Methods for identifying and displaying gaps in health research

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Résumé

Il a été estimé qu'environ 85 % des recherches en santé ne sont pas utilisables. Plus de 50 % des recherches ne sont pas du tout publiées. Parmi les travaux de recherche publiés, la moitié n'est pas utilisable en pratique car des éléments importants ne sont pas rapportés. Enfin, parmi les 25 % restants, la moitié encore a des faiblesses dans sa planification. La planification d'une étude centrée sur une mauvaise question est une cause fréquente du gaspillage de la recherche (*waste in research*). Par conséquent, il est important d'utiliser les recherches publiées et non publiées, achevées et en cours, pour évaluer si un manque ou une lacune dans les recherches existantes justifie de nouvelles recherches. Une telle approche peut aussi éclairer la conception, la conduite et le rapport des recherches ultérieures.

La thématique des méthodes d'identification et de représentation des lacunes dans la recherche en santé n'est pas encore bien établie. Il n'existe ni une définition standardisée du terme « lacunes en recherche » (*research gaps*), ni une méthode standardisée pour identifier ces lacunes. De plus, faute de définition claire, il n'y a pas de consensus sur ce qui constitue les meilleures approches méthodologiques pour identifier ces lacunes, déterminer les priorités de recherche et représenter (par exemple graphiquement) ces lacunes ou priorités en recherche.

Les objectifs de cette thèse de doctorat sont : 1) identifier les différentes définitions rapportées pour le terme « lacunes en recherche » et décrire les méthodes utilisées pour identifier, hiérarchiser et représenter les lacunes dans la recherche en santé ; 2) explorer les points de vue et les expériences des principaux intervenants de la recherche sur la définition, l'identification et la représentation des lacunes dans la recherche en santé ; et 3) élaborer un guide méthodologique pour des travaux visant à identifier et représenter les lacunes dans la recherche en santé.

Dans un premier projet, j'ai effectué une revue panoramique (*scoping review*) pour identifier les définitions rapportées des lacunes en recherche et les méthodes utilisées pour identifier, hiérarchiser et représenter ces lacunes dans la recherche en santé. Les méthodes les plus fréquentes identifiées par la revue panoramique visaient à identifier des lacunes et concernaient des recherches secondaires (recherches réutilisant des données d'autres recherches). La majorité des études correspondaient à des travaux de synthèse de données existantes (80/116 articles, 69 %), en particulier des revues systématiques et des revues panoramiques (58/80 articles, 73 %). Parmi les études visant à prioriser la recherche, les méthodes les plus fréquentes étaient une combinaison de recherche primaire et secondaire (24 articles, 49 %), suivies de recherches secondaires (8 articles, 16 %). Enfin, 37 % des articles décrivaient des méthodes pour représenter ou cartographier les lacunes ou les priorités de la recherche en santé.

Dans cette revue panoramique, j'ai dans un premier temps identifié une liste complète d'articles scientifiques portant sur la définition, l'identification, la hiérarchisation ou la représentation des lacunes en recherche en santé. Ces articles variaient considérablement selon les domaines de recherche. En ce qui concerne les termes et définitions utilisés pour décrire les lacunes de la recherche, j'ai identifié un total de douze définitions différentes en utilisant le titre des articles, mais certaines similitudes existaient dans la description de chacune de ces définitions. Trois thèmes transversaux ont été identifiés : les définitions relatives aux 1) informations manquantes, 2) informations inadéquates et 3) informations insuffisantes. Cette analyse montre que, bien que le terme « lacunes en recherche » soit couramment utilisé dans la recherche en santé, sa signification peut différer selon les travaux.

Les méthodes utilisées pour identifier les lacunes en recherche étaient présentées plus clairement que les termes liés aux lacunes en recherche et les définitions utilisées pour décrire ces lacunes. Les méthodes identifiées ont été regroupées en trois grandes catégories : 1) recherche primaire (enquêtes), 2) secondaire (synthèse de l'évidence existante) et 3) méthodes combinant recherche primaire et secondaire. L'approche la plus fréquemment utilisée était une recherche secondaire, suivie par des méthodes combinées primaires et secondaires.

Parmi les méthodes de recherche secondaires, la synthèse des connaissances était couramment utilisée. La synthèse des connaissances se définit comme une approche scientifique efficace pour identifier et résumer les preuves. Elle permet d'évaluer le caractère généralisable et la cohérence des résultats de recherche et les incohérences des données à explorer. Le but de cette approche est de résumer toutes les études pertinentes portant sur une question spécifique, d'améliorer la compréhension des incohérences des différents résultats et d'identifier les lacunes dans les données de recherche, pour ensuite définir les futurs programmes de recherche. De plus, dans le cadre de la synthèse des connaissances, les revues panoramiques (*scoping reviews*) sont l'une des seules méthodes utilisées pour identifier les lacunes en recherche qui incluent explicitement l'identification des lacunes en recherche dans leur objectif.

J'ai ensuite classé les méthodes utilisées pour identifier les lacunes en recherche. Les méthodes les plus fréquentes de la revue visaient à identifier les lacunes (à la fois pour les identifier et les prioriser) et impliquaient des recherches secondaires, y compris la synthèse des connaissances (80/116 articles, 69 %), en particulier des revues systématiques et des revues de cadrage (58/80, 73 %). Dans l'ensemble, 24/116 (21 %) articles décrivaient l'utilisation de la recherche primaire

et secondaire et 12/116 (10 %) uniquement de la recherche primaire. J'ai trouvé 7 méthodes spécifiques décrites pour identifier les lacunes en recherche.

Un mélange de recherche primaire et secondaire était le plus souvent utilisé pour déterminer les priorités de recherche (par exemple, *James Lind Alliance [JLA] - Priority Setting Partnerships methods*). Ces méthodes impliquent la participation de patients, de soignants et de professionnels de santé et des soins sociaux pour identifier des questions de recherche, puis les hiérarchiser en utilisant une combinaison de recherche primaire et secondaire. La principale approche de recherche primaire pour déterminer des priorités de recherche était l'enquête Delphi, qui est une approche pratique et efficace pour obtenir les opinions d'un grand nombre d'experts pour identifier les domaines prioritaires potentiels pour la recherche.

Pour représenter les lacunes en recherche, la moitié des méthodes utilisaient encore des méthodes traditionnelles pour présenter les résultats (par exemple, un tableau récapitulatif et des diagrammes), et l'autre moitié utilisaient des méthodes plus avancées (par exemple, *graphiques en arbre, ou graphiques en bulles*). Ces méthodes non traditionnelles font davantage usage de couleurs et de formes pour présenter les lacunes ou les priorités de la recherche. Par exemple, les graphiques en bulles utilisent différentes formes, tailles et couleurs pour afficher les informations et peuvent être utilisés pour présenter jusqu'à trois variables différentes dans un même diagramme. Ces caractéristiques pourraient être explorées plus avant pour déterminer la méthode appropriée à utiliser avec des méthodes spécifiées pour identifier les lacunes en recherche ou déterminer les priorités de recherche.

Le deuxième projet est une étude qualitative à partir d'entretiens semi-structurés d'intervenants clés. Ces entretiens avaient pour but d'explorer leur niveau de connaissance, leurs perceptions et expériences avec la définition des lacunes en recherche, et de caractériser les méthodes et pratiques utilisées pour l'identification et la communication des lacunes dans la recherche en santé. Les résultats ont permis de caractériser ce que les participants considéraient comme des lacunes dans la recherche en santé : les termes allaient de « manque d'information », « information inadéquate », « information insuffisante » à « défaut de qualité des preuves » et « incertitude du traitement ». L'étude a montré les expériences des participants et leurs perceptions des différentes méthodologies de recherche utilisées (c'est-à-dire primaire, secondaire, ou une combinaison des deux). La variété des méthodes citées reflétait le large éventail de méthodes actuellement utilisées, sans consensus ni orientation claire. Les chercheurs participants ont également exprimé une difficulté à identifier systématiquement les lacunes en recherche. En ce qui concerne la représentation des lacunes en recherche, les participants ont souligné l'importance de la visualisation des données et la difficulté pour les participants-chercheurs à trouver le bon outil permettant de présenter au mieux les résultats de la recherche.

Cette étude démontre que l'utilisation des deux méthodes primaires et secondaires (par exemple, la méthode développée par la *James Lind Alliance*) pour identifier les lacunes était la méthode la plus robuste. Le principal avantage connu de cette méthode est l'identification de ces lacunes (incertitudes sur le traitement) en impliquant différentes parties prenantes, et notamment la confirmation et la priorisation des lacunes identifiées en impliquant des patients et des intervenants non professionnels experts du domaine (identifiés comme « le public » dans la suite). Les inconvénients principaux sont la nécessité d'une main-d'œuvre abondante (une équipe de différents spécialistes) et les coûts associés (en tenant compte du support administratif, des

salles de réunion, de la restauration…) par rapport aux méthodes primaires (enquête) ou secondaires (synthèse des preuves).

