England who were consulted on the interim outputs of the study. The trial protocol was published on PROSPERO, number CRD42020186506.

Identification of the research question

The purpose of this review was to assess the volume of published literature describing the experience of implementing population testing for COVID-19 and to identify the nature and characteristics of the testing programmes. We sought to elucidate what data were available to assess the outcomes of these testing programmes in terms of processes, participants, quality and/or resource-use. We developed these broad aims in order to generate breadth of coverage and map the literature on this topic so that key concepts and gaps could be identified to inform further practice and policy.

Table 1 outlines the inclusion and exclusion criteria for this review. Studies were included if they described the process of providing or accessing a population testing point for symptomatic and/or asymptomatic individuals for COVID-19, using an antigen or antibody test in any setting, using any testing modality. In order to prioritise research relevant to high-income countries such as the UK, we included literature relating to comparable health services from high and upper-middle income countries only.

Studies of laboratory aspects of testing (including diagnostic accuracy), commentaries, opinion pieces and modelling studies were excluded. Studies that described screening where samples were not taken, or that described the testing of passengers at ports or borders, were also considered out of scope and excluded.

Literature identification and selection

A search strategy (see Appendix 1) was developed to retrieve studies that had evaluated or described the process of providing or accessing a testing point for population testing for COVID-19. An information specialist (AC) searched the electronic databases Medline, Embase and the Cochrane Library (Fig. 1). Searches were originally conducted in May 2020 and updated in August 2020. The search was limited to studies in English and published between January and August 2020.

Extracted titles and abstracts were screened by at least two reviewers (CF, FC, LB). A total of 250 full text articles were reviewed to clarify whether the article met the inclusion criteria given in Table 1, either because no abstract was available or because it was unclear from the title and abstract alone whether the study met the inclusion criteria. Abstracts were often unavailable due to the large number of commentaries and opinion pieces found by the search strategy. A formal quality appraisal of the evidence was not conducted, given the remit of the scoping review.

In addition, a grey literature search for national and international clinical guidelines was conducted during May 2020. The World Health Organization (WHO) and the European Centre for Disease Prevention and Control (ECDC) websites were searched, plus websites in the English language from the UK, USA, Canada, Australia, New Zealand, South Korea, China and Taiwan. This search identified 58 potentially relevant guidelines. On further review, only one guideline was relevant to population testing and detailed an approach to drive-through screening implemented in South Korea.[12] A further 21 guidelines looked at wider aspects of screening.

Charting of data and collation of results

After the screening was completed, relevant content in the included studies was extracted into a spreadsheet. Data extraction was verified by a second reviewer who checked data extraction from a random sample of four articles. The mode of testing was

Table 1
Inclusion and exclusion criteria.

Inclusion criteria	Exclusion criteria
● Any study evaluating or describing the process of providing or accessing a population testing point for COVID-19 ● Symptomatic and/or asymptomatic individuals ● Any setting ● Antibody or antigen testing ● Any testing modality (for example drive through testing or home visiting testing) ● High or upper-middle income countries according to World Bank criteria	● Commentaries, modelling studies or opinion pieces ● Studies describing only laboratory aspects of testing, including studies of diagnostic accuracy. ● Studies describing only testing at ports or borders ● Studies describing only screening in which samples were not taken ● Not written in English

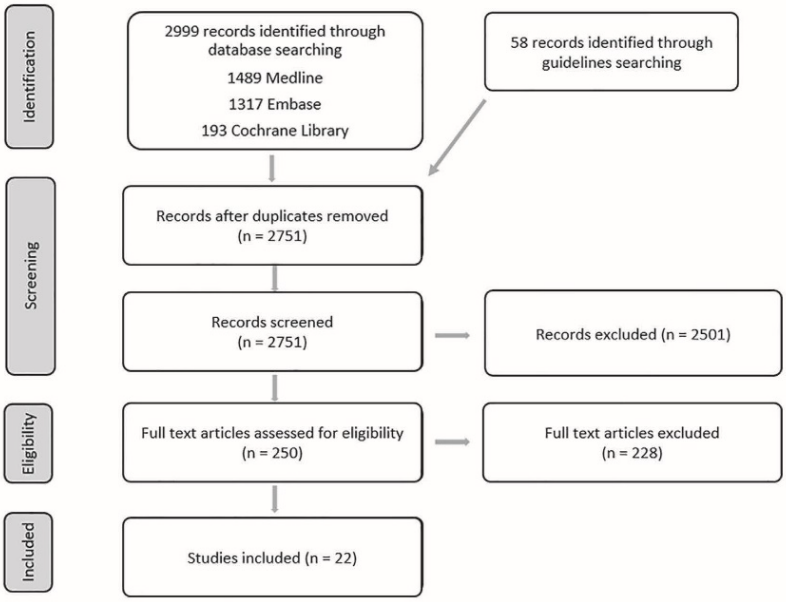

Fig. 1. PRISMA flow diagram.

categorised into one of five different types: drive-through testing, home visiting testing, indoor walk-through centre, outdoor walk-through centre and mobile testing. For outcome data, a thematic framework was used which categorised any quantitative outcomes indicators into one of four groups: process outcomes, participant outcomes, quality outcomes and resource use outcomes.

Results

The database search returned 2999 results (Fig. 1). After automated and manual deduplication, 2751 unique references were screened for relevance to the question. On first screening of titles, 250 references were identified as potentially relevant, and on further reading 22 were categorised as relevant.

Study characteristics

A summary of the studies' characteristics is presented in Table 2.

Most of the published literature on this topic is difficult to assign to a study type. Studies were often referred to as brief reports or short communications. The articles typically comprise a description of the testing modality of interest, often with a diagram of the layout of the testing centre and useful operational details followed by an evaluation of its advantages and disadvantages. In some articles, a comparison group was described; however, there were no published randomised controlled trials (RCTs). One article was a qualitative interview study of early experiences of drive-through testing centres.[13]

All the eligible studies described population testing programmes where samples were taken from symptomatic and occasionally asymptomatic individuals. There were no eligibility studies of mass testing, defined as regular and/or large-scale testing of individuals from defined populations regardless of symptom status using lateral flow tests.

Many articles describing testing programmes were from the USA (43% of studies) and the Republic of Korea (29% of studies). Most articles described testing programmes in high income countries (95% of articles), with only one originating from an upper-middle income country (Malaysia).[15]

Testing modalities

Several different testing modalities were described, which were categorised into five main categories: drive-through, home visiting, mobile testing, indoor walk-through centres and outdoor walk-through centres.

Sometimes described as off-site COVID-19 testing centres (OSCTCs),[13] drive-through testing centres were by far the most common testing modality evaluated (in 72% of articles), in Israel, Malaysia, Korea, Scotland and the USA. This testing modality enabled the use of a vehicle as a self-contained unit, which can reduce the spread of infection. Most were in car parks, stadiums and parks, and one was in an open-air area of a hospital.[26] Some centres enabled individuals without a car to walk in for testing[13] in order to increase accessibility. Six articles described drive-through testing in combination with either home visiting testing, mobile testing and/or walk-through testing, enabling a greater proportion of the population to access testing.

Home visiting testing (18% of articles) typically involved a small number of healthcare workers visiting the home of an individual to perform a test. This enabled individuals who are home-bound, frail or have no means of private transportation to access testing without having to use an ambulance, visit a hospital or rely on the assistance of others to access a drive-through site. These schemes were often used as an alternative to local drive-through testing facilities. Home visiting testing took place in Israel, Scotland and the USA.

Two articles described mobile testing, in which testing staff visited populations rather than expecting the participants to travel to a testing centre. In Korea, testing staff visited workers onsite at their workplaces[28] and in Florida, mobile polymerase chain reaction (PCR) testing laboratories were used to provide point-of-care testing in different cities.[27]

Table 2
Characteristics of included articles.

First Author	Country	Mode of Testing
Appa[14]	USA	Drive-through
Brammer[13]	USA	Drive-through
Chang[16]	Republic of Korea	Drive-through and walk-through
Drees[17]	USA	Drive-through
Flynn[18]	USA	Drive-through and walk-through
Goldberg[19]	USA	Home visiting testing
Halalau[20]	USA	Drive-through
Hill[9]	Scotland	Drive-through and home visiting testing
Kim[21]	Israel	Drive-through and home visiting testing
Kim[22]	Republic of Korea	Indoor walk-through centre
Kwon[4]	Republic of Korea	Drive-through
Kwon[23]	Republic of Korea	Outdoor walk-through centre
Lee[15]	Malaysia	Drive-through
Lin[24]	Taiwan	Outdoor walk-through centre
Lindholm[25]	USA	Drive-through
Manauis[26]	Singapore	Indoor walk-through centre
Ministry of Foreign Affairs[12]	Republic of Korea	Drive-through
Mark[8]	Scotland	Drive-through and home visiting testing
Rivkees[27]	USA	Drive-through, walk-through and mobile testing
Seo[28]	Republic of Korea	Mobile testing
Shah[29]	USA	Drive-through
Ton[30]	USA	Drive-through

Indoor walk-through centres based in healthcare facilities were used in Singapore and Korea. There were several different designs for walk-through centres, which were located inside hospitals or other healthcare facilities: screening centres,[22,26] negative pressure booths[22] and negative pressure tents.[16] Screening centres permit individuals to access testing inside a building. The Singapore Screening Centre was designed to minimise the movement of patients around the building.[26] Patients were assigned a seat number and tagged with a tracker to facilitate contact tracing; staff visited patients in their seats to further reduce contact amongst patients.

Negative pressure booths and tents have been designed to minimise the opportunity for viral spread in an indoor setting. Negative pressure booths[22] were used for sample collection and medical examination procedures in Korea. The booths were inspired by the design of biosafety cabinets and contain a 'glove wall' separating the patient and the medical staff member, who communicate using an interphone. Patients complete registration, questionnaires and payment outside the booths in other sections of the screening centre. Negative pressure booth systems aimed to protect healthcare staff, reduce personal protective equipment (PPE) use and increase throughput compared to other walk-through systems. Negative pressure tents,[16] also located in Korea, were similar to negative pressure booths, but the whole tent is under negative pressure. Staff working in the tent wore full personal protective equipment (PPE) and most of the tent required sterilisation between people tested that took at least 30 min.

The final testing modality described was outdoor walk-through centres. These were located outside the hospitals in Korea and Taiwan.[23,24] The Korean clinic[23] screened all patients and visitors to the hospital with the aim of minimising ward closures due to COVID-19 outbreaks. In Taiwan, a 'multifunctional sampling station' was built outside an emergency department, using a 2-cm thick clear acrylic board to separate emergency department patients and medical personnel, with inbuilt gloves used to conduct sampling.[24]

Populations tested

Table 3 summarises details of the populations tested and the types of test used. Fifty-five percent of studies provided information regarding the population that was eligible to be tested. Forty-one percent of articles described accepting both symptomatic and asymptomatic individuals, and 14% accepted those with symptoms only. Some testing centres had an algorithm for testing eligibility involving symptoms, epidemiological links, occupational risk factors and/or potential for community exposure. For some, the testing criteria changed over time as the pandemic progressed.[20,29]

Types of testing used

The most common method for sampling was through nasal or throat swabs. 45% of articles described the use of nasal swabbing, involving either nasal, nasopharyngeal or mid-turbinate sampling. 27% described the use of throat/oropharyngeal swabbing. 27% stated that swabbing was used but did not specify whether these were nasal and/or throat swabs. Less commonly described testing procedures included sputum sampling (14% of articles) and blood sampling for antibody testing (14% of articles) and one included the use of temperature measurements. 9% of publications described the use of prescreening questionnaires before sampling took place. 18% of articles did not specify the method of testing and 41% of articles described the use of a combination of the above methods.

Outcomes of interest

Outcomes of interest were divided into three categories: process outcomes, quality outcomes and resource use outcomes. No participant outcomes were measured quantitatively in any of the included studies; however, the discussion section of many articles contained rich qualitative data describing participant outcomes such as staff and participant safety and well-being and service equity.

Process outcomes

The process outcomes described comprise throughput, duration of test, decontamination time, time to don/doff PPE and waiting time.

Seventy-seven percent of the articles reported the number of people tested in a specified time period (Table 4). These figures were used to calculate the mean number of people tested each day.[a] Although the different studies are not directly comparable due to

[a] *2 studies reported the time period per 'week' but did not state which days of the week were available for testing; a 7-day week was assumed for calculations.

Table 3
Populations eligible to be tested and types of testing used.

First Author	Population tested	Type of test
Appa[14]	Symptomatic and asymptomatic	Oropharyngeal/midturbinate swab for PCR testing, finger prick blood test for antibody testing
Brammer[13]	N/A	N/A
Chang[16]	Symptomatic and asymptomatic	RT-PCR tests of 'specimens'
Drees[17]	Symptomatic only	Not specified
Flynn[18]	Unclear/unspecified	'In-house COVID-19 test' of nasopharyngeal swabs
Goldberg[19]	Symptomatic and/or epidemiological link and/or risk factors and/or potential for community exposure	Nasopharyngeal swab collection for RT-PCR analysis
Halalau[20]	Unclear/unspecified	Nasopharyngeal swab collection for RT-PCR analysis
Hill[5]	Unclear/unspecified	Combined nose and throat swab
Kim[21]	Symptomatic only	Swab testing
Kim[22]	Symptomatic only	Not specified
Kwon[4]	Symptomatic and asymptomatic	Naso- and oropharyngeal swabs, sputum specimen
Kwon[23]	Symptomatic and/or epidemiological link and/or risk factors and/or potential for community exposure	Pretesting questionnaire
Lee[15]	Symptomatic and asymptomatic	Temperature measurements, nasopharyngeal and oral swabs
Lin[24]	Unclear/unspecified	Nasopharyngeal and oral swab, sputum collection, blood testing for antibodies
Lindholm[25]	Symptomatic and/or epidemiological link and/or risk factors and/or potential for community exposure	Screening questionnaire followed by nasopharyngeal swab collection for RT-PCR analysis for those with symptoms or epidemiological link
Manauis[26]	Symptomatic and/or epidemiological link and/or risk factors and/or potential for community exposure	Swab testing
Mark[6]	Unclear/unspecified	Not specified
Ministry of Foreign Affairs[12]	Unclear/unspecified	Upper respiratory tract sample (all), lower respiratory tract sample (only if participant can expectorate sputum alone into a container)
Rivkees[27]	Symptomatic and asymptomatic	PCR and antibody testing
Seo[28]	Unclear/unspecified	Questionnaire followed by sample collection for RT-PCR testing
Shah[29]	Symptomatic and/or epidemiological link and/or risk factors and/or potential for community exposure	Originally naso- and oropharyngeal swab, later solely nasopharyngeal swab for reverse transcription (RT)-PCR testing (following Centre for Disease Prevention and Control [CDC] advice)
Ton[30]	Unclear/unspecified	Nasal swab

factors such as the different sizes of populations served and staff employed, this section gives a broad overview of the types of throughput that may be experienced for different testing modalities.