Concernant la visualisation des données, les participants ont principalement exprimé son importance dans la communication de la recherche. Cependant, aucun format spécifique pour présenter les lacunes n'a été mentionné. De plus, différents groupes d'intervenants, en particulier les chercheurs, souhaitent utiliser la visualisation des données lors de la communication de la recherche, bien que nous n'ayons trouvé que peu d'exemples d'expériences de développement et d'utilisation de celle-ci. Les participants ont principalement exprimé la difficulté de trouver de bons outils à utiliser pour présenter les résultats de la recherche.

Enfin, bien que les articles scientifiques mentionnent souvent l'existence de lacunes de recherche dans les études, peu de participants ont été en mesure de définir les lacunes de la recherche, sauf s'ils les contextualisent dans une étude ou un domaine spécifique, ou font référence à des méthodes d'identification. Pour mieux comprendre comment réfléchir aux lacunes en recherche en santé et y remédier de manière adéquate, nous avons donc mis en évidence trois éléments clés à prendre en compte : 1) définir clairement les lacunes en recherche fournit un contexte permettant de mieux comprendre quelles sont les lacunes et leurs causes ; 2) une définition claire des lacunes en recherche peut éclairer le choix des méthodes utilisées pour identifier ces lacunes, de la même manière qu'une question de recherche claire peut éclairer la méthodologie de la recherche ; et 3) lors de l'adoption des méthodes les plus appropriées pour identifier les lacunes de la recherche, il est important de trouver la bonne visualisation pour les communiquer efficacement. Enfin, la participation du public, le cas échéant, est nécessaire pour vérifier que les lacunes sont importantes pour lui.

Les derniers résultats de l'étude qualitative ont révélé que diverses méthodes pour identifier les lacunes peuvent être adoptées (c'est-à-dire primaire, secondaire et à la fois primaire et secondaire). De toutes les méthodes pour identifier les lacunes, les recherches secondaires, considérées comme la méthode de référence, sont les plus couramment utilisées, en particulier les revues systématiques. Ce type d'étude aborde une question très ciblée liée aux preuves existantes et présente donc des difficultés pour identifier explicitement les lacunes en recherche dans un domaine général. D'autres méthodes de recherches secondaires rapportées étaient des compilations de revues (*overview of reviews*), de revues panoramiques (*scoping reviews*) et de cartographies des preuves (*evidence mapping*). Les compilations de revues se concentrent sur un domaine beaucoup plus large, rassemblent les preuves issues de multiples revues dans un seul document accessible et utilisable. Elles mettent en évidence d'autres revues dans le domaine de sujet spécifié. Étant donné les besoins en ressources pour des examens de preuves formelles, la priorisation du sujet est nécessaire pour mieux allouer les ressources aux domaines jugés les plus pertinents pour le système de santé. Quel que soit le sujet, le processus de priorisation dépendra probablement des parties prenantes. Les priorités de la synthèse des données probantes varieront en fonction de la mission du système de santé et des besoins locaux.

En résumé, l'utilisation des méthodes de recherche primaires et secondaires est la plus robuste car elle implique la participation de patients, de soignants, et de travailleurs sociaux pour l'identification des questions de recherche, puis leur hiérarchisation.

Dans le troisième projet, je me suis concentrée sur le développement d'un guide méthodologique, visant à identifier systématiquement et rapporter les lacunes en recherche, pour donner une vision

plus claire de l'état des connaissances et du niveau de preuve. Ce guide méthodologique a été construit à partir des résultats de la revue panoramique et de l'étude qualitative. Mon premier résultat est que, pour identifier et combler les lacunes en recherche dans un domaine thématique, l'étape la plus importante est d'abord de clairement définir ce que l'on entend par une lacune de recherche, et de le rapporter de façon explicite. Je recommande d'adopter une définition existante qui décrit la nature de ces lacunes. En décrire la nature peut impliquer différents éléments, comme :

1) Énoncer l'ampleur du déficit de recherche (c'est-à-dire spécifique ou étendu)

2) Définir clairement le déficit de recherche

3) Préciser la cause de ces lacunes de recherche

En combinant les résultats de la revue panoramique et de l'étude qualitative, une liste de termes clés liés à l'identification de lacunes en recherche a été élaborée, ainsi qu'une liste des méthodes principales pour identifier et représenter ces lacunes en recherche. Ces résultats ont ensuite été combinés et utilisés pour développer un guide méthodologique pour les études visant à identifier des lacunes dans la recherche en santé. Six étapes sont à considérer : 1) spécifier le domaine thématique et / ou la question de recherche ; 2) cartographier et énoncer clairement les lacunes en recherche existantes, et prendre en considération les avis d'experts ; 3) identifier les lacunes en recherche ; 4) décrire clairement les lacunes en recherche identifiées ; 5) caractériser ces lacunes en recherche ; et 6) représenter (par exemple graphiquement) ces lacunes en recherche. Pour être en mesure de déterminer la faisabilité et l'utilisabilité de ce guide méthodologique en pratique, l'importance, la pertinence et l'applicabilité de chaque étape proposée doivent être discutées entre les différents intervenants, puis mises en œuvre et évaluées.

Le guide méthodologique développé devra être évalué pour déterminer son applicabilité et sa facilité de mise en œuvre dans la recherche en santé. Il sera ensuite possible d'envisager des adaptations pour son extension à d'autres domaines.

Abstract

Statistics on the use of the available health research is troubling: an estimated 85% is wasted and more than 50% never gets to be published. From the published research, 50% is not usable in practice because items are missing. In addition, among the remaining half of the published research, 50% contains design flaws. Planning a study focusing on the wrong research question is a frequent cause of waste in research. Hence, published, unpublished, completed, and on-going research should be used to assess whether research gaps justify new research that can inform the design, conduct and reporting of further research.

The overall topic area of methods to identify and display gaps in health research is still not well established; also, there is no standard definition for the term "research gaps" nor standardized methods to identify research gaps. Furthermore, with a lack of a clear definition, consensus is lacking on what constitutes the best methodological approaches to identify research gaps, determine research priorities and display research gaps or priorities. Therefore, with an aim to improve understanding research gaps as a whole, and specifically defining, identifying and displaying research gaps, I undertook this PhD project. With the specific objectives to 1) identify different definitions reported for the term "research gap" and describe the methods used to identify, prioritize and display gaps in health research; 2) explore key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research; and 3) develop methodological guidance for identifying and displaying gaps in health research.

In the first project, I conducted a scoping review to map reported definitions of research gaps and methods to identify, prioritize and display gaps in health research. The study provided an overview of different definitions and methods used to identify, prioritize, and display gaps or priorities in health research. The most frequent methods in the review aimed at gap identification

and involved secondary research, which included evidence synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80 articles, 73%). Among studies aimed at research prioritization, the most frequent methods were combined primary and secondary research, accounting for 24 (49%) articles, followed by secondary methods, 8 (16%) articles. Finally, 37% articles described methods for displaying gaps and/or priorities in health research.

The scoping review revealed a comprehensive list of scientific articles that reported the definition, identification and/or prioritization, and display of research gaps. These articles related to defining research gaps and methods used to identify, prioritize, and display research gaps varied significantly across topic areas in health research. For the studies that reported on terms and definitions used to describe research gaps, I identified 12 different definitions based on the report title, yet with some similarities in the description provided for each. Three similar cross-cutting themes were identified: definitions related to missing information, inadequate information, and insufficient information. This analysis shows that despite the term "research gap" being commonly used in health research, its meaning can differ. Therefore, having a clear term and description supports the clarity in reporting health research and subsequently communication of what research gaps exist.

The methods to identify research gaps were more clearly presented than were the terms related to research gaps and the definitions used to describe the research gaps. The methods identified were grouped in three main categories: primary, secondary, and combined primary and secondary research. The most frequently used method was secondary methods, followed by both primary and secondary, then finally primary methods.

Among the secondary methods, knowledge synthesis was commonly used. Knowledge synthesis is an efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored. The purpose of knowledge synthesis is to summarize all pertinent studies on a specific question, improve the understanding of inconsistencies in diverse evidence and identify gaps in research evidence to define future research agendas. Moreover, within knowledge synthesis, the scoping review is one of the only methods used to identify research gaps that explicitly included identification of research gaps as part of the purpose of the method.

I then classified the methods used to identify research gaps. The most frequent methods in the review were aimed at gap identification (including both identification and prioritization) and involved secondary research, including knowledge synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80, 73%). Overall, 24/116 (21%) articles described the use of both primary and secondary research and 12/116 (10%) only primary research. I found 7 specific methods for identifying research gaps that are described, along with the purpose of the method and further elaborated in the thesis.

A mix of primary and secondary research was most frequently used to determine research prioritization, namely, priority setting (e.g., James Lind Alliance [JLA]-Priority Setting Partnerships methods). These methods involve the participation of patients, caregivers and healthcare and social-care professionals in identifying research questions, then prioritizing them by using a combination of primary and secondary research. The main method for determining research prioritization with primary research was the Delphi survey, which is a practical and

productive approach to obtaining opinions from a wide number of relevant experts for identifying potential priority topic areas for research.

To display research gaps, half of the methods still used traditional ways to present findings (e.g., summary table and bar charts), and the other half used more advanced ways to display information (e.g., tree map charts, radial bar plots and bubble plots). The non-traditional methods used more colors and diagrams to present research gaps or priorities. For example, bubble plots use different shapes, sizes, and colors to display information and can be used to present up to three different variables in one diagram. These characteristics could be further explored to determine the appropriate method to be used along with specified methods to identify research gaps and/or determine research priorities.

The second project was a qualitative study involving semi-structured interviews of key informants to investigate their knowledge, perceptions, and experiences with defining research gaps and characterizing methods/practices used for identifying and displaying gaps in health research. The results provided evidence on what participants reported as gaps in health research: the terms ranged from “lack of information”, “inadequate information”, “insufficient information”, “quality of evidence” and “treatment uncertainty”. The study showed detailed participants’ experiences with and perceptions of different research methodologies used (i.e., primary, secondary, both). The variety of identified methods reflected the state of the field in the sense of the wide array of methods currently used with no clear consensus or guidance. Researcher participants also expressed a difficulty in systematically identifying research gaps. With experiences in displaying research gaps, participants expressed the importance of data visualization and the difficulty in researcher-participants finding the right tool to use to present research findings.

Overall the qualitative study found that various methods to identify gaps can be adopted (i.e., primary, secondary and both primary and secondary). Of all the methods to identify gaps, secondary methods were the most commonly used, specifically systematic reviews, considered the gold standard: these address a highly focused question related to the existing evidence and thus present difficulties for explicitly identifying research gaps in a general area. Hence using both primary and secondary methods (e.g., JLA method) was the most robust because it involves the participation of patients, caregivers and health and social care professionals in identifying research questions, then confirming, and prioritizing them by using a combination of primary and secondary methods. The main reported advantage of the JLA method is that it identified gaps (treatment uncertainties), and involved different stakeholders including patients and the public to confirm and prioritize the gaps. The main disadvantages are that it is labor-intensive (requires a team of different specialists) and expensive (administrative support, meeting rooms, catering, among others) as compared with secondary methods (evidence synthesis) or primary methods (survey). Concerning data visualisation, the study showed different stakeholders, particularly researchers, desire the use of data visualisation when communicating research, although few examples of experiences with developing and using them. The participants also expressed the difficulty in finding the right tool to use to present research findings.

In the third project, I focused on the development of methodological guidance, aimed at systematically identifying and reporting research gaps, to provide a clearer picture of the status of the evidence base. The guidance highlights the importance of clearly reporting research gaps for subsequence clear identification of research gaps. The methodological guidance merged findings from the scoping review and the qualitative study. It demonstrates when identifying and addressing

research gaps in a topic area, the most important step is to first define clearly what is meant by a research gap in the text. This can be done by either adopting an existing definition that best describes the research gap or explicitly describing the nature and type of the research gap. Describing the nature of the research gap can involve different items, as follows:

1) State the scale of the research gap (i.e., specific or broad)
2) Clearly define the research gap
3) Specify the cause of research gap

The methodological guidance involves six steps: 1) specify the topic area and/or research question; 2) map and clearly state the existing research gaps ; 3) identify research gap(s); 4) clearly describe the research gaps(s) identified; 5) characterize the research gaps; and 6) present the research gaps. To be able to determine the feasibility and usability of this methodological guidance in practice, the importance, relevance, and applicability of each step proposed needs to be thoroughly discussed among different stakeholders, then implemented and evaluated accordingly. The evaluation of the methodological guidance will help determine its applicability and future implementation and adaption in health research and other fields.

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List of publications

Paper1: Nyanchoka L, Tudur-Smith C, Thu VN, Iversen V, Tricco AC, Porcher R: A Scoping Review describes Methods Used to Identify, Prioritize and Display Gaps in Health Research. *J Clin Epidemiol* 2019.

Paper 2: Nyanchoka L, Tudur-Smith C, Porcher R, et al: Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study protocol *BMJ Open* 2019;9: e027926. doi: 10.1136/bmjopen-2018-027926

Paper 3: Nyanchoka L, Tudur-Smith C, Porcher R, Hren D: Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study (BMJ Open – Accepted October 2020)

Related publications

Nyanchoka L, Tudur-Smith C, Nguyen VT, *et al* 34 Methods for identifying and displaying research gaps *BMJ Evidence-Based Medicine* 2018;**23:**A17-A18.

Other Publications

Al-Shamsi M, Moitinho de Almeida M, Nyanchoka L, Guha-Sapir D, Jennes S. Assessment of the Capacity and Capability of Burn Centers to Respond to Burn Disasters in Belgium: A Mixed-Method Study. *J Burn Care Res*. 2019;40(6):869-877. doi:10.1093/jbcr/irz105

Presentations

Poster

- *Methods to Identify and Display Gaps in Clinical Research,* Evidence Live Conference, Oxford (UK) - June 21–22, 2017- https://zenodo.org/record/1165596#.XGQwuFxKiM8

- *Methods to Identify and Display Gaps in Clinical Research,* Global Evidence Summit, Cape Town (South Africa) - September 13–16, 2017 https://zenodo.org/record/1165628#.XGQwnlxKiM8

Oral

- *Methods for Identifying and Displaying Research Gaps: Scoping Review* - Evidence Live Conference, Oxford (UK) - June 21–22,2018 https://zenodo.org/record/1303904#.XGQvnlxKiM8
- *Methods for identifying and displaying gaps in clinical research*, Cochrane Colloquium, Edinburgh (UK) - September 2018

Invited

Presentation on Methods for Identifying Gaps in Health Research at the following-:

- The European Clinical Research Infrastructure Network (ECRIN), Paris, France – January 2017, https://zenodo.org/record/1165946#.XGQwQVxKiM8
- Annual meeting of Pierre Louis Doctoral School of Public Health of Paris, Saint Malo (France) – October 2017 and October 2018
- The Centre of Research in Epidemiology and Statistics Sorbonne Paris Cité, Paris (France) – November 2016 and January 2017
- The European Clinical Research Infrastructure Network, Paris (France) – January 2017
- The Evidence Based Research Network representative, Remote presentation – February 2017
- National Institute for Health Research (NIHR), UK – November 2018

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Chapter 1: Introduction

The general topic of this PhD focuses on methods to identify gaps in health research. This is a cross-cutting area that explores definitions of research gaps and the importance of identifying and understanding gaps in health research. It focuses on understanding the existing body of evidence and what is missing, needed and should be prioritized to improve how we use evidence to inform health practice, policy and future research.

1.1. Rationale for thesis

The term "research gap" is not standardized, and its meaning can differ depending on the research context. In this study, we adopted the definition from the National Collaborating Centre for Methods and Tools (NCCMT) in Canada, which describes a research gap as a research question for which missing or insufficient information limits the ability to reach a conclusion [1]. To further understand research gaps and their causes, we also refer to a paper by Robinson et al. that developed a framework for identifying research gaps from systematic reviews by characterizing the gap with use of Population, Intervention, Comparison, Outcome and Setting (PICOS) elements and identifying reason(s) for why the gap exists, including insufficient or imprecise information, biased information, inconsistency or unknown consistency, and incorrect information [2].

Upon identifying a research gap, prioritizing research based on the gap is essential to determine its importance and relevance, especially based on feedback from key stakeholders such as patients, clinicians, researchers, advocates and funders. Research priority-setting is not consistently defined, although it has been described as any interpersonal activity that leads to the selection of topics or key questions to further investigate [3]. Research prioritization processes can help researchers and policy-makers effectively target research that has the greatest potential health benefit. Consensus

is lacking on what constitutes the best methodological approaches to identify research gaps [2, 4], determine research priorities [3, 5] and display research gaps or priorities.

1.2. Aims and objectives for thesis

The overall aim of this PhD project was to provide methodological guidance on approaches to identify gaps in health research. The research was guided by three main objectives:

1) Identify different definitions reported for the term "research gap" and describe methods for identifying and displaying gaps in health research. This involved a scoping review of studies describing or reporting methods to identify, prioritize, and display gaps or priorities in health research.

2) Explore key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research. This was achieved by a qualitative study conducted with semi-structured interviews.

3) Develop methodological guidance for identifying gaps in health research. This was achieved by combining the scoping review and qualitative study findings.

1.3. Thesis structure

The thesis is structured in 6 chapters. Chapter 1 introduces the aim and objectives of the thesis. Chapter 2 provides background literature to describe the context of this thesis. Chapter 3 presents the scoping review, which describes different definitions reported for the term "research gap" and methods for identifying and displaying gaps in health research. Chapter 4 presents the qualitative study that explored key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research. Chapter 5 describes the development of methodological guidance on identifying gaps in health research. Chapter 6 concludes with a summary of the main findings and recommendations for future work.