Drive-through testing centres tested between 22 and 539 individuals per day. Indoor walk-through centres tested 9–500 people per day. One outdoor walk-through centre tested 300 people per day. Home visiting testing teams tested 6–15 people per day.

Some studies compared the throughput of different testing modalities (Table 5). Three studies compared drive-through testing with walk-through or home visiting testing.[5,6,16] For similar settings, a higher throughput of individuals could be achieved in a drive-through setting compared to walk-through testing[16] or home visiting testing.[5,6] For the same time period, one indoor walk-through screening centre using negative pressure booths tested more patients than a walk-through centre with no negative pressure booths (>70 people per day compared to 9–10 people per day).[22] Multiple booths could be installed and decontamination time between individuals could be reduced to 3–5 min from over 30 min. It is difficult to compare different studies, as several variables other than the testing modality can affect the number of people tested per day, such as the number of staff present, the procedures used and the number of individuals who could be tested concurrently.

The mean duration of a drive-through test was between 3 and 15 min. One study reported a median time per test of 28 min (interquartile range [IQR] 17–44 min).[20] Some centres allowed multiple people to be tested per vehicle, whereas others allowed only one person per vehicle. The layout of drive-through testing sites can allow several individuals to be tested at one time, for example one drive- and walk-through centre could test two patients every 5 min.[18]

Twenty-seven percent of articles compared testing modalities, calculating a range of indicators for process outcomes (Table 5). Drive-through testing was found to be faster than walk-through testing using a negative pressure tent[16] or a screening centre.[4] Testing using an outdoor walk-through centre (2 min per test) was faster than using traditional sample collection in a single negative-pressure isolation room (5 min per test).[24] Home visiting testing (30 min per test) was also quicker than transporting patients to hospital for tests with a specialist ambulance (<1 h).[6] However, waiting times, defined as the time between arrival at the drive-through centre and testing, was reported to be as high as 7 h at peak volume.[20]

Testing in an outdoor walk-through centre dramatically reduced time to don/doff PPE compared to traditional sample collection in a single negative-pressure isolation room (1 min per patient compared to 21 min per patient).[24]

Several studies measured the time to disinfect equipment between individuals (decontamination time). The use of drive-through testing eliminated the need for a 30 min decontamination time between patients in a walk-through centre using a negative pressure tent.[16] Drive-through and home visiting testing also required no decontamination time, compared to up to 6 h of decontamination time needed if a patient was tested in the emergency department.[6] Decontamination time was much shorter when using an outdoor walk-through centre (10 min per patient) compared to a single negative-pressure isolation room (35 min per patient). The use of negative pressure booths reduced decontamination time from >30 min with no negative pressure booths to 3–5 min between patients.[31]

Quality outcomes

Two different indicators were used to describe quality outcomes: the median time from referral to test and the test

Table 4
Number of people tested per day.

Number tested per day (calculated)	Prescreening (Questionnaire/Temperature only)	Testing (Samples taken)	Ref
Drive-through		22*	6
	107	45	20
		60	12
		192	30
		>100	4
	163	122	25
		200 (max 400)	29
		242	16
		460	14
		539	17
Drive-through and home visiting		2000 country-wide	21
Drive-through and Walk-through		65 (range 11–127)	18
Indoor Walk-through centres		9–10 (no negative pressure booths)	22
		>70 (with negative pressure booths)	
		41	16
		50–500	26
Home visiting testing		6 (max 11)*	6
		15	5
		15	19
Outdoor Walk-through centre		300	23

turnaround time. Only one study of home visiting testing calculated the median time from referral to test, which was 1 day with a maximum of 3 days.[6] One drive-through testing study calculated the test turnaround time, defined as the time between testing and communication of results. This was found to be 25 h (IQR 21–29) in-house and 221 h (IQR 161–269) if outsourced.[25]

Resource use outcomes

Resource use outcomes were measured using cost per patient, use of PPE and impact on hospital closure. Home visiting testing reportedly costed much less (£55 per patient) than the use of a specialist ambulance and hospital sampling (£768 per patient)[6] (Table 6).

Another study reported that staff in an outdoor walk-through centre used fewer items of PPE than staff working in negative-pressure isolation rooms.[24] Similarly, drive-through testing can reduce PPE use (96% reduction in mask use, 97% reduction in gown use and 47% reduction in glove use) compared to emergency department based testing.[30]

One study reported the effect of a screening and testing clinic on maintaining the functioning of a tertiary hospital.[23] Before the clinic was opened, an average of 36 beds per day were closed due to COVID-19 patients entering the hospital, whereas after the clinic was open and operating well, there was only one closure event (25 beds).

Discussion

This scoping review provides an overview of the literature describing the experience of implementing population testing for SARS-CoV-2 in high and upper-middle income countries. Whilst a range of modalities were reported, the most commonly evaluated were drive-through services using naso- and/oropharyngeal swabbing. Drive-through testing provided a rapid and scalable method of testing for COVID-19, reducing the risk of exposure to staff and patients within healthcare settings and minimising PPE use. However, this approach raises questions regarding equity of access for those

Table 5
Differences in process outcomes when comparing different testing modalities.

Ref	Testing Modality	PROCESS OUTCOMES			
		Throughput (mean number tested per day)	Duration of test (mean time per test)	Decontamination time (time to disinfect room between individuals)	Time to don/doff PPE
Chang[16]	Drive-through	242	5–7 min	N/A	
	Walk-through (negative pressure tent)	41	30 min	At least 30 min between patients	
Hill[5]	Drive-through	92 patients in 18 h			
	Home visiting testing	15 patients per three staff members per day			
Kwon[4]	Drive-through		10 min		
	Screening centre		30 min		
Lin[24]	Outdoor walk-through centre (multifunctional sample collection station)		2 min	10 min per patient	1 min
	Traditional sample collection (single negative-pressure isolation room)		5 min	35 min per patient	21 min
Mark[6]	Home visiting testing	79 in 2 weeks	30 min + <1 h travel time	0	
	Drive-through	313 in 2 weeks		0	
	Specialist ambulance and hospital sampling	Not specified	<1 h + overnight stay may be required	<6 h	
Kim[22]	Indoor walk-through screening (negative pressure booths)	>70 per day		3–5 min	
	Indoor walk-through screening (no negative pressure booths)	9–10 per day		>30 min	

Table 6
Resource use outcomes.

Ref	Testing modality	Resource use outcomes	
		Cost per patient	PPE use
Lin[24]	Outdoor walk-through centre (multifunctional sample collection station)		0 PPE items used*
	Traditional sample collection (single negative-pressure isolation room)		24 PPE items used
Mark[6]	Home visiting testing	£55	
	Drive-through	Not specified	
	Specialist ambulance and hospital sampling	£768	
Ton[30]	Drive-through		1152 masks, 960 gowns/pairs of gloves for 192 patients
	Emergency department testing		42 masks, 24 gowns, 504 pairs of gloves for 192 patients

who do not have access to a private vehicle or are not well enough to drive. The addition of other testing options such as home visiting, mobile testing or walk-through services may help address this issue. Recently, home-based diagnostic and screening tests for SARS-CoV-2 have become much more widely available, which may reduce the need for large scale testing facilities in the future.

However, the evidence base for population testing lacks robust studies and the heterogeneous nature of the testing programmes described in the literature makes it difficult to compare between studies. Many were simply an evaluation of a testing programme with a discussion of its advantages and limitations rather than robust research studies with control groups. Prospective randomised controlled trials (RCTs) of testing centres would give higher quality data, however the researchers would need to overcome challenges such as adversely impacting the expediency and evolution of site practices in real time, and keeping pace with the rapid development of testing methods in response to the pandemic. Conversely, the studies described in this review are rich in qualitative data which could be synthesised to produce valuable insights into the lessons that have been learned in a variety of different settings. Such a review may be a better use of public health resources to identify translatable and implementable best practices.

There is a paucity of published literature on the implementation of mass testing for SARS-CoV-2, defined as regular and/or large-scale testing of individuals from defined populations regardless of symptom status, using lateral flow tests. There were no studies on mass testing that fit the inclusion criteria of this review, although a few articles have described mass testing of residents in facilities such as care homes and prisons.[32,33] A recent review by the European Centre for Disease Prevention and Control (ECDC) highlights the need for further studies on the assessment and impact of mass testing.[34] Indeed, some countries have rushed to adopt mass testing before the benefits, risks and costs of this approach is fully understood.[35] It is therefore pertinent to draw on the international literature on population testing to inform decision making in order to ensure that testing is deployed in a safe and effective way as part of the overall COVID response.[36] It is imperative that future studies assess the cost-effectiveness, specificity and sensitivity of home-based testing, in conjunction with assessing possible scenarios for ending or reducing access to home-testing in the future.

A distinction needs to be made between population testing and screening for COVID-19. As COVID-19 is a new disease, it is unsurprising that definitions for screening and testing in this context have not yet been standardised and often appear to be arbitrary. We found the term 'screening' is loosely used in different ways in the literature, including the testing of symptomatic and/or asymptomatic individuals, assessment of risk factors via a questionnaire, and temperature measurement of individuals travelling past a screening post.[37 39]

Our literature search aimed to be comprehensive but timely and expedient. Whilst we used rigorous and transparent search methods, we had to limit our search to articles published in English. Therefore it is possible that some relevant studies have not been included. Although we did not formally assess the quality of included articles, much of the literature was not robust as stated earlier. Any preference for one testing modality over another cannot be extrapolated from the data collected in this review, as there is considerable heterogeneity between studies and outcomes will reflect factors such as the local population, geography and site protocols.

Further exploration is needed of population testing using different SARS-CoV-2 tests as the strengths and limitations of the various SARS-CoV-2 tests could influence the yield, cost-effectiveness and viability of the population testing programmes. Additionally, research into the wider consequences of testing programmes is needed especially on population behaviours as a result of testing. Finally, further study of the cost-effectiveness of population testing compared to other pandemic control measures is also required. As with most public health interventions, there is no 'one-size-fits-all' approach, and whatever population testing approach is adopted it will need to be tailored to the local context and target population.

Author statements

Ethical approval

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Competing interests

AL is the co-editor for the journal, but had no involvement in the peer review and editorial decision for this article.

Appendix A. Supplementary data

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EVIDENCE AND GAP MAP

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Non-familial intergenerational interventions and their impact on social and mental wellbeing of both younger and older people—A mapping review and evidence and gap map

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Abstract

Background: Opportunities for social connection between generations in the UK have diminished over the last few decades because of changes in the way that we live and work. The decline in communal spaces such as libraries, youth clubs and community centres mean that there are fewer opportunities to meet and mix socially with other generations outside our own families. Increased working hours, improved technology, changes in family patterns, relationship breakdowns within families and migration are also believed to be contributory factors to generation segregation. There are many potential economic, social and political impacts of generations living separate and parallel lives, for example, higher health and social care costs, an undermining of trust between generations reduced social capital, a reliance on the media to form understanding of others' viewpoints and higher levels of anxiety and loneliness. Intergenerational programmes and activities can take many forms and are delivered in many settings. Evidence suggests that intergenerational activity can have a positive impact on participants, for example, in reducing loneliness and exclusion for both older people and children and young people, improving mental health, increasing mutual understanding and addressing important issues such as ageism, housing and care. There are currently no other EGMs that exist that address this type of intervention; however, it would complement existing EGMs addressing child welfare.

Objectives: To identify, appraise and bring together the evidence on the use of intergenerational practice, to answer the following specific research questions:

What is the volume, nature and diversity of research on, and evaluation of, intergenerational practice and learning?

Fiona Campbell and Rebecca Whear are joint first authors.

What approaches have been used to deliver intergenerational activities and programmes that may be relevant to providing such services during and in the subsequent recovery from the COVID-19 pandemic?

What promising intergenerational activities and programmes have been developed and are being used but have not yet been subject to formal evaluation?

Search Methods: We searched MEDLINE (via OvidSp), EMBASE (via OvidSp), PsycINFO (via OvidSp), CINAHL (via EBSCOHost), Social Policy and Practice (via OvidSp), Health Management Information Consortium (via OvidSp), Ageline (via EBSCOhost), ASSIA (via ProQuest), Social Science Citations Index (via Web of Science), ERIC (via EBSCOhost), Community Care Inform Children, Research in Practice for Children, ChildData (via Social Policy and Practice), the Campbell Library, the Cochrane Database of Systematic Reviews and the CENTRAL database between 22 and 30 July 2021. We searched for additional grey literature via the Conference Proceedings Citation Index (via Web of Science) and ProQuest Dissertation & Theses Global and via relevant organisation websites, for example, Age UK, Age International, the Centre for Ageing Better, Barnado's, Children's Commission, UNICEF, Generations Working Together, the Intergenerational Foundation, Linking Generations and The Beth Johnson Foundation) and the Ottawa initiative called Older Adults and Students for Intergenerational support.

Selection Criteria: Any intervention that brings older and younger people together with the purpose of interacting to achieve positive health and/or social and/or educational outcomes from any study design including systematic reviews, randomised controlled studies, observational studies, surveys and qualitative studies are included. The titles and abstracts, and later full texts, of records identified by the search methods were screened against inclusion criteria by two independent reviewers.

Data Collection and Analysis: Data extraction was undertaken by one reviewer and checked by a second with any inconsistencies identified and resolved through discussion. The data extraction tool was developed on EPPI reviewer and was modified and tested through stakeholder and advisor consultation, and piloting of the process. The tool was informed by the research question and the structure of the map. We did not undertake quality appraisal of the included studies.