Chapter 2: Background Literature

2.1. Health research and research gaps (defining research gaps)

An estimated 85% of health research is avoidably wasted. More than 50% of all health research does not get published at all. From the published research, 50% is not usable in practice because items are missing. Also, among the remaining 25%, half contains design flaws. Planning a study focusing on the wrong question is a frequent cause of waste in research[6]. Hence, published, unpublished, completed and on-going research should be used to assess whether research gaps justify new research and can inform the design, conduct and reporting of further research.

The term “research gap” is commonly referred to in the literature, yet the criteria used seem to be ambiguous and vague. Different researchers have highlighted the importance of identifying gaps in health research and reported the lack of systematic methodological approaches for identifying, prioritizing and addressing gaps [7-9]. Robinson et al. argued that a clear and explicit identification of research gaps is a necessary step in developing a research agenda, including decisions about funding and the design of informative studies. The authors provided reasons for research gaps such as insufficient or imprecise information, biased information, inconsistency or unknown consistency and not the right information [2, 7]. Their study focused on a literature search for published articles that described the identification of research gaps from systematic reviews or related processes such as health technology assessments. The authors subsequently developed a framework for identifying and prioritizing research gaps from systematic reviews using the PICOS method[2]. This framework has since been widely adopted mainly in health research. It provides a basis for identifying research gaps; although it mainly focuses on using systematic reviews, other methods are applicable.

Carey et al., conducted a study that evaluated 19 Cochrane collaboration systematic reviews; the authors did not find any implemented structured approach to research gap identification or prioritization [8]. Trikalinos and colleagues performed a study to assess reporting of recommendations for future research needs in secondary research publications. The authors empirically assessed 50 randomly selected peer-reviewed systematic reviews published in high-impact peer-reviewed journals between 2005 and 2010. Most included some discussion of future research needs (40/50, 80%) and many identified specific research questions that should be addressed by future studies (36/50, 72%). However, specific research designs were suggested in 23 (46%) reviews. In 20 of these 23 papers, the recommendation was that more randomized controlled trials are necessary. Only 13 (26%) reviews devoted a whole paragraph to discuss future research needs. None of the reviews reported whether any specific methodology was used to identify or prioritize future research needs [10]. Hence, this study showed how little attention is given to gap identification, addressing gaps and future research.

Another key study is by Ballini et al., which developed and proposed a method evaluating and ranking scientific uncertainty. The authors used using a 5-step evaluation process: 1) definition of the technology's evidence profile and all relevant clinical outcomes; 2) systematic review of the scientific literature and outline of the *uncertainty profile* differentiating research results into *steady* results (results that are highly unlikely to be changed by further studies)*, plausible results* (consistent results from sufficiently numerous high-quality observational studies and related to outcomes for which comparative evaluations are not strictly necessary), *uncertain results* (results that would most probably change, in both size and direction of estimate, if evaluated in randomised clinical trials) and *unknown results* (unreported/non-existent results on outcomes judged by the panel to be relevant for evaluating the technology); 3) definition of the acceptable

level of uncertainty for investing research resources; 4) analysis of local context; and 5) identification of clinical indications with promising clinical return[11]. The principle they used to differentiate levels of uncertainty was an adaptation of the grading of the level of evidence for developing recommendations for clinical practice developed by the GRADE group. The Grades of Recommendation, Assessment, Development, and Evaluation (GRADE) method involves assessment of both the level of evidence and the strength of each recommendation [11, 12]. The study also found a variety of approaches used but did not identify any empiric work to recommend a "best practice" and recommended future work on evaluating scientific uncertainty using the proposed steps.

A study by Li and colleagues aimed to test a framework for prioritizing clinical questions and identifying evidence gaps by using existing systematic reviews and clinical practice guidelines. The framework included the following steps: 1) deriving clinical research questions from clinical practice guidelines to reflect issues that clinicians encounter frequently, 2) asking clinicians to prioritize questions for research to incorporate opinions from evidence users, 3) determining whether high-quality systematic reviews of all previous research exist for each clinical question, and 4) identifying evidence gaps. By mapping evidence gaps to clinicians' priorities, they proposed a comparative effectiveness research agenda[13].These studies all showed the complexities of gap identification and combination of methods being used and implemented.

2.2. Distinguishing research gaps

The first step in better understanding how to identify research gaps is to explore the existing definitions and related terms currently used. In this section, we explore what different authors have documented on definitions related to gaps in research. Robinson et al. define that a research gap

arises when the ability of the systematic reviewer to draw conclusions is limited and that research gaps represent an output (of literature reviews)[7]. Müller-Bloch et al. added that a research gap also holds a function as a starting point for research and also perceived it as an input because it can motivate further research [9].

Müller-Bloch et al. distinguished a research gap, research problem and research agenda. They highlighted that the term research problem might occasionally be used as a synonym for a research gap. A problem statement represents "a gap in sets of information that, when examined carefully, results in a call for action or resolution", and "research seeks to resolve the disparate sets of information through the generation of new knowledge and the introduction of theory" [9, 14]. A research gap was described as arising when there is a gap in sets of information derived from a literature synthesis and requires further research to be resolved [2, 9, 14]. Müller-Bloch et al. argued that the definition is narrow and limiting, given that one can derive research gaps from sources other than literature syntheses [9], which I agree with and observed from our study findings. A research agenda is defined as a set of questions for further research [15]. Müller-Bloch et al. argued that "the literature does not provide information on the link between a set of research gaps, and whether or not they may be called a research agenda. Whereas research gaps can directly lead to questions for further research, this does not necessarily mean that all questions for further research are derived from research gaps. Thus, it can be assumed that research gaps are a part of research agendas, but not necessarily exclusively"[9].

Figure 1. Key terms related to research gaps

Research Gaps	Research Problems	Research Needs	Research Agenda
• "research gap arises when there is a gap in sets of information derived from a literature synthesis and requires further research to be resolved"[2, 12, 13]	• The term research problem might occasionally be used as a synonym for research gaps[12]. • "a research problem is a problem statement that is resolved by means of research. Furthermore a problem statement represents 'a gap in sets of information that, when examined carefully, results in a call for action or resolution', and 'research seeks to resolve the disparate sets of information through the generation of new knowledge and the introduction of theory.' " [12, 13]	• Research needs are those areas in which the gaps in the evidence limit decision making by patients, clinicians, and policy makers [2].	• "a research agenda is defined as a set of questions for further research." [14] • "a research agenda is a gap in sets of information that, when examined carefully, results in a call for action or resolution", and "research seeks to resolve the disparate sets of information through the generation of new knowledge and the introduction of theory." [12, 13]

As we can see from Figure 1, the terms related to research gaps vary and are rather confusing to distinguish the differences. Evidence shows that characterizing research gaps deepens the understanding of how research gaps may be constituted and may thus help in identifying research

gaps in literature reviews [2, 9]. Similar to the link of the research agenda with research gaps, Jacobs (2011) identified six kinds of research problems as presented below[9, 14].

1) **Provocative exception** - When new research findings contradict widely accepted conclusions. This is not evident in the literature and requires critical appraisal, to carefully analyze and scrutinize even subtle discrepancies [9, 14].
2) **Contradictory evidence** – The contradictory evidence is related to the provocative exception. It occurs if results from studies allow for conclusions in their own right but are contradictory when examined from a more abstract point of view [9, 14].
3) **Knowledge void** - First, knowledge may not exist in the actual field of research but in a related research domain. In this case, it may be necessary for scholars to refer to theories and literature from related research domains. Second, results of a study may differ from what was expected [9, 14].
4) **Action-knowledge conflict** - Arises when the actual behavior of professionals differs from their advocated behavior [14]. Also, a methodological conflict may occur due to the influence of methodology on research results [9, 14].
5) **Methodological conflict** - When use of one or another research methodology may contribute a source for a research problem [9, 14].
6) **Theoretical conflict** - If one phenomenon is being explained with various theoretical models, there might be a theoretical conflict [14].

Another characterization of research gaps by Robinson et al. is the classification of research gaps based on the most important reason(s) for the existence of the gap. The reason(s) indicated would be those that most preclude conclusions being drawn. The following are the proposed classifications of the reasons for research gaps:

1) **Insufficient or imprecise information -** Can arise if no studies are identified, if a limited number of studies are identified or if the sample sizes in the available studies are too small to allow for conclusions about the question of interest[2, 9].
2) **Biased information** - Various criteria exist for assessing the risk of bias of studies of different designs. The aggregate risk of bias depends on the risk of bias of the individual studies[16]. In addition to considering methodological limitations of studies, the appropriateness of the study design should also be considered[2].
3) **Inconsistency or unknown consistency** - Consistency is defined as the degree to which reported effect sizes from included studies appear to go in the same direction. Statistical measures of heterogeneity may be used to help in evaluating consistency. If there is only one available study, even if considered to have a large sample size, the consistency of results is unknown[2, 16].
4) **Not the right information** - First, results from studies might not be applicable to the population and/or setting of interest. Second, the optimal or most important outcomes might not be assessed. Third, the study duration might be too short, and patients might not be followed up for long enough to adequately assess some outcomes that might be most important [2].