Main Results: Our searches identified 12,056 references, after screening 500 research articles were included in the evidence gap map conducted across 27 countries. We identified 26 systematic reviews, 236 quantitative comparative studies (of which 38 were randomised controlled trials), 227 were qualitative studies (or had a qualitative element), 105 were observational studies (or had elements of observational methods) and 82 used a mixed methods approach. The outcomes reported in the research cover mental health ($n = 73$), physical health ($n = 62$), attainment and knowledge ($n = 165$), agency ($n = 174$), mental wellbeing ($n = 224$), loneliness and social isolation ($n = 54$), attitudes towards the other generation ($n = 283$), intergenerational interactions ($n = 196$), peer interactions ($n = 30$) and

health promotion ($n = 23$) and including mutual outcomes such as the impact on community ($n = 37$) and perceptions on the sense of community ($n = 43$). Gaps in the evidence that were identified include: research that reports on mutual, societal and community outcomes of intergenerational interventions; more research on interventions classified as levels 1–4 and level 7 on the Intergenerational Engagement Scale, mental health, loneliness, social isolation, peer interactions, physical health and health promotion outcomes in children and young people; health promotion in older people; outcomes centred on care giver wellbeing, mental health and attitudes; economic outcomes; process outcomes and adverse or unexpected outcomes.

Authors' Conclusions: Whilst a substantional amount of research on intergenerational interventions has been identified in this EGM, as well as the gaps identified above, there is a need to explore promising interventions not yet formally evaluated. Research on this topic is gradually increasing, and systematic reviews will be important to determine how and why interventions are or are not beneficial. However, the primary research needs to build more cohesively so that the findings can be comparable and avoid research waste. The EGM presented here will nevertheless be a useful resource for decision-makers allowing them to explore the evidence with regard to the different interventions that may be relevant to their population needs and the settings or resources available to them.

1 | PLAIN LANGUAGE SUMMARY

1.1 | Large evidence base for impact of intergenerational interventions involving young and old, but many gaps in research

There is a considerable body of research evidence on intergenerational interventions and their impact on older people and children and young people. However, there are still many research gaps, and primary research could benefit from more consistency in outcome reporting.

1.2 | What is this evidence and gap map about?

Opportunities for social connection between generations in the UK have diminished over the last few decades because of changes in the way that we live and work. The Office for National Statistics Community Life Survey 2020-2021 reports that 6% of adults in the UK said they often or always felt lonely. People aged 16 to 24 were significantly more likely to report feeling lonely often or always, which is 11% of that age group. Nine percent of people aged 65 years and over reported the same.

Evidence suggests that intergenerational activity can have a positive impact on participants, for example, in reducing loneliness and exclusion for both older people and children and young people, improving mental health, increasing mutual understanding, and addressing important issues such as ageism, housing and care.

However, knowing what to implement, how and for whom is complex due to the lack of evidence about their effectiveness, transferability of effects across settings and cost-effectiveness. This evidence gap map (EGM) identifies the nature, volume and types of intergenerational interventions found in the research literature. It identifies areas for future research and evidence synthesis to help decision makers make more informed choices.

1.3 | What is the aim of this evidence and gap map (EGM)?

The aim of this EGM is to identify all the existing research evidence on intergenerational interventions to improve understanding about intergenerational activities in terms of the health and social care outcomes of older people, younger people and children, and to inform future research.

1.4 | What studies are included?

The EGM includes 500 research articles of any design on intergenerational interventions that do not include family members. The evidence comes from 27 countries.

We identified 26 systematic reviews, 236 quantitative comparative studies (of which 38 were randomised controlled trials), 227 qualitative studies (or had a qualitative element), 105 observational studies (or had elements of observational methods) and 82 with a mixed-methods approach.

1.5 | What are the main findings of this EGM?

The most commonly reported outcomes for children and young people were attitudes towards older people, knowledge and attainment, and intergenerational interactions.

For older people the most commonly reported outcomes were mental wellbeing, agency, attitudes towards younger people, and intergenerational interactions.

We identified several gaps in the research, including research on mutual, societal and community outcomes, young people's mental health, loneliness, social isolation, peer interactions, physical health and health promotion, outcomes centred on caregiver wellbeing, mental health and attitudes, and adverse or unexpected outcomes, including economic outcomes.

Interventions were most commonly delivered in schools, in the community or in care homes.

Interventions most commonly involved activities related to sharing perspectives of being an older or younger person/child, spending time together, helping with chores, helping more generally within a school environment, mentoring, art and crafts to engage the generations together, learning or sharing music and playing games.

1.6 | What do the findings of the map mean?

The EGM provides a starting point for researchers and decision makers to access the available research evidence on the effectiveness of intergenerational interventions.

The map demonstrates considerable diversity in the types of intergenerational activity. It also shows that it is mainly demonstration projects that are evaluated.

The quality of the evaluations makes analysis of their effectiveness, and hence their impact on shaping practice and policy, limited.

Methods of supporting useful evaluations of these types of interventions – so they are measuring meaningful outcomes – is needed. This EGM identifies many areas where there are still gaps in research.

1.7 | How up-to-date is this EGM?

The authors searched for studies published up to July 2021.

2 | BACKGROUND

2.1 | Introduction

2.1.1 | The problem, condition or issue

Opportunities for social connection between generations in the UK have diminished over the last few decades because of changes in the way that we live and work (Kingman, 2016; United for all Ages, 2017). Housing and economic trends have seen younger people move to live in city centres whilst the older generation live in towns and rural areas. A report published by the Intergenerational Foundation in 2016 (Kingman, 2016) suggests that in the 25 biggest cities within the UK only 5% of people aged over 65 live in the same neighbourhood as someone under the age of 18. Furthermore, even when people from different age groups do live in the same area, the decline in spaces such as libraries, youth clubs and community centres mean that there are fewer opportunities to meet and mix socially with other generations outside our own families. Increased working hours, improved technology, changes in family patterns, relationship breakdowns within families and migration are also believed to be contributory factors to generation segregation (Generations Working Together, 2019). There are many potential economic, social and political impacts of generations living separate and parallel lives, for example, higher health and social care costs, an undermining of trust between generations (Brown & Henkin, 2014; R. L. Jones, 2011; Laurence, 2016; Vitman et al., 2013); reduced social capital (Laurence, 2016); a reliance on the media to form understanding of others' viewpoints (Edström, 2018; Vasil & Wass, 1993) and higher levels of anxiety and loneliness. A review of the prevalence of loneliness in 113 countries found high levels of loneliness for a substantial proportion of the population in many countries (Surkalim et al., 2022). For example, in the Office for National Statistics Community Life Survey, 2020 to 2021 (ONS, 2021); 6% of adults in the UK reported feeling lonely often or always. Those aged 16–24 were also significantly more likely to report feeling lonely often or always (11% of that age group) with 9% of those aged 65 years and over report the same.

2.1.2 | The intervention

Intergenerational programmes and activities can take many forms and are delivered in many settings, very often by third sector organisations. Although evidence suggests that intergenerational activity can have a positive impact on participants (e.g., reducing loneliness and exclusion for both older people and children and young people, improving mental health, increasing mutual understanding and addressing important issues such as ageism, housing and care), commissioning decisions are complex due to the apparent wealth of options available, and yet limited and varying resources with which to provide them. This evidence gap map brings together all the available research evidence on intergenerational interventions.

2.2 | Why it is important to develop the EGM

Intergenerational programmes and activities are promising interventions that can address some of the needs of both children and young people and older people. The outcomes for children and young people and older people will form one of the key dimensions for the EGM—the list of which were developed from the frameworks listed below and through discussion with our stakeholder advisory group. The other dimension will be type of intergenerational intervention as categorised by the Depth of Intergenerational Engagement Scale (Kaplan, 2004). These two dimensions will give an overall picture of broad types of interventions and outcomes that have, and have not, been researched. Intergenerational interventions can take many forms and are delivered in diverse settings, therefore it will be important to be able to distinguish which aspects and characteristics of the interventions are supported by the evidence. We will therefore use the filter function in the EGM to identify the research design, intervention setting, age of the children/young people involved, the focus or activities involved, and any participant characteristics that have been targeted by an intervention.

Although evidence suggests that intergenerational activity can have a positive impact on participants, commissioning decisions are complex due to the lack of evidence about their effectiveness, transferability of effects across settings, and cost-effectiveness. This evidence and gap map (EGM) will identify the nature, volume and types of intergenerational interventions that have been undertaken and evaluated. It will identify areas for future research and evidence synthesis.

There are currently no other EGMs that exist that address this type of intervention; however, it would complement existing EGMs addressing child welfare.

3 | OBJECTIVES

We aim to use existing evidence to improve understanding about intergenerational activities in terms of the health and social care outcomes of older people, younger people and children.

Our objectives are to:

Identify and bring together the evidence on the use of intergenerational practice, to answer the following specific research questions:

- What is the volume, nature and diversity of research on, and evaluation of, intergenerational practice and learning?
- What approaches have been used to deliver intergenerational activities and programmes that may be relevant to providing such services during and in the subsequent recovery from the COVID-19 pandemic?
- What promising intergenerational activities and programmes have been developed and are being used but have not yet been subject to formal evaluation?

4 | METHODS

4.1 | EGM: Definition and purpose

EGMs are maps of a specific sector or subsector which typically includes both systematic reviews and primary studies. Produced using the same systematic approach as systematic reviews, EGMs usually show what evidence is there, not what the evidence says (White et al., 2018).

The EGM framework will inform the inclusion and exclusion criteria of the EGM. Here, we describe the population, intervention, comparison, outcomes (indicators) and study designs for the map.

4.2 | Framework development and scope

The aim of this EGM is to capture the broad range of evidence from systematic reviews and primary research that has investigated intergenerational practice.

The EGM will enable policymakers and practitioners in the field to take account of the research evidence in the commissioning and use of intergenerational practice in health and social care. It will also highlight opportunities for intergenerational activities and programmes during and in the subsequent recovery from the COVID-19 pandemic and direct the commissioning of appropriate research where there are evidence gaps.

The scope of the EGM is defined by a framework of interventions and outcomes presented as two dimensions: the rows include interventions with sub-categories, and the columns outcome domains. The framework was developed in consultation with our stakeholders who identified how the interventions could be helpfully defined using an existing framework which categorises interventions based on the level of engagement they promote Depth of Intergenerational Engagement Scale (Kaplan, 2004). We identified several outcomes that the research literature in this area already reports on, however we were aware that using the literature alone does not help us to identify outcomes that may be of interest but are not reported on. To address this issue, we asked our stakeholders to review the list of outcomes we had drawn from the literature and suggest additional outcomes that they felt were also of interest/importance. All these outcomes were then captured in the framework for the map. For the benefit/ease of those using the map the outcomes were grouped into the following subsections, outcomes for children and young people, outcomes for older people, mutual outcomes, for example, community, outcomes for others, for example, carers, economic outcomes, process outcomes and adverse or unexpected outcomes, so that they could be expanded or collapsed depending on the preferences of the user.

Further attributes can be considered and used to filter the results, such as the research design of the included studies or characteristics of the included populations, for example, age of the younger people, any people with vulnerable or protected characteristics. Each cell shows studies which contain evidence on that

combination of intervention and outcome. Study characteristics including, for example, study design, setting, intervention level and intervention activity/focus are coded, and the evidence can be filtered by these characteristics.

4.3 | Stakeholder engagement

The following individuals have contributed to the project through the advisory group:

Ronald Amanze; Iain Lang—University of Exeter; Vicki Goodwin—University of Exeter; Jo Day—University of Exeter; Aideen Young - Centre for Ageing Better; G.J. Melendez Torres—University of Exeter; Dylan Kneale—UCL; Ruth Garside—University of Exeter; Claire Goodman—University of Hertfordshire; Tracey Howe—Cochrane Campbell Global Ageing Partnership; Kelvin Yates—AgeUK Cornwall; Nathan Hughes—University of Sheffield; Debbie Hanson—Sheffield City Council; Laura Abbott—Chilypep; Hannah Fairbrother—University of Sheffield; Kerry Albright—Unicef; Rachel Staniforth—Public Health; Girish Vaidya—Sheffield Children's NHS Foundation Trust; Sally Pearse—Sheffield University.

Members of the 'Only Connect!' Network have contributed throughout the project. The group includes local, national and international members from the care sector, local government, academia, people living with dementia, schools and leading organisations involved in providing intergenerational activities. Members of the group also facilitated discussion of the project with older people, people living with dementia, and young people with experience of taking part in intergenerational activities.

We convened three virtual whole project meetings to include stakeholders and advisory group members (during Months 1 and 3), which assisted with understanding and presentation of the evidence in the EGM. We used break-out rooms and other methods of sharing ideas and suggestions such as a JamBoard and individual meetings to ensure that as many views and perspectives were captured as possible. We followed large meetings up with smaller meetings/phone calls where necessary.

Between meetings we involved people through email, telephone and video conferencing, depending on the nature of the involvement and the preference of individuals.

During the stakeholder meeting in month one the stakeholder group informed the development of the framework, which helped to form the matrix for the EGM. Working in smaller groups, we encouraged participants to identify outcomes and types of intervention. This was used, along with the wider literature to inform the components of the framework.

4.4 | Conceptual framework

We developed a broad logic model to portray the general theory/pathway expected in any intergenerational intervention (Figure 1).