In this PhD thesis, I also identified other characteristics of gaps that are based on the scale of their existence (i.e., broad or specific). For example, a study of health research and development explored the use of data from registered clinical trials to identify gaps in health research and development. This study demonstrated how mapping studies can be used to identify research gaps in broad areas[5]. The study focused on the International Clinical Trials Registry Patform (ICTRP) database, established by the World Health Organization (WHO) in 2005 to create a platform for

linking clinical trial registries and provide a single point of access to information on all clinical trials conducted globally[17]. It explored what can be learned from the clinical trial records available in the ICTRP database. It also explored the current composition of the global landscape of health research and development, particularly in the distribution of trials across different diseases and countries and the identification of any major gaps in the landscape[5].

A similar study by Atal et el. evaluated the alignment between the research effort (measured as the number of randomised controlled trials conducted) and the burden of disease across all world regions and a broad range of diseases[18]. The study was on a global level; it estimated the research effort across non–high-income regions and identified the regions for which the research effort was too low as compared with the regional disease burden. The authors highlighted that the research gaps shown may be considered by local funders or health authorities to drive research toward local needs. Furthermore, analyses to identify what research type is more likely to help reduce the largest amount of burden. Both of these studies demonstrate mapping gaps on a broad scale and not specific to an individual study. Therefore, an additional characteristic of research gaps is whether they are specific or broad in nature.

2.3. Summary

The literature shows complexity and variability on defining research gaps. I also found a need to synthesize the evidence on defining research gaps and methods to identify research gaps. In efforts to address this, the next chapter focuses on my first study, a scoping review to map reported definitions of research gaps and methods to identify, prioritize and display gaps in health research.

Chapter 3: Mapping Methods to Identify, Prioritize Gaps in Health Research

3.1. Background

The current body of research is growing, with more than 1 million clinical research papers published from clinical trials alone [19]. Planning a study focusing on the wrong question is a frequent cause of waste in research [6]. Hence, completed and on-going research should be used to assess whether research gaps justify new research and can inform the design, conduct and reporting of further research [20]. Initiatives such as the James Lind Alliance, UK Database of Uncertainties about the Effects of Treatments, Cochrane Agenda and Priority Setting Methods Group and Evidence-based Research Network are some examples of existing efforts to identify and prioritize research gaps in health.

The term "research gap" is not standardized, and its meaning can differ depending on the research context. In this study, I adopted the definition from the NCCMT in Canada, which describes a research gap as a research question for which missing or insufficient information limits the ability to reach a conclusion [1]. To further understand research gaps and their causes, we also referred to a study by Robinson et al. that developed a framework for identifying research gaps from systematic reviews by characterizing the gap with use of PICOS elements. The study also identified reason(s) for the gap's existence, including insufficient or imprecise information, biased information, inconsistency or unknown consistency, and incorrect information [2].

Upon identifying a research gap, prioritizing research based on the gap is essential to determine its importance and relevance, especially based on feedback from key stakeholders such as patients, clinicians, researchers, advocates and funders. Research priority setting is not consistently defined, although it has been described as any interpersonal activity that leads to the selection of topics or

key questions to further investigate [3]. Research prioritization processes can help researchers and policy-makers effectively target research that has the greatest potential health benefit.

Consensus is lacking on what constitutes the best methodological approaches to identify research gaps [2, 4], determine research priorities [3, 5] and display research gaps or priorities. Therefore, we considered that a scoping review on this topic area was warranted. Our objectives were to 1) identify different definitions reported for the term "research gap", 2) explore methods used to identify research gaps, 3) describe methods used to determine research priorities, and 4) map methods used to display research gaps or research priorities.

3.2. Method

The analytic framework for this scoping review involved the methodology outlined by Arksey and O'Malley [21] and further refined by members of the Joanna Briggs Institute [22]. It entails identifying the research question; expert consultation on conceptualizing the research topic, identifying the different key terms for the search strategy; developing the items for the data extraction form and reviewing the manuscript; searching for relevant studies by using key terms; selecting studies; charting the data; collating, summarizing, and reporting the results; and consulting with stakeholders to inform study findings. Experts played a major role in this study; their role was important because of the uncharted nature of this topic area.

3.2.1. Search methods for identification of documents

The scoping review aimed to identify and include a wide range of article types, including original research, protocols, conference proceedings and website content. The goal of the search strategy was to identify a diversity of methods used to identify, prioritize and display gaps or priorities in

health research. To build the search terms for the search strategy, because of the variability in terminology used, we began by contacting experts to ask for the terms, descriptions and definition they use to refer to research gaps. Upon compiling different terms, we built our search terms with the assistance of a research librarian. The final search terms included "identifying gaps in research", "research gaps", "evidence gaps", "research uncertainties", "research gaps identification", "research gaps prioritization" and "methods" in health research including public health and clinical research. Two reviewers (Linda Nyanchoka, LN and Van Nguyen, VN) conducted the searches by using the Peer Review of Electronic Search Strategies (PRESS) guideline checklist, which aims to improve the quality of database searches [23].

The databases searched were MEDLINE, PubMed, EMBASE, Cochrane Library, Scopus, Web of Science, PROSPERO register, TRIP, Google Scholar and Google. To focus on the most current research, database searches were limited to the past 10 years (2007-2017). Additional searches involved hand searches, web searches, expert suggestions, and checking reference lists of highly relevant articles. Only studies reported in English and involving humans were included to increase the feasibility of this scoping review. See **Appendix B** for complete search strategies.

3.2.2. Eligibility criteria

Studies were included if they aimed to describe a methodology and/or applied some methodology to identify gaps, determine research priorities, and/or display gaps or priorities in health research. All study designs were eligible, including those that used qualitative or quantitative methods, methodology or guideline reports. We focused our inclusion criteria to capture reports within the domain of health, reporting on and/or describing methods for identifying, prioritizing and/or

displaying research gaps. We excluded publications that did not explicitly describe how they aimed to identify, prioritize and/or display research gaps. For additional information, see **Appendix C.**

3.2.3. Abstract and full-text screening and selection of articles

Abstract and full-text screening was performed by two authors. The first reviewer (LN) performed the entire screening of 1938 abstracts, and a second reviewer (VN) screened 10% (194/1938) of all abstracts. Agreement on selection of abstracts was 174/194 (90%). In total, 237 articles were selected for full-text screening: LN performed the entire screening and VN screened 10% (24/237) of articles. Among the 24 articles that were doubled screened, agreement was reached on 20 (85%). Title and abstract screening involved use of the software package Covidence for conducting systematic reviews. Full-text screening involved using EndNote to manage and retrieve full texts. Discrepancies in both abstract and full-text screening were resolved in a meeting with senior researchers.

3.2.4. Data charting and synthesis

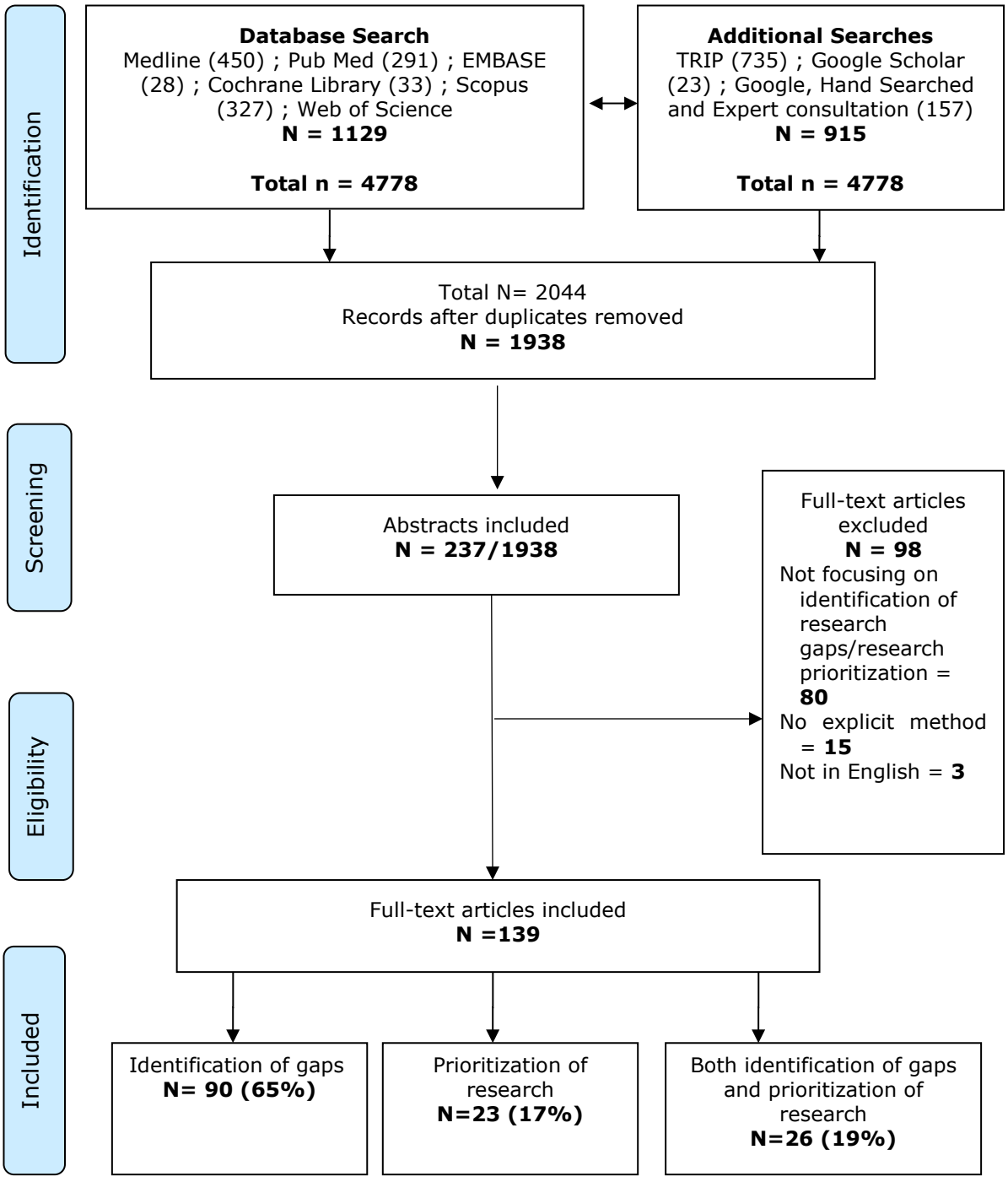
Data charting involved the use of a Google Form developed by LN with expert consultation, guidance from senior researchers and reviewing a previous methodological study[24]; the form was calibrated by LN and VN. See **Appendix D** for the complete data extraction form. All data collection and analyses were conducted by LN, and VN extracted 10% (14/139 of all full-text articles). As an additional data cleaning step, two senior researchers then verified and discussed the 14 articles extracted by the second reviewer, to ensure data accuracy. A 95% agreement was achieved; disagreements were mainly on interpretation of methods used to identify gaps and/or determine research priorities. Disagreements were resolved in a meeting with senior researchers. We extracted the following data from articles: type of article, main objective of the study, main

study methodology, definition of research gaps, and specific methods to identify research gaps, determine research priorities and display research gaps or priorities [24]. The synthesis included a quantitative analysis (i.e., frequency analysis) and qualitative analysis (i.e., thematic analysis) of the components of the methods to identify, prioritize and/or display gaps in health research and conceptual definitions of gaps in health research [25].

3.2.5. Results of the search

The literature search retrieved 2,044 citations, and after duplicates were removed, 1,938 remained. Overall, 247 references were considered potentially eligible. After full-text assessment, 98 articles were excluded, and 139 were included. Figure 2 shows the flow of articles through the scoping review.

Figure 2. Scoping review flow diagram



3.3. Results

3.3.1. Summary of study designs

Among the 139 included articles, 90 (65%) aimed to identify gaps, 23 (17%) aimed to determine research priorities and 26 (19%) focused on both identifying gaps and determining research priorities.

Table 1. Study designs used

Study Design	All Articles	Identification of research gaps	Research prioritization	Both identification and prioritization
	(N=139)	**(N=90)**	**(N=23)**	**(N=26)**
Primary research	**25 (18%)**	**8 (9%)**	**13 (56%)**	**4 (15%)**
Qualitative study	3 (12%)	1 (13%)	2 (15%)	0
Quantitative survey	2 (8%)	1 (12%)	2 (15%)	1 (25%)
Both qualitative study and quantitative survey	20 (80%)	6 (75%)	9 (69%)	3 (75%)
Secondary research	**85 (61%)**	**77 (86%)**	**5 (22%)**	**3 (12%)**
Knowledge synthesis				
Systematic review*	36 (42%)	33 (43%)	1 (20%)	2 (67%)
Scoping review	25 (29%)	23 (30%)	2 (40%)	0
Evidence mapping	4 (4%)	3 (4 %)	0	1 (33%)
Mapping study	2 (2%)	2 (3%)	0	0
Literature review	4 (4%)	4 (5%)	0	0
Umbrella review	4 (4%)	4 (5%)	0	0
Other (integrative review, critical interpretive synthesis)	8(9%)	6 (8%)	2 (40%)	0

Bibliometric analysis	2(1%)	2(3%)	0	0
Both primary and secondary research	**29 (21%)**	**5 (6%)**	**5 (22%)**	**19 (73%)**
Review of evidence and quantitative study	6 (21%)	0	1 (20%)	5 (26 %)
Review of evidence and qualitative study	3 (10%)	2 (40%)	0	1 (5%)
Review of evidence and both quantitative and qualitative study	20 (69%)	3(20%)	4 (80%)	13 (68%)

*Including methods used in Health Technology Assessments.

3.3.2. Definitions of research gaps reported in articles

We explored the definitions as reported in the included studies. We identified a total of 12 different definitions, some of which overlapped, as presented in Box 1. Three cross-cutting themes were identified: definitions related to missing information, inadequate information and insufficient information.

Box 1: Definitions of research gaps as reported by the scoping review

Research/evidence/knowledge gaps

- not much information is available and/or there is a lot of uncertainty about the accuracy of the existing estimates/evidence [4]
- additional research is needed, from policy-makers perspectives, to address the evidence gap in the available primary research [26]
- evidence is missing from a body of research on a particular topic that could otherwise potentially answer the questions of decision-makers (clinicians, other practitioner groups, administrators, policy-makers) [26]
- the evidence base inadequately addresses a key question [27]

- desired research findings do not exist [9, 22, 28]
Synthesis/unidentified gaps - little or no evidence from systematic reviews is available and could be a valuable resource to inform the evidence base in a particular area [29, 30] - lack of up-to-date and conclusive systematic reviews at low risk of bias mapped to a clinical question [31]
Treatment uncertainty - lack of up-to-date, reliable systematic reviews of research evidence addressing the uncertainty about the effects of treatment, and/or up-to-date systematic reviews of research evidence show that uncertainty exists [32]
Absolute evidence gaps - little or no evidence from primary studies is available [29]
Practical knowledge gap (action–knowledge conflict gap) - professional behavior or practices deviate from research findings or are not covered by research [9, 22, 28]
Empirical gap (evaluation void gap) - research findings need to be evaluated or empirically verified [9, 22, 28]
Population gap - research regarding a population that is not adequately represented or under-researched in the evidence base or prior research (e.g., sex, race/ethnicity, age) [2]
Methodological gap (method and research design gap) - a variation in research methods is required to generate new insights or to avoid distorted findings [9, 22, 28]
Theoretical gap (theory-application void gap) - theory should be applied to certain research issues to generate new insights; theory is lacking, so a gap exists [9, 22, 28]

3.3.3. Specific methods for identifying research gaps

I then classified the methods used to identify research gaps. The most frequent methods in the review were aimed at gap identification (including both identification and prioritization) and involved secondary research, including knowledge synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80, 73%) (Table 1). Overall, 24/116 (21%) articles described the use of both primary and secondary research and 12/116 (10%) only primary research. I found 7 specific methods for identifying research gaps that are described along with the purpose of the method (Table 2).

Table 2. Overview of specific methods to identify research gaps

Methods to identify research gaps	Definition	Purpose
Primary research methods		
Quantitative Survey [31]	A scientific procedure for collecting information and making quantitative inferences about a pre-defined population	Determine evidence gaps by using a Likert-type response scale with scoring from 0 (not important at all) to 10 (highly important)
Academic crowd-sourcing [4]	An emerging paradigm that is based on harnessing the power of the crowd to solve problems [4]	Aims to reach a wider range of people, which may sometimes be required to solve a problem correctly and efficiently, including identifying research gaps [4]

Needs assessment [33, 34]	A systematic process for determining and addressing needs, or "gaps" between current conditions and desired conditions or "wants" by using various techniques including primary or secondary research methods (e.g., reviewing evidence, guidelines, and conducting interviews) [28]	Clarify problems and identify appropriate interventions or solutions [28]
Secondary research methods		
Knowledge synthesis Systematic review [35-38]* Scoping review [39-41] Evidence mapping [29, 42, 43] Mapping study [44-46] Umbrella review [3] Integrative review [47] Critical interpretive synthesis [48]	Efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored [49]	Summarize all pertinent studies on a specific question; improve the understanding of inconsistencies in diverse evidence and identify gaps in research evidence to define future research agendas [49]
Bibliometric study [50, 51]	The quantitative study of bibliographic material used to examine the knowledge structure and development of research fields based on analysis of related publications [52]	Provide a general picture of a research field that can be classified by papers, authors and journals [52]
Both primary and secondary research methods		

Priority setting [31, 32, 53-63] James Lind Alliance priority setting partnership (JLA PSP), Cochrane Priority Setting (*consists of four steps: the first two aim at gap identification and the last two at research prioritization*)	JLA PSP methods were designed to allow clinicians, patients and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that could be answered by research by gathering research questions, checking existing research evidence, interim prioritization and a final consensus meeting to reach agreement on the top 10 research priorities [53]	Raise awareness of research questions that are of direct relevance and potential benefit to patients and the clinicians who treat them, to lead to changes in how research funding is granted [53]
Global evidence-mapping methods [42, 64]	Maps available research and provides an overview of a broad range of research questions and identifies evidence gaps [42]	Characterize the breadth, depth, methodology of relevant evidence and make this readily accessible [65]; identify research gaps

*including methods used in Health Technology Assessments

3.3.4. Specific methods for determining research priorities

Among the 49 studies aimed at research prioritization, the most frequent method involved both primary and secondary research, accounting for 24 (49%) studies, followed by primary research 17

(35%), and secondary research 8 (16%) (Table 3). We identified five specific methods for determining research priorities.