Our conceptual framework is informed by the following: the five essential elements of wellbeing described by Nazroo and colleagues adopted by the Institute for Public Policy Research (IPPR) (Nazroo et al., 2005); the seven outcomes outlined in the Department of Health Social Care Green Paper, Independence, Well-being and Choice (DOH, 2005) and the six domains identified in which actions

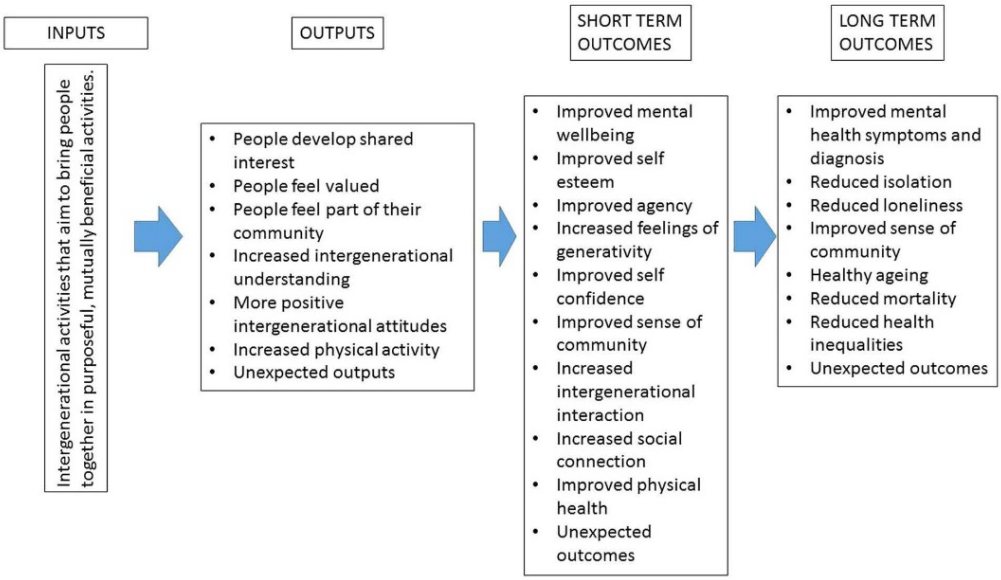

FIGURE 1 Logic model.

are required for child and adolescent health and wellbeing by the World Health Organisation and UNICEF (UNICEF & WHO, 2020).

These were then further considered and discussed with our stakeholders to identify the relevant outcomes of interest for one dimension of the framework. The other dimension of the framework was informed by the Depth of Intergenerational Engagement Scale (Kaplan, 2004) which gives a broad category on intervention based on the level of engagement it requires between the two generations. As intergenerational interventions are delivered using different formats and in diverse settings, it is important to be able to easily identify intervention characteristics such as research design, intervention setting, age of the children/young people involved, the focus or activities involved and any participant characteristics that have been targeted by an intervention. We will use the filter function in the EGM to capture and present these and these are further detailed below. We expect the interventions to cover both universal and targeted approaches, and whilst these definitions are not explicitly used as a filter in the map, targeted approaches will be identifiable by the filters used to describe particular characteristics of the populations involved in the intervention.

Five essential elements of wellbeing (Nazroo et al., 2005)	Seven outcomes in the social care Green Paper, Independence, Well-being and Choice (DOH, 2005)	Six domains identified in which actions are required for child and adolescent health and wellbeing (UNICEF & WHO, 2020)
Resilience	Improved health and emotional well-being	Good health
Independence	Improved quality of life	Adequate nutrition
Health	Making a positive contribution	Opportunities for learning and education
Income and wealth	Increased choice and control	Securing, safety and a supportive clean environment
Having a role and having time	Freedom from discrimination or harassment	Responsive relationships and connectedness
	Economic well-being	Realisation of personal autonomy and resilience
	Maintaining personal dignity and respect	

4.5 | Dimensions

4.5.1 | Types of study design

We wanted to capture all the available evidence (not just intervention effectiveness) regarding intergenerational interventions for users to be able to use the EGM to identify any research they were interested in and where the gaps in evidence still lie. Therefore, all study designs including systematic reviews, randomised controlled studies, observational studies, surveys and qualitative studies are included. Due to the substantial amount of research literature found we did not include news items describing intergenerational activities and programmes even if they reported innovative interventions not otherwise represented within the evidence base (as in the protocol).

4.5.2 | Types of intervention/problem

We included any intervention that brings older and younger people together with the purpose of interacting to achieve positive health and/or social and/or educational outcomes. These include reminiscence programmes, buddy systems, storytelling, school-based interventions and arts-based interventions as well as others. We used the Depth of Intergenerational Engagement Scale (Kaplan, 2004) as the framework for the interventions. This is described below:

4.5.2.1 | *The Depth of Intergenerational Engagement Scale*

The Depth of Intergenerational Engagement Scale places programmes and activities on a continuum, with points that correspond to different levels of intergenerational engagement, ranging from initiatives that provide no direct contact between age groups (point 1) to those that promote intensive contact and ongoing opportunities for intimacy (point 7). Examples of intergenerational initiatives fitting into each point on the scale are described.

1. Learning about other age groups

   Participants learn about the lives of persons in other age groups, although there is no direct or indirect contact.

   Example: 'Learning about Aging' programmes designed to teach youth about aspect(s) of the aging process.
2. Seeing the other age group at a distance

   These initiatives facilitate an indirect exchange between individuals of two or more age groups. Participants might exchange videos, write letters, or share artwork with each other,

but never actually meet in person.

Example: A pen-pal programme in which youth in an after-school club exchange letters with residents of a nursing home.

3. Meeting each other

Initiatives culminate in a meeting between the young participants and older adults, generally planned as a one-time experience.

Example: A class of students plan for and visit a local senior centre in which all engage in activities during a July 4th picnic.

4. Annual or periodic activities

Often tied to established community events or organisational celebrations, intergenerational activities occur on a regular basis. Although infrequent, these activities might symbolise intergenerational and community unity and influence attitudes and openness towards additional or ongoing activities.

Examples: Intergenerational activities at a school on Grandparent's Day, an annual community dance in which youth and older adults are actively involved, and Christmas caroling at assisted-living homes.

5. Demonstration projects

Demonstration projects generally involve ongoing intergenerational activities over a defined period of time. Depending on project goals and objectives, the intergenerational exchange and learning can be quite intensive. These initiatives are often implemented on an experimental or trial basis, and frequently depend on external funding.

Example: A 6-month pilot programme, sponsored by an agency that provides teen parenthood support services. Senior adults who have successfully raised children are enlisted to mentor and provide support for pregnant and parenting teens.

6. Ongoing intergenerational programmes

Programmes from the previous category that have been deemed successful and valuable from the perspective of the participating organisations and the clientele are incorporated as an integral part of their operation. This extends to programme and staff development such as preparing individuals to work with populations of various age groups.

Example: Based on a partnership forged between a senior centre, a community youth centre, and an environmental education centre, senior adults and youth plan and execute the town's environmental improvement campaign. Systems are established to organise numerous projects, train and assign participants, and provide continuing support and recognition.

7. Ongoing, natural intergenerational sharing, support and communication

There are times when the intergenerational reconnection theme transcends a distinct programme or intervention. This is evident when the social norms, institutional policies and priorities of a particular site, community, or society reflect values of intergenerational reciprocity and interdependence. Intergenerational engagement takes place as a function of the way community settings are planned and established. In this context, opportunities for meaningful intergenerational engagement are abundant and embedded in local tradition.

Example: A YMCA facility houses a senior citizen centre. Older adults and youth participate in a variety of age-integrated activities.

Programmes fitting into all points on this continuum provide positive experiences for interacting with persons in other age groups. However, if the aim is ambitious, such as changing attitudes about other age groups, building a sense of community, enhancing self-esteem, or establishing nurturing intimate relationships, it becomes important to focus on programmes that fit into Levels 4–7 on the scale. Programmes would take place over an extended period of time, would last anywhere from a few months to many years, and would provide extensive interaction opportunities (Kaplan, 2004).

4.5.3 | Types of population

Older adults and children and young people. No age boundary restrictions were applied but we sought studies that suggest at least one skipped generation between the older and younger participants. Studies in which participants were related by family or marriage were excluded. Inclusion was not determined by prior age cut-offs but by the included studies own definition of 'older people' and 'young people'.

4.5.4 | Types of outcome measures

We included all reported outcomes. Outcomes did not form part of the criteria for including studies in the EGM since we are keen to explore all of the available evidence.

4.5.5 | Other eligibility criteria

Types of settings

Any setting or context. No restrictions on language.

Status of studies

We included studies irrespective of their publication status and their electronic availability. We also aimed to include ongoing studies where it was feasible to ascertain when the study will be completed.

4.6 | Search methods and sources

We searched MEDLINE (via OvidSp), EMBASE (via OvidSp), PsycINFO (via OvidSp), CINAHL (via EBSCOHost), Social Policy and Practice (via OvidSp), Health Management Information Consortium (via OvidSp), Ageline (via EBSCOhost), ASSIA (via ProQuest), Social Science Citations Index (via Web of Science), ERIC (via EBSCOhost), Community Care Inform Children, Research in Practice for Children, ChildData (via Social Policy and Practice), the Campbell Library, the

Cochrane Database of Systematic Reviews and the CENTRAL database between 22nd and 30th July 2021.

We used terms covering intergenerational practice, or terms for older adults combined with terms for children and intergenerational activities. The full search strategies for every database are available in Supporting Information: Appendix 1. We searched for additional grey literature via the Conference Proceedings Citation Index (via Web of Science) and ProQuest Dissertation & Theses Global.

We expected that some relevant reports would not be published in academic sources so we also searched for grey literature via relevant organisation websites, for example, Age UK, Age International, the Centre for Ageing Better, Barnado's, Children's Commission, UNICEF, Generations Working Together, the Intergenerational Foundation, Linking Generations and The Beth Johnson Foundation and the Ottawa initiative called Older Adults and Students for Intergenerational support (OASIS, https://www.oasis-aesi.com/) between 28 January 2022 and 4 February 2022 by either examining the resources section of the website or entering 'intergenerational' into the search box.

Due to the amount of research literature found we limited our additional searches (forwards and backwards citation chasing) as follows: we carried out backward citation chasing on the included systematic reviews to identify any randomised controlled trials (RCTs) and other systematic reviews not already included in the EGM; we did not check the citations of older key papers (forward citation chasing); we hand searched one key—journal the Journal of Intergenerational Relationships. Although we did not conduct the horizon scanning process described in the protocol we expect to conduct that in subsequent reviews.

We published the agreed protocol with Campbell (Thompson-Coon et al., 2022).

4.7 | Analysis and presentation

4.7.1 | Report structure

The report provides tabulations or graphs of the number of studies, with accompanying narrative description, by

- Intervention category and subcategory
- Outcome domain and subdomain
- Table of 'aggregate map' of interventions and outcomes
- Country (designated by country of first author)
- Year
- Study type
- Population subgroups.

The interactive EGM can also be used to explore the data using the filters presented below.

4.7.2 | Filters for presentation

In addition to the interventions and outcomes, the following filters have been coded:

Characteristics of the participants that the intervention targets (this was an iterative list that also aimed to include characteristics included in Progress Plus (O'Neill et al., 2014)

Progress plus:

- Minority groups (in either generation based on race, ethnicity, culture, language, LGBTQ)
- Low socioeconomic status (in either generation)
- Unemployment (in either generation)
- Educational needs (in either generation
- Social isolation (in either generation)

Other important characteristics (discussed with the Stakeholder advisory group):

- Mental health difficulties (*in either generation*)
- Physical health difficulties (*in either generation*)
- Age category of the *children/young people*—0–5 years, 6–12 years, 12–18 years, 19–30 years.
- *Children* experiencing childhood adversity
- *Older people* with cognitive impairment

Contextual factors:

- Country/region—country of the first author
- Setting—where the intervention took place, for example, in school, care home, retirement village, university/higher education, shared facility, day care centre, hospital, assisted living centre or community setting

Study design factors:

- Study design—RCTs, non-RCTs, interrupted time series, controlled before and after studies, observational studies, qualitative studies, mixed methods and systematic reviews

Focus of the intervention (the activities involved in the intervention):

- Education—where older or younger generations teach the other generation a skill or share educational knowledge
- Art—generations share in arts or crafts
- Music—generations share musical activities or teach a musical skill
- Interaction—interaction between the generations like conversation, spending time/communication, helping tasks
- Cooking—generations cooking together
- Dance—generations sharing and working together in dance performances

- Drama—generations sharing and working together in dramatic performances
- Environmental activities—generations sharing environmental activities
- Exercise—generations exercising together or helping the other generation to exercise more
- Gardening—generations gardening together
- History—older generations helping to share history with younger generations
- IT—younger generations helping older generations to learn and use technology
- Language—older generations helping younger generations to learn/practice language
- Letter writing—generations writing to each other and to help learn to write
- Literature—generations sharing literature together
- Living together—generations living in the same space (usually students/young adults living with older generation—with no familial connection)
- Maths—older generation helping younger generation to learn Maths
- Playing games—generations playing games together
- Professional education—older generation involved in professional education of students working with older generations
- Reading—older generation helping younger generation to learn to read
- Reminiscence—older generations encouraged to reminisce by presence of younger generation
- Science activities—generations conduct science activities together
- Sharing meals—generations share a meal together
- Sharing perspectives (of being and older person/a child/young person)
- Story telling—one generation tells a story to another
- Trips and excursions—generations visit places or attend events together
- Other—any intervention not covered by the descriptions above, for example, general presence/assistance in a school context.

4.7.3 | Dependency

Each entry in the map is a systematic review or a primary study of effectiveness. The final EGM identifies the number of studies covered by the map in each sector or subsector. We have included all relevant systematic reviews and primary studies irrespective of whether there is overlap between reviews and studies. Similarly, studies with multiple interventions or multiple outcomes may appear multiple times within the map.

4.8 | Data collection and analysis

4.8.1 | Screening and study selection

The titles and abstracts of records identified by bibliographic and supplementary search methods were screened against inclusion criteria by two independent reviewers (FC, JTC, RW, MR) looking for reasons for exclusion. The full text of records retained at this stage were retrieved and screened for inclusion against the inclusion criteria using the same process. All included studies were saved in a master library using EndNote X8 Endnote X8. These studies were then entered on to EPPI reviewer where the remaining data extraction and management was conducted. These are the studies that form the basis for the EGM and that can also be used in the next phase of this project, for example, subsequent review topics.

4.8.2 | Data extraction and management

Data extraction was undertaken by one reviewer and checked by a second (FC, JTC, RW, MR) with any inconsistencies identified and resolved through discussion. The data extraction tool was modified and tested through stakeholder and advisor consultation, and piloting of the process. The tool was informed by the research question and the structure of the map. Data extraction was conducted using EPPI reviewer (Thomas et al., 2022).