Table 3. Overview of specific methods to determine research priorities

Methods to determine research priorities	Description	Summary of steps if specified
Primary research		
Delphi survey [63, 66-68]	A group facilitation technique that seeks to obtain consensus on the opinions of relevant stakeholders by a series of structured questionnaires (commonly referred to as rounds). The questionnaires are completed anonymously by the experts (commonly referred to as panelists, participants or respondents) [69]	Involves a series of questionnaires that are completed anonymously by experts. A process of group communication without the group ever meeting face to face. The responses from each set of questionnaires are analyzed, summarized, and then sent back to the participants until a large degree of consensus is reached in the area of interest.
Quantitative survey [31]	Adapted to determine participant research priorities by using forced raking of research questions and Likert-type scale for responses	Developing and testing questionnaires to address research questions Forced ranking of research questions Likert-type scale
Secondary research		
Knowledge synthesis	Efficient scientific approach for identifying and summarizing evidence that allows for assessing	Summarizes all pertinent studies on a specific question; can improve the

Systematic review [36] * Scoping review [39, 40]	the generalizability and consistency of research findings and exploring data inconsistencies [49]	understanding of inconsistencies in diverse evidence and identify gaps in research evidence to define future research agendas [49]
Both primary and secondary research		
Priority setting [19, 31, 32, 55, 56, 58, 59, 61, 62, 66, 70-79] *Example: JLA PSP methods*	Designed to enable clinicians, patients and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that could be answered by research [32]	Survey to identify treatment uncertainties Review of existing systematic reviews to explore existing evidence and address treatment uncertainties Interim prioritization to identify the priorities of relevant individuals and stakeholder groups Focus groups to discuss the research priorities based on missing or inadequate evidence A final consensus meeting to reach agreement on the top 10 research priorities [32]
Global evidence-mapping method [42, 64]	Maps available research and provides an overview of a broad range of research questions and identifies evidence gaps [42]	Question development involving expert consultation, preliminary literature search, mapping workshop, online survey and development of clinical question Question prioritization Evidence search and selections

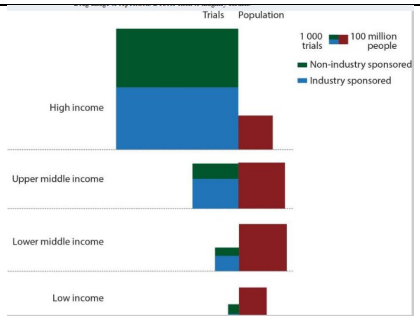
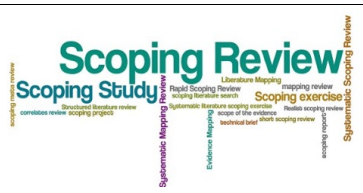
*including methods used in Health Technology Assessments

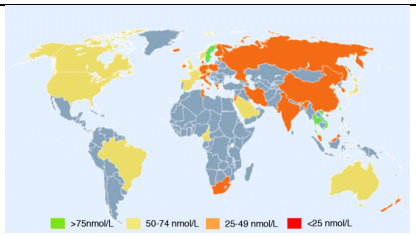
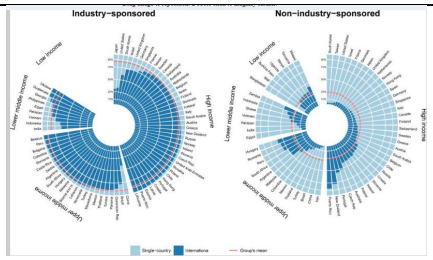
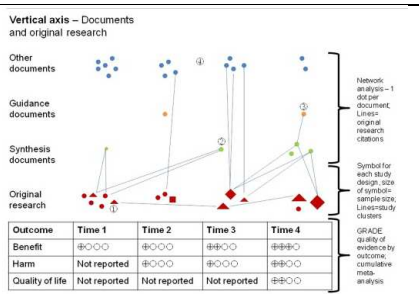
3.3.5. Specific methods for displaying research gaps and research priorities

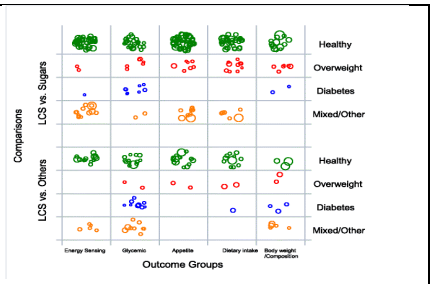
We identified 14 unique methods used to display research gaps and/or research priorities and give some examples of these methods (Table 4). We provide some illustrations of non-traditional methods. An illustration of all methods can be found in **Appendix E**.

Table 4. Overview of methods to display gaps and research priorities

Format	The information on research gaps and priorities displayed	Ways of displaying research
Table [19, 26, 31, 32, 40, 42, 57, 59-61, 64, 69, 71, 80-82]	List of clinical questions, gaps and research priorities Ranking quality of evidence Study designs to address research questions Scoring of each research gap List of research questions Prioritization of research questions Metric of ranking information	Table format *
Box plot [31]	List of research questions Ranking of research questions	Box plot format *
Bar graph/horizontal bar graph [31, 44, 75, 83-89]	List of research priorities Frequency of questions prioritized Number of studies and categories studied Frequency of research questions Quality of evidence metric	Bar graph format *

Scatter plot [90]	Numeric values of desired research and current research	Scatter plot format *
Funnel plot [91]	Number of studies included in the review Effect sizes of studies on the x-axis	Funnel plot format *
Pie chart [18, 85]	Proportional size of health problems being investigated by trials registered in a registry platform	Pie chart format *
Mind maps [92]	Diagram used to represent concepts, ideas or tasks linked to and arranged radially around a central key word or idea Primary branches represent the major ideas or themes around the central topic, and secondary branches tend to include more concrete illustrative examples	Mind map format *
Tree map chart [44]	Number of clinical trials, population and income group Comparison study of clinical registry data vs global health research data from the Global Burden of Disease	 [44]
Word cloud [93]	Frequency of words Frequency of words between two groups	

		[93]
Geographic map [8, 35]	Studies mapped around the world using colors on a pre-defined health outcome. Different shapes and sizes also used for additional information on a map.	 [35]
Dot plot [94]	Number of studies Quality of evidence Different colors to show different study designs	 [94]
Radial bar plot/ polar histogram [44]	Proportion of trials in several countries using one color per country among two groups over a period of time Comparison of proportion of trials in several countries using one color per country among two groups over a period of time	 [44]
Schematic representation [50]	Horizontal axis represents time, and vertical axis represents different documents	 [50]

Bubble plot/chart [29, 95, 96]	Bubbles represent studies; size indicates the relative number of studies and color the study design Number of studies by intervention type and health status Compares three sets of values	 [96]
* Examples available in **Appendix E.**		

3.4. Discussion

These findings demonstrate that the term “research gap” significantly differs across research contexts and there is no common definition. It also reveals no clear methodological guidance on which methods should be used to identify research gaps or determine research priorities. This situation leads to a wide variety in methodology, for difficulties in comparing results across studies. Also, many studies aimed at identifying gaps relied on secondary research, primarily systematic reviews. Systematic reviews are considered the gold standard in providing the highest level of evidence for the relative efficacy and safety of interventions [97] and summarizing the overall quality and results of research. A study of identifying and prioritizing research gaps corroborated that systematic reviews are the standard for evaluating the existing state of scientific knowledge regarding a specific clinical or policy question [8]. Robinson et al. also developed a framework for using systematic reviews to identify research gaps [2]. Although these two studies show that systematic reviews can identify research gaps, most systematic reviews address a highly focused question related to the existing evidence and thus present difficulties for explicitly identifying research gaps in a general area [2, 8, 98].

Other secondary research methods identified in this review were overviews of reviews, also known as umbrella reviews, scoping reviews and evidence mapping. Overviews of reviews focus on a much broader area, compiling evidence from multiple reviews into one accessible and usable document and highlighting other reviews within the specified topic area [99, 100]. Moreover, scoping reviews and evidence mapping are designed to describe existing evidence in a broader content area [96, 101, 102]. They descriptively summarize results, which can be presented in a user-friendly format, often a visual figure called an evidence gap map [103], or a searchable

database, to improve research planning, strategic research prioritization, and evidence-informed policies.