We extracted data on study design, geographical location, setting, population (age, gender, health condition/status, equity characteristics), intervention (type, mode of delivery, setting) and outcomes.

We used the PROGRESS-Plus framework (O'Neill et al., 2014) to identify studies that measured effects of interventions by gender or other factors that may lead to health inequalities (e.g., ethnicity; etc.).

4.8.3 | Tools for assessing risk of bias/study quality of included reviews

We did not undertake quality appraisal of the included studies.

4.8.4 | Methods for mapping

We used EPPI-Reviewer software (Thomas et al., 2022) for data extraction and coding, and to generate the online EGM (EPPI Mapper 2022). The map is interactive so that users can click on (i) cells within the matrix to show a list of the relevant studies and on (ii) study names to access the study or a reference and database link for the study.

5 | RESULTS

5.1 | Description of studies

5.1.1 | Results of the search

Our search strategy identified 12,056 references (reduced to 8638 after removal of duplicate studies). After both stages of screening had

been completed a total of 500 research articles were included in the EGM. Figure 2—PRISMA flow diagram provides further details on the screening process and decisions at each stage (Page et al., 2021).

Studies were conducted in 27 countries (based on country of first author). Studies were conducted in the US ($n = 326$), Canada ($n = 33$), the UK ($n = 29$), Australia ($n = 27$), Japan ($n = 15$), Spain ($n = 8$), Hong Kong ($n = 7$), Italy ($n = 7$), South Korea ($n = 5$), Brazil, France, Portugal, Singapore, Taiwan, Israel ($n = 4$ in each), Sweden, the Netherlands ($n = 3$ in each), Germany, Ireland, China ($n = 2$ in each), one each in Austria, Finland, Greece, Malta, New Zealand, South Africa and Switzerland.

The 500 research studies were published over a period of 46 years from 1975 to 2021. All study designs were included, we identified 26 systematic reviews, 236 quantitative comparative studies (of which 38 were RCTs), 227 were qualitative studies (or had a qualitative element), 105 were observational studies (or had elements of observational methods) and 82 used a mixed methods approach. We did not record the age of the older generations involved in the intergenerational interventions as we were looking more closely for evidence of a generational gap between the two populations; however, we did record the ages of the young people and children involved in the interventions which spanned from 0 to 30 years. One hundred and twenty-two interventions involved children aged between 0 and 5 years, 182 interventions involved children aged 6–12 years, 137 interventions involved young people aged 12–18 years, and 155 interventions involved young people aged 19–30 years. In 39 intervention studies the age range could not be established.

Outcomes included (but were not limited to) social isolation, engagement, interacting, perception of people living with dementia, social inclusion, psychological outcomes, depression, anxiety, social skills, self-confidence, creativity, school performance, relationship building, attitudes, empathy, personal growth, community responsibility, activity levels (physical activities), mood, quality of life, stimulation of memory and mind, digital inclusion (helping people to get online). Figures 2–4 depict snapshots of how the EGM looks and how the studies are presented across the dimensions of intervention level and outcomes for children and young people (Figure 3), older people (Figure 4) and outcomes other people (e.g., carers), mutual outcomes (e.g., sense of community), economic outcomes, process outcomes, and adverse or unexpected outcomes (Figure 5).

5.1.2 | Excluded studies

Of the 794 reports assessed for eligibility, 303 reports were excluded. One-hundred and eighty-six reports were excluded as they were considered the wrong study type, for example, reports that did not detail their research methods, descriptions or summaries of interventions, or were personal reports/descriptions of an intervention; 47 were excluded because they included the wrong population, for example, where 'intergenerational' referred to a spread across

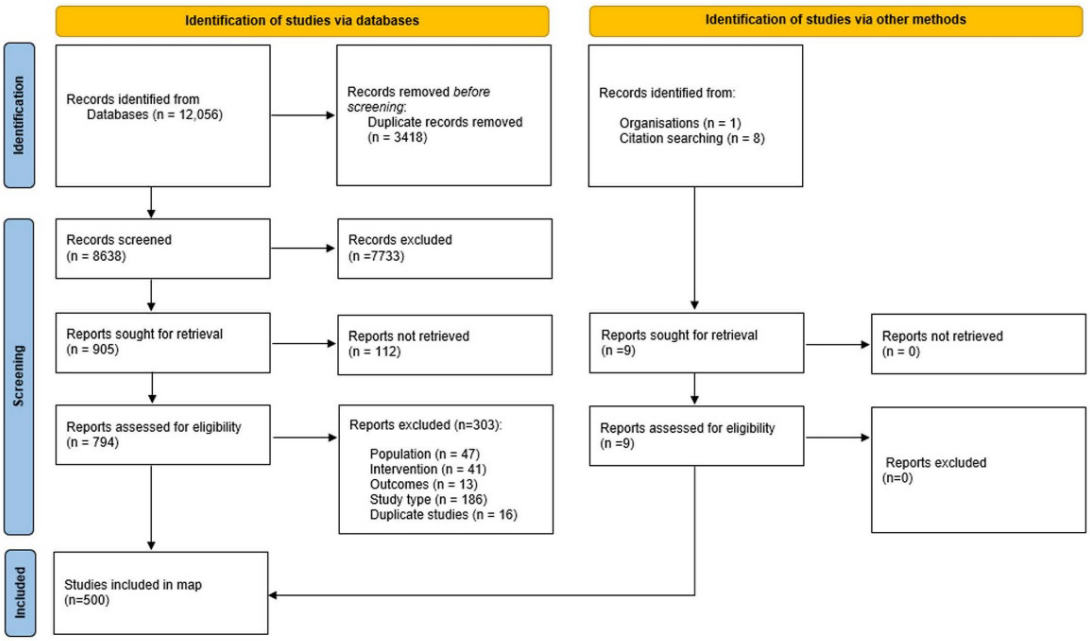

FIGURE 2 Prisma flow diagram.

FIGURE 3 Figure 3 EGM aggregate map interventions × outcomes (children and young people).

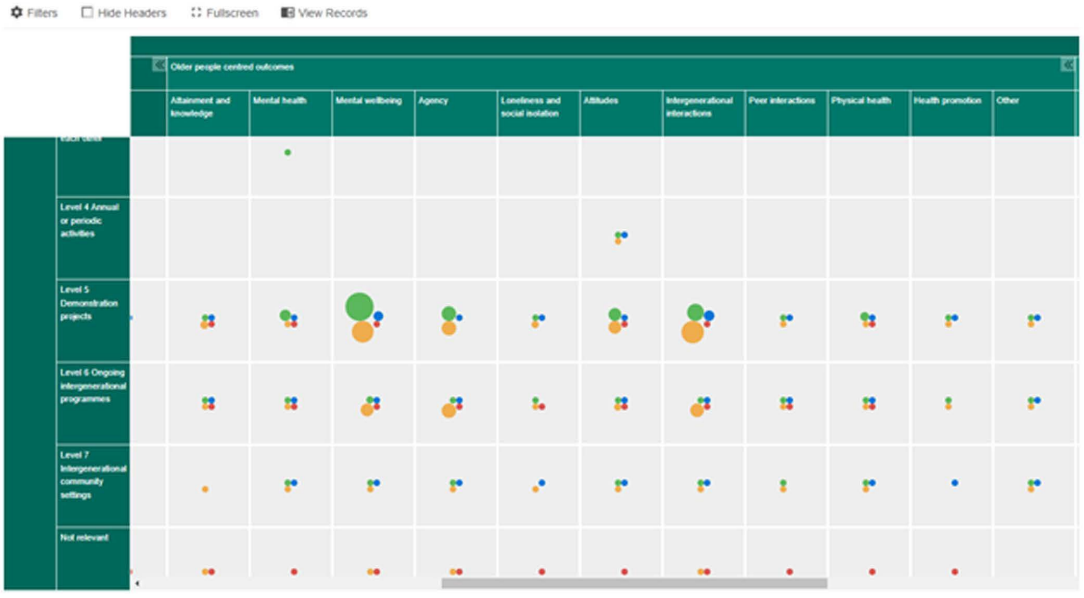

FIGURE 4 Figure 4 EGM aggregate map interventions × outcomes (older people).

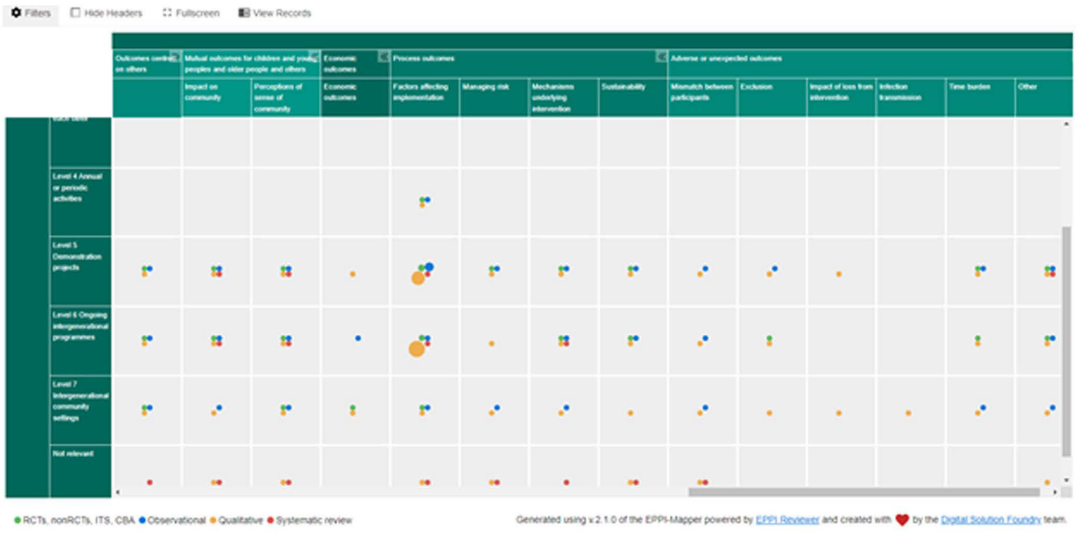

FIGURE 5 Figure 5 EGM aggregate map interventions × outcomes (other people, mutual outcomes, economic outcomes, process outcomes, adverse or unexpected outcomes).

generations or where a generational gap could not be ascertained; 41 were excluded based on ineligibility of the intervention, for example, where the generations did not have direct contact or interact; 13 were excluded because they did not report on participant outcomes; 16 were excluded as they were duplicate reports. All 303 excluded studies are listed in Supporting Information: Appendix 2.

5.1.3 | Studies awaiting classification (if applicable)

None identified.

5.2 | Synthesis of included studies

The interactive map can be found here.

5.2.1 | Intervention level

We used the Depth of Intergenerational Engagement Scale (Kaplan, 2004) as the framework for describing the interventions identified in this EGM. By the nature of the eligibility criteria for this EGM interventions that would have been classified as Level 1 (Learning about other age groups—participants learn about the lives of persons in other age groups, although there is *no direct or indirect contact*) or level 2 (Seeing the other age group at a distance—these initiatives facilitate an *indirect* exchange between individuals of two or more age groups. Participants might exchange videos, write letters, or share artwork with each other, but never actually meet in person) are not represented as they did not meet the eligibility criterion with regard to the generations having direct contact/interaction with each other.

In Table 1 we can see that the included interventions most commonly fall within Level 5 (Demonstration projects—generally involve ongoing intergenerational activities over a defined period of time, $n = 284$) or Level 6 (Ongoing intergenerational programmes—Programmes from the previous category that have been deemed successful and valuable from the perspective of the participating organisations, $n = 155$) with a seemingly increasing (based on the frequency of published studies in the last 5 years) number of Level 7 interventions (Ongoing, natural intergenerational sharing, support and communication—evident when the social norms, institutional policies and priorities of a particular site, community, or society reflect values of intergenerational reciprocity and interdependence, $n = 35$). This is what we would expect to see when looking for *research* in this area because interaction between generations described in interventions in Levels 3 and 4 is less likely to conform to an intervention that could be tested in a research study. However, this doesn't mean that this type of interaction is not being facilitated by organisations in practice.

Some examples of the interventions identified in Levels 3–7 are:

Level 3—Developing one-one relationships via instagram (Lytle et al., 2020) or the Intergenerational Partners Project where 4th Grade students share activities with older people to develop friendships (Aday et al., 1996).

Level 4—An intergenerational dinner event where medical students and older people attended together and participated in dancing and games together (Diachun et al., 2007; Dumbrell et al., 2007).

TABLE 1 Study design of evidence present in each intervention level.

Intervention level/study design	Systematic review	RCT	Non-RCT	Qualitative	Observational	Mixed methods	Total studies in EGM *
1	0	0	0	0	0	0	0
2	0	0	0	0	0	0	0
3	0	1	3	0	0	0	4
4	0	0	1	1	0	1	1
5	0	23	131	122	63	52	284
6	0	14	53	82	28	25	155
7	0	0	11	19	14	4	35
Total	26	38	198	227	105	82	

Note: *This is the number of studies at this level in the EGM some studies are represented in more than one study design category hence this number does not represent the total number in the relevant row.

Abbreviation: RCT, randomised controlled trial.

TABLE 2 Broad outcomes reported across included studies.

Age group/ outcome	Children/younger people's outcomes	Older people's outcomes	Other people's outcomes (e.g., carers)	Mutual outcomes	Economic outcomes	Process outcomes	Adverse outcomes
0–5	71	94	17	16	3	47	19
6–12	137	115	18	26	1	65	15
12–18	103	99	12	26	0	55	9
19–30	129	89	10	12	0	56	11
Not described	25	35	1	10	0	20	8

Level 5—These are demonstration projects aiming to see if an intervention can become a more permanent/sustainable intergenerational activity including, but not exclusively, projects that might target specific populations. For example, an intervention aiming to increase the citizenship experience of young children and their awareness of what it means to live with stroke tackling social isolation and self-confidence in older people with stroke whilst encouraging mutual fine motor skill development such as handwriting (Lane, 2016).

Level 6—Ongoing interventions that are relatively well established, such as service learning opportunities for students studying topics where intergenerational interactions will aid their learning and development of personal skills related to future employment (Howell et al., 2021); for example or the 'Through their Eyes Project' where health sciences students are partnered with older adults to explore and assess the age-friendliness of their neighbourhood (Gardner & Alegre, 2019); or 'Active Generations' an intergenerational nutrition education and activity programme implemented in out-of-school environments (after school and summer camps) where older adult volunteers implement a version of the evidence-based childhood obesity prevention programme, 'Coordinated Approach to Child Health' (Werner et al., 2012).

Level 7—Where younger generations might live with older generations in intergenerational housing projects (Hock & Mickus, 2019; Kilaberia & Ratner, 2018; Labit & Dubost, 2016) or where very young children (0–5years) have their nursery/kindergarten located within a care home setting (Doll & Bolender, 2010; Rosa Hernandez et al., 2020; Skropeta et al., 2014).

5.2.2 | Outcomes reported

Table 2 summarises broad categories of outcomes reported across the included studies and also shows how these varied depending on the age of the young people or children involved in the study. Interestingly we found that not all research in this area reported on the outcomes for both generations; some intervention studies only reported on outcomes or experiences for one of the generations with the opposite generation being considered part of the intervention itself.

More specifically the outcomes reported in the research identified in this EGM cover mental health (*n* = 73), physical health (*n* = 62), attainment and knowledge (*n* = 165), agency (*n* = 174), mental wellbeing (*n* = 224), loneliness and social isolation (*n* = 54), attitudes towards the other generation (*n* = 283), intergenerational interactions (*n* = 196), peer interactions (*n* = 30) and health promotion (*n* = 23) and including mutual outcomes such as the impact on community (*n* = 37) and perceptions on the sense of community (*n* = 43). The most commonly reported children/younger people's outcomes were attitudes towards older people, knowledge and attainment and intergenerational interactions. For older people the most commonly reported outcomes were mental wellbeing, agency, attitudes towards younger people and intergenerational interactions.

Economic outcomes (*n* = 3) and adverse or unexpected outcomes (*n* = 47) were not commonly reported but process outcomes such as factors affecting implementation, and mechanisms of interventions were reported across 183 studies.

Of those reporting adverse or unexpected outcomes (mostly from studies that used qualitative methods), 14 report time being a burden associated with the running of the intervention, 12 report a mismatch between the pairing of participants across the generations, which negatively impacted on the effects of the intervention, eight reported that some participants (or those around them) still felt excluded, three were concerned with the impact that loss might have on participants (particularly the loss of an older person with whom a younger person was interacting) and one study reported concerns about the risk around transmitting infections between older and younger participants. Other unexpected or adverse outcomes were also reported across 25 studies including negative behaviours and attitudes during interactions, and careful requirements for the design and implementation of interventions to ensure positive experiences and interactions.

Of the 183 studies reporting on process outcomes, 155 reported on factors affecting the implementation of the intervention being studied. The factors reported are dependent on the type of intervention being offered but, for example, some studies found that it was necessary to carefully select the activities available for older people and very young children (0–5years) to engage with together so as to ensure the generations were able and willing to mix, others found they needed to make sure there was a choice of activities available, whilst others working with older young people (19–30) found that sometimes extra preparation was needed for those groups to feel confident or ready to engage with their older adult counterparts. Approximately 55 studies explored mechanisms underlying the intervention being studied. Elements such as valuing interactions that incorporate learning and insights in both generations (Lane, 2016); how promoting positive experiences was key to developing meaningful and satisfying relationships (Kamei et al., 2021); how characteristics of either generation can impact on success/engagement, and how success/engagement in these interventions can impact on the characteristics of both generations. Sustainability factors were explored by 46 studies, these factors overlap with factors affecting implementation but also look forward towards resolving challenges for future interventions. Very few studies explored managing risk within the intervention (*n* = 7), and of those that did, the concerns were related to the circumstances where young people shared accommodation with older people or where young children entered an older person's setting like a day care centre or care home.

5.3 | Risk of bias in included reviews

Risk of bias was not assessed as part of this EGM as per the protocol.

5.4 | Additional dimensions (if applicable)

5.4.1 | Participant characteristics

We were able to identify studies that targeted specific participant characteristics, and these are described below.

Progress plus characteristics

Fifty-one studies targeted children and young people with vulnerable characteristics. Of these, 6 involved minority groups (institutionalised children, those affected by race or cultural differences), 13 involved children and young people from low socioeconomic backgrounds, 2 involved those experiencing social isolation, 11 involved children with educational needs and 5 involved young people who were unemployed. Eighty-eight studies targeted older people with vulnerable characteristics. Of these two involved minority groups (those affected by race or cultural differences), ten involved those from low socioeconomic backgrounds, five involved those experiencing social isolation and no interventions specifically involved older people who were unemployed. We did not identify any research that looked at other Progress Plus characteristics such as gender, LGBTQ, religion or place of residence.

Other important characteristics (discussed with the stakeholder advisory group)

Of the 51 studies that targeted children and young people with vulnerable characteristics, 6 involved those with mental health difficulties, 6 involved children with physical difficulties and 22 involved children and young people experiencing childhood adversity. Of the 88 studies that targeted older people with vulnerable characteristics, 14 involved those with mental health difficulties, 25 involved older people with physical difficulties and 49 involved older people with cognitive impairment.

Only 12 interventions involved participants with multiple vulnerability characteristics across the generations. For example, one intervention involved older people from a low income background (and some with additional physical health conditions) and young people with mental health problems (E. D. Jones et al., 2004); or young unemployed people and older people with a physical health condition (Schindler, 1992); or children with educational needs and

TABLE 3 List of settings for interventions in included studies.

Setting	Number of studies
School	162
Community	135
Care Homes	110
Higher education	70
Day care centres for older people	31
Retirement community	31
Shared site facilities	31
Assisted living facilities	25
Hospital	7
Other	28

older people with mental or physical health difficulties (Kamei et al., 2020) or where both generations shared the same vulnerability such as a physical health condition (Macmillan-Smith 1999; Sherman, 1997); low SES (Alcock et al., 2011; Carney, 1985; Kerrigan & Stevenson, 1997; La Porte, 1999; Rogers, 1994); social isolation (Jackson et al., 2019); or multiple vulnerabilities (Barbosa et al., 2020).

5.4.2 | Setting

The intergenerational interventions identified in this EGM took place across at least 10 different settings described below (Table 3). The descriptions in 25 studies were unclear where (which setting) the intervention was conducted in and where 'other' is reported in the setting (n = 28) 10 are systematic reviews covering more than one setting, eight are interventions that used digital interventions such that the true 'setting' may be mixed or unclear, seven are interventions that took place in mixed settings, and three are interventions that were conducted in a holiday/retreat type setting. None of the studies were conducted in secure institutions.

5.4.3 | Intervention focus

Approximately 25 different intervention activities (or focuses) were recorded in this EGM (Table 4). Some interventions involved multiple activities to engage the generations but others have specifically concentrated on one main approach. The most commonly reported activities were those that included sharing perspectives of being older (n = 200), in part reflecting the fact that many of these interventions have been designed to address negative stereotypes and perceptions of older or younger age groups. The limited number of evaluations of older and younger people sharing living accommodation (n = 9) possibly reflects the few examples of these types of innovations. One-hundred and sixty-four interventions also included other forms of interaction such as spending time together, helping with chores, helping more generally within a school environment and mentoring.

TABLE 4 Reported activities in intergenerational interventions.

Activity	Number of studies
Involved sharing perspectives of being an older or younger person/child	200
Interventions also included other forms of interaction such as spending time together, helping with chores, helping more generally within a school environment, and mentoring	164
Interventions used art and crafts to engage the generations together	154
Learning or sharing music	127
Involved play games together	109
Involved supporting children to learn to read	82
Involved students interacting with older people to improve their professional education and skills	68
Storytelling	66
Exercise	65
Learning or sharing history	59
Sharing meals together	58
Learning or sharing IT skills	51
Used drama	41
Dance	36
Cooking activities	36
Gardening activities	30
Joint trips, events and excursions	27
Sharing literature or learning literacy	21
Writing letters	14
Reminiscence	13
Learning or practicing a new language	12
Sharing science activities	12
Used environmental activities such as developing sustainable communities or forest school activities	11
Learning or helping with maths	10
Students or young people sharing accommodation with older people	9

5.4.4 | Bibliometric analysis

In Figure 6 we can see there has been a steady increase in the number of studies evaluating intergenerational interventions published, with the first and single study published in 1975, to 35 in

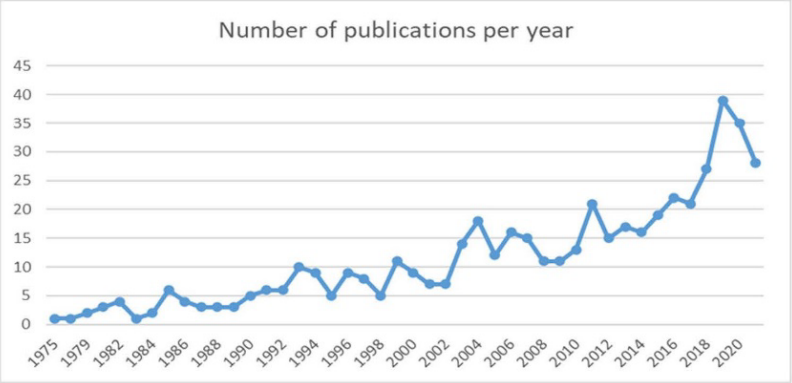

FIGURE 6 Number of publication per year.

2020. This may reflect a growing trend in evaluating these types of interventions and publishing the results, or an increase in the number of intergenerational interventions (Figure 6).

6 | DISCUSSION

6.1 | Summary of main results

This EGM presents the available evidence on non-familial intergenerational interventions that involve direct contact or interaction between younger and older people two generations apart (at least one skipped generation in between). Below we address the literature in accordance with our three research questions:

RQ1—What is the volume, nature and diversity of research on, and evaluation of, intergenerational practice and learning?

We found a substantial amount of research literature ($n = 500$ studies) in this area of varying design, setting, focus, content and outcome. There are 26 systematic reviews, 38 RCTs, 198 non-RCTs, 227 qualitative studies, 125 observational studies and 82 mixed methods studies. Most interventions include in this map are at Level 5 ($n = 284$), Level 6 ($n = 155$) or Level 7 ($n = 35$) of the Intergenerational engagement scale (Kaplan, 2004)—these are interventions with the most/deepest intergenerational engagement structures in place, and that may offer more lasting impacts on participant outcomes and be more sustainable and integrated in the future. These interventions take place in a range of settings assisted living facilities ($n = 25$), care homes ($n = 110$), community setting ($n = 135$), day care centres for older people ($n = 31$), hospital ($n = 7$), retirement community ($n = 31$), school ($n = 162$), university or higher education institution ($n = 70$) and shared site facilities ($n = 31$). The most commonly reported outcomes amongst the studies in this EGM are attainment and knowledge ($n = 165$), agency ($n = 174$), mental wellbeing ($n = 224$), attitudes towards the other generation ($n = 283$) and intergenerational interactions ($n = 196$), although mental health, physical health, loneliness and social isolation are also commonly reported. Interventions that involve people with vulnerability characteristics are also identified within this EGM.

RQ2—What approaches have been used to deliver intergenerational activities and programmes that may be relevant to providing such services during and in the subsequent recovery from the COVID-19 pandemic?

The interventions themselves report using at least one of 25 different activities as the focus for an intervention but in many occasions multiple activities are used Table 4. Some of these activities ($n = 8$) were conducted online which would enable these activities in particular to carry on amid a pandemic. Such activities included but were not limited to sharing learning or perspectives and gaming online or mentoring through videoconferencing or email, or letter writing. Some activities that can be conducted either online or in outside spaces may work for pandemic recovery periods such as gardening activities, physical exercise or leisure activities conducted outside, excursions or trips or environmental activities. Other activities that need direct in person contact through music, drama, arts and crafts might be more suited to non-pandemic times.

RQ3—What promising intergenerational activities and programmes have been developed and are being used but have not yet been subject to formal evaluation?

We were unable to answer this research question first due to the amount of research literature identified and so we were unable search for news items that would have identified interventions that exist but do not yet have research evidence available for them. Secondly, the complexity of the interventions are that 'named' interventions are not common and so what is identified in the literature are combinations of activities rather than interventions with specific models and structures.

6.2 | Areas of major gaps in the evidence

This EGM has highlighted approximately ten areas in which research evidence is lacking (evidence gaps):

1. Many of the included studies evaluated the impact of intergenerational interventions on only one of the generations, often measuring and reporting outcomes for older people only. This finding was a surprise to our stakeholders, particularly those involved in the delivery of intergenerational activities, since in their experience benefits are often observed not only in terms of personal outcomes but also mutual or societal outcomes. Future research should consider how best to measure the broader impact of intergenerational activities.
2. Research evidence for interventions categorised as Levels 1–4 and 7 in Kaplans Intergenerational Engagement Scale (Kaplan, 2004). This could be due to interventions in Levels 1–2 being excluded from this EGM as they do not involve direct/personal contact or that research on interventions at these Levels (1–4) is less frequently conducted. Level 7 interventions are larger scale and more complex to study and therefore may not have been tested or implemented so frequently.
3. Mental health outcomes in children and young people—whilst there are some studies looking at this outcome ($n = 14$) the general lack of studies measuring this outcome seems to be at odds with the amount of intergenerational research available more generally
4. Loneliness and social isolation in children and young people, both as an outcome ($n = 14$) but also as a targeted characteristic ($n = 2$)
5. Peer interactions ($n = 11$), physical health outcomes ($n = 10$) and health promotion ($n = 9$) in children and young people
6. Health promotion in older people ($n = 19$)
7. Outcomes centred on others, for example, carers, care givers… mental health ($n = 0$), mental wellbeing ($n = 12$) and attitudes ($n = 21$)
8. Economic outcomes ($n = 3$)
9. Process outcomes—such as those related to managing risk ($n = 7$)
10. Adverse/unexpected outcomes whilst often reported ($n = 47$) are not consistently measured or reliably reported.

6.3 | Potential biases in the mapping process

6.3.1 | Limitations of the EGM

Due to the amount of research literature available we did not include news items describing intergenerational activities and programmes even if they reported innovative interventions not otherwise represented within the evidence base. Whilst we recognise that this might mean the EGM is not comprehensive in terms of capturing all the existing intergenerational interventions, we are confident the EGM captures all the robust research in this area.

By nature of our inclusion criteria that specifies that 'Any intervention that seeks to bring older and younger people together to intentionally with the purpose of interacting', the EGM does not include interventions at level 1-2 where there is no direct contact between the generations. This does not mean that these types of interventions are unlikely to have an impact but they are not the focus of our research interest.

We did not conduct quality appraisal of the research studies identified. We deemed this an appropriate approach as we wanted the EGM to be as comprehensive as possible in capturing the research picture without being confusing for the viewer (quality appraisal of different study designs would have been difficult to present in the EGM without oversimplifying the appraisal, which would undermine the usefulness of the information). The subsequent reviews that involve the use of this research map and that focus on intervention effectiveness, should ensure that quality appraisal is undertaken before making recommendations with regard to policy and practice.

Whilst the design of our framework may have limitations (other approaches may have been possible)—the design of our framework was led by the stakeholders. We used a framework that they were familiar with and is used by major intergenerational organisations—we were keen to use a framework that made sense to the people who we hoped would use the map. The level of engagement in an intervention was also seen to be a key driver for successful interventions and is also an indicator of the potential resource level required for implementation which may be helpful for some users. We felt that using the aims of an intervention would have been difficult to capture in the space of a map and would have been complex as interventions may have more than one aim. This might have made the map more difficult for users to access.

6.3.2 | Stakeholder engagement throughout the EGM process

We liaised with our stakeholders to confirm the details of the protocol before submitting this to Campbell. We were unable to meet with our stakeholders in person and conducted our first meeting online in one large group. At this meeting it was decided that subsequent meetings would be better conducted over two events within the same week to enable some flexibility in attendance and to ensure the meeting could be better facilitated for all attendees. At any point if any stakeholders could not attend the planned meetings they were given the opportunities to have one-to-one meetings with one of the project team or to share their thoughts and feedback over email. Stakeholders were also consulted about the structure of the EGM and how best to capture the outcomes they thought were important as well as the outcomes actively reported in the research. Two stakeholders have not engaged with the project so far but we hope to reconnect with them in the next stages. Details of the two meetings are in Table 5.

TABLE 5 Stakeholder engagement.

Meeting	No. of attendees	Content	Impact on the EGM/research
Stakeholder meeting 1	26 July 2021 (20 researchers, providers, commissioners, third sector and public perspectives represented) Individual meetings arranged where possible/necessary	- Introduction to the project (Jo Thompson Coon) - What are intergenerational activities? (Ellie Robinson-Carter) - What is an evidence gap map? (Fiona Campbell) - Small group discussion in break out rooms to answer (using Jamboard): Q1: What are intergenerational activities? Do you know of any? What has been your experience of them? Q2: What are the potential positive and negative outcomes that can come from intergenerational activities and what do you feel should be measured?	Discussion and stakeholder contributions to the jamboard: - Enabled the research team to understand what type of intergenerational interventions there are and are likely to be identified in research. - Helped to inform the EGM about the outcomes that were important to capture and incorporate in the framework.
Stakeholder meeting 2	Held over two meetings: 27 Sept (17 researchers, providers, commissioners and third sector perspectives represented) 28 Sept (16 researchers, commissioners, third sector and public perspectives represented)	- Welcome and Introductions (Jo Thompson Coon) - Project update (Rebecca Whear and Morwenna Rogers) – numbers of screening and coding, initial map - Purpose of meeting (Rebecca Whear) – share what we have done so far, share map, explore it and think about the kinds of questions it raises but particularly thinking about research questions for the two reviews that we will be conducting as a result of this mapping exercise. - Present the map (Fiona Campbell) – - Any questions about the map? - Discuss potential questions for next reviews	Discussion and stakeholder contributions helped to: - Understand how the EGM was interpreted and how its presentation could be improved - Helped to understand what the most useful next steps would be - Helped to determine the most relevant research questions for the second stage of the project

7 | AUTHORS' CONCLUSIONS

7.1 | Implications for research, practice and/or policy

Based on the research identified in this EGM the implications for research are:

- A need to explore gaps in terms of promising interventions not yet formally evaluated.
- Further primary research needs to build on the evidence for existing interventions exploring a more consistent set of outcomes relevant to both generations engaged in the intervention. This should include the wider impact of the intervention on their families and/or carers and the wider community.
- More primary research is needed on mental health and the mental wellbeing of children and young people, and also loneliness and social isolation in both generations.
- Further primary research should also focus on issues with regard to intervention implementation and sustainability including economic outcomes so that policy makers and commissioners as well as service providers can make better informed decisions as to what intervention might work well and be sustainable for the community with which they are working.
- Further research needs to be conducted on Level 7 type interventions in which the interactions between the generations are built into the community and part of every-day communication, interaction and general living, with the potential thereby for demonstrating lasting positive impacts for everyone involved. These interventions could potentially be more costly and therefore decision-makers need to be confident about the individual, social, economic and community benefits (as well as costs).

Implications for policy are:

- Uncertain in many circumstances because much of the available research does not currently tell us what the impact of the interventions are on both generations (i.e., where there may be positive outcomes for one generation there is a need to be mindful of the outcomes experienced by the other generation). Research that explores the outcomes of an intervention for only one generation need to be further explored before being implemented.
- The research on this topic is gradually increasing, and systematic reviews will be important to determine how and why interventions

are or are not beneficial. However, the primary research area needs to build more cohesively so that the findings can be comparable and avoid research waste.

- The EGM presented here will nevertheless be a useful resource for decision-makers allowing them to explore the evidence with regard to the different interventions that may be relevant to their population needs and the settings or resources available to them.

CONTRIBUTIONS OF AUTHORS

Content: ERC is a socially engaged creative practitioner and consultant based in Plymouth, founder of The Photobook Project and Project Manager at The Sensory Trust where she works on the dementia and intergenerational project Creative Spaces. This project uses nature and outdoor spaces to encourage older people with dementia to become more active, build social networks and foster independence. Previously she founded the multi-award winning Penryn Memory Café and led a memory café in York for 2 years whilst at University. She has recently completed the International Certificate in Intergenerational Practice provided by Generations Working Together and the University of Granada. SC is Commissioning Manager at NHS Kernow Clinical Commissioning Group and has an interest in the role of intergenerational programmes and activities in health and social care. RS is an advanced public health specialist at Cornwall Council with an interest in the role of intergenerational programmes and activities in health and social care specifically in relation to the mental health of older adults. JB is an expert in the mental and social wellbeing of children and young people and also has expertise in evidence synthesis methodology.

EGM methods: JTC is an expert in evidence synthesis and health policy research. She is co-chair and editor of the Ageing Group of the Campbell Library and co-director of the Cochrane Campbell Global Ageing Partnership. RW is an expert in evidence synthesis methods. FC is editor of the Children and Adolescent Group of the Campbell Collaboration. She has over 20 years of experience in evidence synthesis and leads a short course in scoping, mapping and EGM reviews.

Information retrieval: MR is an information specialist with experience in health services research, methods editor for the Ageing Group of the Campbell Library and a member of the Campbell Information Retrieval Methods Group. AS is a Senior Information Specialist, with extensive experience of literature searching and information management for systematic reviews and other types of evidence syntheses on a wide range of topics, including integrated care, art therapy and quality of life. AS is the joint lead of a module on systematically reviewing the research literature for postgraduate students, and the joint author of the textbook *Systematic Approaches to a Successful Literature Review*, 2nd Edition published by Sage in 2016. Anthea is also the Reviews Editor for *Health Information and Libraries Journal*.

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DECLARATIONS OF INTEREST

ERC, members of our advisory group and members of the Only Connect steering group are involved in the delivery of intergenerational activities and programmes.

PLANS FOR UPDATING THE EGM

Once completed the evidence gap map will be updated as resources permit.

DIFFERENCES BETWEEN PROTOCOL AND REVIEW

Due to the substantial amount of research literature found we did not include news items describing intergenerational activities and programmes if they reported innovative interventions not otherwise represented within the evidence base (as in the protocol). However, this will be conducted in the subsequent reviews directly related to this EGM.

Due to the amount of research literature found we limited our additional searches (forwards and backwards citation chasing) as follows: we carried out backward citation chasing on the included studies within identified systematic reviews specifically looking for RCTs and systematic reviews not already included in the EGM; we did not check the citations of older key papers (forward citation chasing); we identified one key journal the Journal of Intergenerational Relationships and hand-search the contents; we did not conduct the horizon scanning process (we will search Nexus for relevant international news articles about intergenerational practices and Google for relevant reports, blogs, news articles and links to other relevant organisations) mentioned in the protocol but expect to conduct that in subsequent reviews.

In addition to the filters mentioned in the protocol additional amendments were made to include the following:

- Characteristics of the participants: Childhood Adversity, Age category, Disability (physical heath difficulties), Mental health difficulties, Low socioeconomic status, Minority groups, Social isolation, Unemployed, Educational needs, Cognitive impairment.
- Contextual factors: Setting and Country.
- Study design factors.
- Focus of the interventions: Education, Art and craft, Music, Interaction, Cooking, Dance, Drama, Environmental activities, Exercise, Gardening, History, IT, Language, Letter writing, Literature, Living together, Maths, Playing games, Professional education, Reading, Reminiscence, Science activities, Sharing meals, Sharing perspectives (of being and older person/a child/young person), Story telling, Trips and excursions and Other.

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Internal sources

- No sources of support provided

External sources

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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Paper G

Histopathology

Histopathology 2022, **81**, 420–425. DOI: 10.1111/his.14648

Editorial

Evidence-levels in pathology for informing the *WHO classification of tumours*

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Introduction

The *World Health Organization classification of tumours* (WCT), published as the *WHO Blue Books* (Figure 1) and the *Blue Books online*, are essential resources for pathologists across the globe and provide the standards for cancer diagnosis, research, treatment and prognosis. The classification describes the characteristics of each tumour type, defining the aetiology, pathogenesis, epidemiology, clinical features, macroscopic appearances, histology, cytology, molecular pathology, essential and desirable diagnostic features, staging, prognostic factors and predictive biomarkers. Revision of the WCT in each edition requires extensive review of the scientific literature so that decisions made by the editorial board are informed by the best available evidence. Systematic review (SR) is a methodology used in evidence-based medicine (EBM) to identify and rigorously summarize evidence. Clearly, it is not always possible to use this comprehensive approach due to the resources required and the large number of questions to be addressed in revising the WCT. The current approach therefore relies largely upon subject experts performing limited searches of published literature according to their individually perceived need.[1] However, such potentially biased, non-systematic searches may directly affect the classification, and hence the diagnosis and management of cancer patients worldwide. To minimize the risk of including or excluding biased information, the evidence presented is weighed and decisions made by an editorial board consisting of standing members with broad expertise and expert members selected for their specific knowledge.[1] However, there is a lack of consensus about what constitutes good evidence in pathology.

Challenges in assessing evidence for tumour classification

The WCT contributors are mainly practising pathologists, who face important challenges when assessing the evidence to agree on definitions and core criteria for each tumour type. They have to navigate the multidimensional nature of tumour classification and a large volume of heterogeneous evidence from fields as diverse as genetics, histopathology, epidemiology, radiology and others. For each topic to be assessed, a vast number of publications providing potentially relevant information needs to be considered. The research evidence comes from multiple disciplines and may therefore use very different research methodologies reporting on different characteristics of a given tumour type. On top of this, records of the publications may be dispersed through different databases, making the task of locating all relevant evidence very difficult. The editorial process has tight time constraints and it may be uncertain whether contributing authors have conducted an exhaustive search of the literature or whether potentially skewed decisions may be made without considering the most comprehensive, relevant and latest evidence. Inevitably, the expertise of participating experts is varied and may not always include a broad knowledge of research and evidence synthesis methodology.

Evidence-based medicine principles

Pathology has been slower than some areas of medicine to incorporate the principles of EBM. EBM uses systematic review methods to ensure a rigorous, transparent and exhaustive approach to finding and integrating the best available evidence with clinical expertise for decisions related to health-care (Table 1). EBM is well established in medicine and has demonstrated its utility and effectiveness in improving health outcomes in numerous medical fields.[2] While it is acknowledged by the WCT leadership that there is a need for EBM approaches to inform the WCT process, the incorporation of such methods has been gradual. This hesitancy may be due in part to a lack of training and expertise in SR methodology.[1,3–5] It can be laborious and time-consuming to complete such training, particularly for high-level experts engaged in clinical practice, teaching, management and research with editorial work for the WCT. In addition, best practice guidelines such as Cochrane[3] and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[4,5] are closely aligned with meta-analytical reviews for medical interventions, the methods for which are not necessarily appropriate to pathology.[6,7] Similarly, the traditional hierarchy of evidence levels used in EBM (see below), with randomized controlled trials (RCT) preferred, and case reports and opinion articles considered to be low-level evidence, does not adapt well to the reality of research published in the field of histopathology, and has therefore not been enthusiastically embraced by expert groups. Revision of

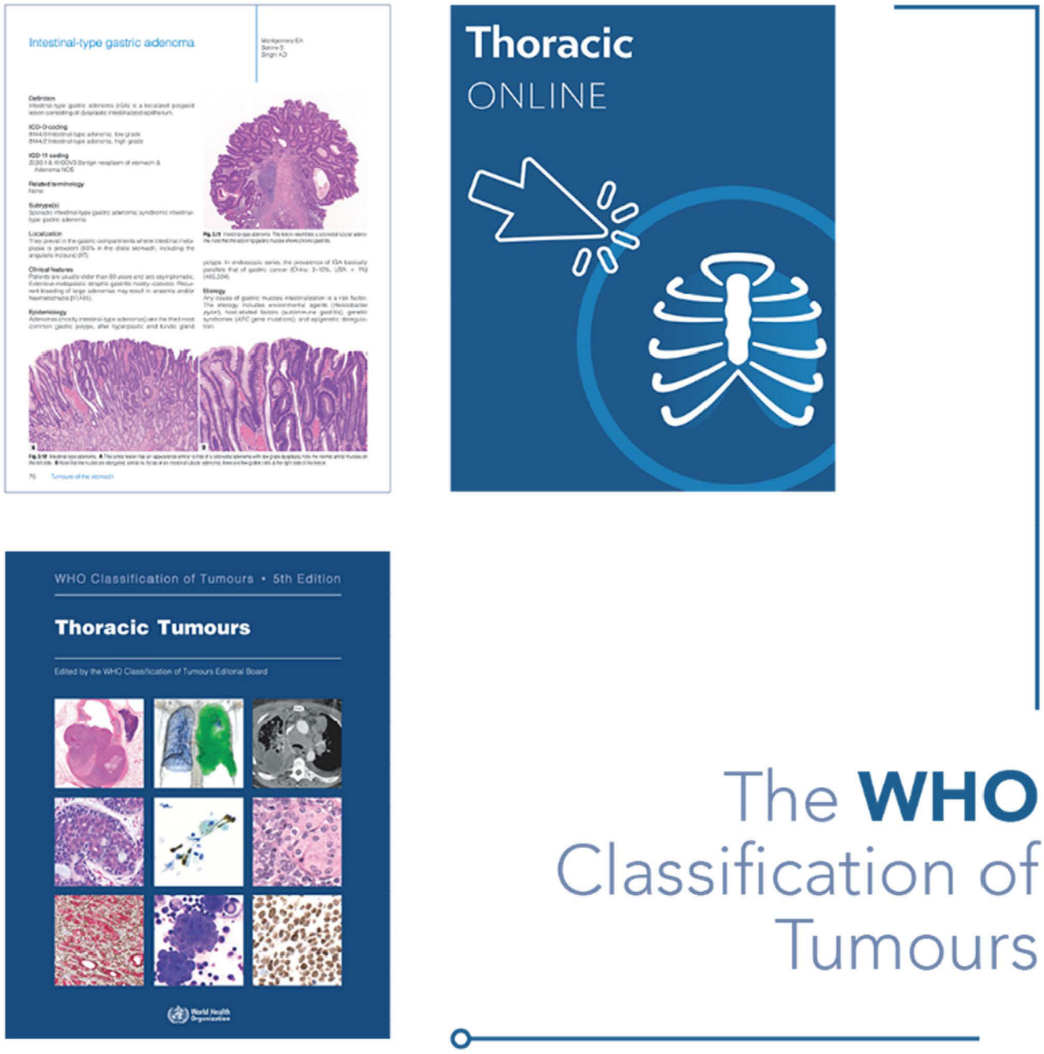

Figure 1. The *WHO classification of tumours* published as the *WHO Blue Books* and BB Online.

this system tailored for pathology may help the uptake of EBM in pathology.

The hierarchy of evidence levels and its recent evolution

The hierarchical system of classifying evidence, known as the levels of evidence and often presented as an evidence pyramid[8] (Figure 2), is a cornerstone of EBM and decision-makers are encouraged to prefer the highest level of evidence to inform health related decisions.[8] This principle highlights that not all evidence is equal and describes study designs (Table 2) with a higher risk of including bias at the bottom (case reports and series), followed by study designs with moderate risk of bias (first case–control, cohort studies in the middle and RCTs) and finally, at the very top, SR and meta-analysis, as both include an

Table 1. Definitions of evidence-based medicine and practice

Definition of evidence-based (Cambridge Dictionary):
Supported by a large amount of scientific research.
Definition of evidence-based medicine (EBM; definition by Dave Sackett in BMJ, 1996):
EBM is the conscientious, explicit, and judicious use of current best evidence in making decisions about the care of individual patients.
Definition of evidence-based practice:
The term evidence-based practice has developed from the term evidence-based medicine (EBM) and refers to an approach to health wherein professionals use the best evidence possible, i.e. the most appropriate information available, to make relevant decisions.

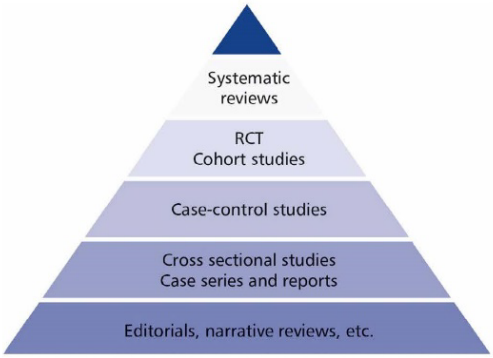

Figure 2. Traditional evidence hierarchy based on internal validity of the different study designs.

Table 2. Overview of epidemiological study designs [Colour table can be viewed at wileyonlinelibrary.com]

Experimental studies	
Clinical (or community) trials *Study design in which the investigator determines through a controlled process the exposure for each individual (clinical trial) or community (community trial) and follows the participants up to analyse effects.*	Analytic study methods
Observational studies	
Cohort studies *Study design in which the investigator observes exposed and unexposed participants, and follow them up to see if they develop the disease of interest. In a cohort study, the investigator observes and does not determine the participants' exposure status as in the experimental design. There are prospective and retrospective cohort studies, depending if exposure and the outcomes are being followed up or have already occurred.*	Analytic study methods
Case-control studies *Study design in which the investigator selects participants with disease (cases) and comparisons without disease (controls) to compare previous exposures between groups. An appropriate control group should be comparable to the case group in most respects to provide a correct estimate of the exposure.*	Analytic study methods
Cross sectional studies *Study design in which a population sample is selected and their exposures and health outcomes are measured at one moment in time without follow-up. The cross-sectional study aims to describe the presence (prevalence) of an outcome at that point of time without regard to duration.*	Analytic study methods
Case series *A report of patient cases containing detailed demographic (age, gender, ethnic origin) and clinical information (symptomatology, diagnosis, treatment, response to treatment, and follow-up) about the individual patients. There are prospective and retrospective case series, depending if they are followed up or collecting information on cases that already occurred.*	Descriptive study methods

evaluation of the risk of bias and/or methodological quality in their methods and usually comprise the highest quality studies available. This description is appropriate in most scenarios; however, it is heavily focused upon interventional studies. Over time, the evidence pyramid has been adapted to include more diverse sources of evidence.[9,10] The Grading of Recommendations Assessment, Development and Evaluation (GRADE) working group developed a framework in which the certainty in evidence was not solely based on study design.[11] In addition to the risk of bias inherent to the study design, GRADE included the assessment of certain methodological limitations that can affect the quality of evidence derived from any study design and recommended in their approach to downgrade quality of evidence if such limitations were detected. The Oxford Centre for Evidence-Based Medicine (OCEBM) also published evidence levels, updated in 2011 (Figure 3) in a table format, with the clear aim to permit the users specifically to focus on answering clinical questions for various contexts.[12] While most ranking schemes focused upon strength of evidence for interventions, the OCEBM system classifies evidence by type of research in prevalence, accuracy of diagnostic tests, prognosis, therapeutic effects, rare harms, common harms and usefulness of (early) screening.[13] This scheme addresses the inflexibility of the evidence pyramid very well and although the Oxford levels of evidence can be applied to pathology, they may appear complex to those untrained or inexperienced in EBM.

Evidence levels for pathology

Application of EBM principles to the field of pathology has been ad hoc, with limited contributions from the pathology field to its development. Marchevsky and colleagues[14–17] have promoted the idea of evidence-based pathology (EBP) in the last decade, and the WCT has been working steadily in recent years to initiate a strong EBP movement.[1,18] Some researchers in the field have recognized that application of the current evidence hierarchies to histopathology poses a problem, as most scientific studies in this speciality are case reports and case series, relegating most histopathology literature to lower-levels of evidence.[15,19] This encourages a negative view of such studies and may lead many to assume that SRs of these types of study would be of limited value. However, literature generated by histopathology has been contributing extensively to the body of medical knowledge for many decades, and even though most studies are observational in nature, with an overwhelming majority of reports of case series, this body of evidence still needs to be assessed systematically.

Oxford Centre for Evidence-Based Medicine 2011 Levels of Evidence

Question	Step 1 (Level 1*)	Step 2 (Level 2*)	Step 3 (Level 3*)	Step 4 (Level 4*)	Step 5 (Level 5)
How common is the problem?	Local and current random sample surveys (or censuses)	Systematic review of surveys that allow matching to local circumstances**	Local non-random sample**	Case-series**	n/a
Is this diagnostic or monitoring test accurate? (Diagnosis)	Systematic review of cross sectional studies with consistently applied reference standard and blinding	Individual cross sectional studies with consistently applied reference standard and blinding	Non-consecutive studies, or studies without consistently applied reference standards**	Case-control studies, or "poor or non-independent reference standard**	Mechanism-based reasoning
What will happen if we do not add a therapy? (Prognosis)	Systematic review of inception cohort studies	Inception cohort studies	Cohort study or control arm of randomized trial*	Case-series or case-control studies, or poor quality prognostic cohort study**	n/a
Does this intervention help? (Treatment Benefits)	Systematic review of randomized trials or *n*-of-1 trials	Randomized trial or observational study with dramatic effect	Non-randomized controlled cohort/follow-up study**	Case-series, case-control studies, or historically controlled studies**	Mechanism-based reasoning
What are the COMMON harms? (Treatment Harms)	Systematic review of randomized trials, systematic review of nested case-control studies, *n*-of-1 trial with the patient you are raising the question about, or observational study with dramatic effect	Individual randomized trial or (exceptionally) observational study with dramatic effect	Non-randomized controlled cohort/follow-up study (post-marketing surveillance) provided there are sufficient numbers to rule out a common harm. (For long-term harms the duration of follow-up must be sufficient.)**	Case-series, case-control, or historically controlled studies**	Mechanism-based reasoning
What are the RARE harms? (Treatment Harms)	Systematic review of randomized trials or *n*-of-1 trial	Randomized trial or (exceptionally) observational study with dramatic effect			
Is this (early detection) test worthwhile? (Screening)	Systematic review of randomized trials	Randomized trial	Non -randomized controlled cohort/follow-up study**	Case-series, case-control, or historically controlled studies**	Mechanism-based reasoning

Figure 3. The Oxford Levels of Evidence. Source: OCEBM Levels of Evidence Working Group.[1] 'The Oxford Levels of Evidence 2'. Oxford Centre for Evidence-Based Medicine. Available at: https://www.cebm.ox.ac.uk/resources/levels-of-evidence/ocebm-levels-of-evidence [Colour figure can be viewed at wileyonlinelibrary.com]

The use of SRs and meta-analyses for these types of evidence would significantly enhance the evidence-base for histopathology. This critical assessment of evidence needs to take into consideration specificities of the field, as most common publishing practices, sources of bias that might be specific to the field, such as the well-known interobserver variation in histopathology, or differences between laboratories in the techniques used. Most importantly, methods need to be developed or adapted for accurate synthesis and evaluation of evidence provided by case series in diagnostic and prognostic research.

Histopathology has long been considered the 'gold standard' for medical diagnosis and the contribution of pathology research to medicine should be reflected in EBM. There is a need to provide the methods and training to integrate evidence into the WCT. A necessary first step is to consider how to evaluate evidence in pathology. The best chance of this new consensus being developed and widely adopted would be if the change comes primarily from pathologists. Basic training in EBM and epidemiology, emphasizing the recognition of evidence levels and the common biases in pathology research, is also needed. The WCT programme and the International Collaboration for Cancer Classification and Research (IC3R) at the International Agency for Research on Cancer (IARC) have set themselves the challenge of conducting a Delphi study to define levels of evidence suited to pathology as part of the WCT EVI MAP project recently funded by the European Union. This will be carried out using an adapted e-Delphi method[20,21] to reach consensus among a representative panel of experts in the field and potential end-users. This expert consensus on a set of generic core items/criteria for the assessment of evidence-levels in pathology related research will then be used to define a new, adapted evidence level hierarchy for the field and propose a set of reporting recommendations for future research. The aim is not only to raise awareness in the field, but also to invite discussion of the potential challenges and solutions proposed to ensure the development of a robust EBP movement that moves the WCT towards a more evidence-based approach. As a secondary product of the Delphi consensus study, the IC3R will draft a set of reporting recommendations in pathology research that could guide researchers and editors to improve the level of evidence generated in the field.

Application of the new EBP framework

The development of new levels of evidence for pathology would help histopathologists in practice (as well as WCT contributors) to recognize and evaluate good evidence in histopathology. It is hoped that this will also promote and encourage more systematic reviews in histopathology, as well as encourage more

histopathologists to seek training in EBM and clinical epidemiology.

For future classifications, the WCT and IC3R will pursue strategies to assist in identifying relevant evidence and existing information gaps, as well as recognizing pockets of low-level evidence showing where research is needed. The WCT EVI MAP project will adapt the recently developed evidence gap map (EGM) methodology and software, pioneered in social sciences and apply this to biomedicine, producing EGMs for the whole WCT. These EGMs will provide a visual, easy-to-interpret summary of what evidence has been published for different tumour types for use by WCT decision-makers, research commissioners, researchers and practising histopathologists. It will also highlight where evidence gaps lie in order to inform future research priorities. To overcome the obvious challenges to such a broad project, adaptative strategies will be applied, such as using recent advances in technologies, following a strict multidisciplinary approach, and integrating this project into WCT strategic planning for the 6th edition. The WCT EVI MAP will represent a ground-breaking advance for the WCT.

Conclusions

Integration of EBM into clinical practice has been shown to have a positive impact in other medical specialities. When the barriers discussed here have been overcome, the EBP approach is likely to gain popularity in histopathology. We are confident that this practice will increase the quality of research and rigour of decisions feeding into tumour classification, addressing critical questions and identifying research gaps to direct future research. Such an approach will help to maintain the reliability of tumour classification and will provide solutions to challenges such as the rapid growth in the number of scientific publications, and the need to manage new types of information, including evidence from genetic and big data studies.

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Conflicts of interest

The authors have no conflicts of interest to declare.

Author contribution

BII, PHT conceived the paper and structured the idea. FC and RC provided methodological support. BII and PHT developed a first draft. RC, IAC and FC revised and improved it. All authors contributed significantly to the final manuscript.

Data availability statement

Data sharing is not applicable to this article, as no new data were created or analysed in this study. B I Indave, R Colling, F Campbell, P H Tan & I A Cree.

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