A mix of primary and secondary research was most frequently used to determine research prioritization, namely, priority setting (e.g., JLA PSP methods). These methods involve the participation of patients, caregivers and healthcare and social-care professionals in identifying research questions, then prioritizing them by using a combination of primary and secondary research [19, 31, 32, 55, 56, 58, 59, 61, 62, 66, 70-79]. The main method for determining research prioritization with primary research was the Delphi survey, which is a practical and productive approach to obtaining opinions from a wide number of relevant experts for identifying potential priority topic areas for research [63, 66-68].

To display research gaps, half of the methods still used traditional ways to present findings (e.g., summary table and bar charts), and the other half used more advanced ways to display information (e.g., tree map charts, radial bar plots and bubble plots). The non-traditional methods used more colors and diagrams to present research gaps or priorities. For example, bubble plots use different shapes, sizes and colors to display information and can be used to present up to three different variables in one diagram. These characteristics could be further explored to determine the appropriate method to be used along with specified methods to identify research gaps and/or determine research priorities.

Finally, the study confirmed that the various methods identified consist of both emerging and established approaches. Nonetheless, these methods can provide rich contextual details for establishing methodological guidance. I propose more work to improve the understanding of the

methods and investigate ways to give the public, patients, clinicians, health researchers, decision-makers and funders more opportunities to know what methodologies are available and applicable.

The scoping review had some limitations. How the term "research gap" was used and defined varied widely among different authors and articles, and our exploration of methods for identifying research gaps, determining research priorities and displaying research gaps and priorities relied on definitions used by authors of included studies. Therefore, it may have missed some methods due to lack of clear definitions. Also, the study included only documents written in English, relying on key articles to identify the steps involved in each method and as presented by the authors of the included articles. As such, it may have missed important methods that were published in other languages. Additionally, the time restriction to the last 10 years provided a comprehensive list of recent methods used rather than an exhaustive list of all methods used.

Finally, I anticipate that the results will be of interest to knowledge users, including patients, public, clinicians, researchers, decision-makers, funders, key organizations such as the JLA, the UK Database of Uncertainties about the Effects of Treatments, Joanna Briggs Institute, Campbell Collaboration, Africa Evidence Network, Cochrane Priority Setting Methods Group and Evidence-based Research Network, and methodologists focused on identifying and displaying gaps, and determining priorities in health research.

3.5. Summary

This study provided an overview of different methods used for and/or reporting on identifying gaps, determining research priorities and displaying both gaps and research priorities. The findings can inform the development of standardized methods to identify, prioritize and display gaps. They

can inform further research and evidence-based decision-making by providing descriptions of different methods that can identify research gaps. These methods will also guide the development of a qualitative study to explore key stakeholders' perceived needs in identifying, communicating and displaying gaps in research.

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REVIEW

A scoping review describes methods used to identify, prioritize and display gaps in health research

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Abstract

Background and Objectives: Different methods to examine research gaps have been described, but there are still no standard methods for identifying, prioritizing, or reporting research gaps. This study aimed to describe the methods used to identify, prioritize, and display gaps in health research.

Methods: A scoping review using the Arksey and O'Malley methodological framework was carried out. We included all study types describing or reporting on methods to identify, prioritize, and display gaps or priorities in health research. Data synthesis is both quantitative and qualitative.

Results: Among 1,938 identified documents, 139 articles were selected for analysis; 90 (65%) aimed to identify gaps, 23 (17%) aimed to determine research priorities, and 26 (19%) had both aims. The most frequent methods in the review were aimed at gap identification and involved secondary research, which included knowledge synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80, 73%). Among 49 studies aimed at research prioritization, the most frequent methods were both primary and secondary research, accounting for 24 (49%) reports. Finally, 52 (37%) articles described methods for displaying gaps and/or priorities in health research.

Conclusion: This study provides a mapping of different methods used to identify, prioritize, and display gaps or priorities in health research.

Keywords: Evidence synthesis; Knowledge synthesis; Scoping review; Evidence mapping; Gaps in health research; Treatment uncertainties; Research gaps; Research priorities; Displaying gaps; Evidence gap maps

1. Introduction

The current body of research is growing, with more than 1 million clinical research papers published from clinical trials alone [1]. Planning a study focusing on the wrong question is a frequent cause of waste in research [2]. Hence, completed and on-going research should be used to assess whether research gaps justify new research and can inform the design, conduct, and reporting of further research [3]. Initiatives such as the James Lind Alliance (JLA), UK Database of Uncertainties about the Effects of Treatments, Cochrane Agenda and Priority Setting Methods Group and Evidence-based Research Network are some examples of existing efforts to identify and prioritize research gaps in health.

The term ''research gap'' is not standardized, and its meaning can differ depending on the research context. In this study, we adopted the definition from the National

Ethics approval and consent to participate: This study did not need ethics approval because it used secondary data.

Conflict of interest statement: The authors have declared that no conflict of interest exists.

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What is new?

Key findings

- We identified 12 different definitions of the term research gaps. We found seven specific methods for identifying research gaps involving secondary and primary research: quantitative survey, academic crowd-sourcing, needs assessment, knowledge synthesis, bibliometric study, priority setting, and global evidence-mapping methods. We found five specific methods for determining research priorities involving secondary and primary research: Delphi survey, quantitative survey, knowledge synthesis, priority setting, and global evidence mapping method. We also identified 14 unique methods used to display research gaps and/or research priorities.

What this adds to what was known?

- This is the first study to describe methods used to identify research gaps, determine research priorities, and display research gaps or research priorities.

What is the implication and what should change now?

- The term ''research gap'' is not standardized, and its meaning can differ depending on the research context.
- The study findings can be adopted to inform the development of standardized methods to identify, prioritize, and display gaps in health research.
- We propose convening an international group of leaders in the field to clarify the methods for identifying, prioritizing, and displaying gaps in health research.

Collaborating Center for Methods and Tools in Canada, which describes a research gap as a research question for which missing or insufficient information limits the ability to reach a conclusion [4]. To further understand research gaps and their causes, we also referred to an article by Robinson et al. that developed a framework on identifying research gaps from systematic reviews by characterizing the gap with use of PICOS (population, intervention, comparison, outcomes, setting) elements and identifying reason(s) for why the gap exists, including insufficient or imprecise information, biased information, inconsistency or unknown consistency, and incorrect information [5].

On identifying research gaps, prioritizing research based on the gaps is essential to determine its importance and relevance, especially based on feedback from key stakeholders such as patients, clinicians, researchers, advocates, and funders. Research priority setting is not commonly defined in a consistent way, although it has been described as any interpersonal activity that leads to the selection of topics or key questions to further investigate [6]. Research prioritization processes can help researchers and policy-makers effectively target research that has the greatest potential health benefit.

Consensus is lacking on what constitutes the best methodological approaches to identify research gaps [5,7], determine research priorities [6,8] and display research gaps or priorities. Therefore, we considered that a scoping review on this topic area was warranted. Our objectives were to (1) identify different definitions reported on the term ''research gap''; (2) explore methods used to identify research gaps; (3) describe methods used to determine research priorities; and (4) map methods used to display research gaps or research priorities.

2. Materials and methods

The analytic framework for this scoping review involved the methodology outlined by Arksey and O'Malley [9] and further refined by the Joanna Briggs Institute [10]. It entails identifying the research question; expert consultation on conceptualizing the research topic, identifying the different key terms for the search strategy, developing the items for the data extraction form and reviewing the article; searching for relevant studies using key terms; selecting studies; charting the data; collating, summarizing, and reporting the results; and consulting with stakeholders to inform study findings. Experts played a major role in this study; their role was important because of the uncharted nature of this topic area. A detailed study protocol is included as supplementary material.

2.1. Search methods for identification of documents

The scoping review aimed to identify and include a wide range of article types, including original research, protocols, conference proceedings, and website content. The goal of the search strategy was to identify a diversity of methods used to identify, prioritize, and display gaps or priorities in health research. To build the search terms for the search strategy, because of the variability in terminology used, we began by contacting experts for the terms, descriptions, and definitions they use to refer to research gaps. On compiling different terms, we built our search terms with the assistance of a research librarian. The final search terms included ''identifying gaps in research'', ''research gaps,'' ''evidence gaps,'' ''research uncertainties,'' ''research gaps identification,'' ''research gaps prioritization,'' and ''methods'' in health research including public health and clinical research. Two reviewers (LN and VN) conducted the searches by using the Peer Review of Electronic Search

Strategies (PRESS) guideline checklist, which aims to improve the quality of database searches [11].

The databases searched were MEDLINE, PubMed, EMBASE, Cochrane Library, Scopus, Web of Science, PROSPERO register, TRIP, Google Scholar, and Google. To focus on the most current research, database searches were limited to the past 10 years (2007–2017). Additional searches involved hand searches, web searches, expert suggestions, and checking reference lists of highly relevant articles. Only studies reported in English and involving humans were included to increase the feasibility of this scoping review. See Appendix B for complete search strategies.

2.2. Eligibility criteria

Studies were included if they aimed to describe a methodology and/or applied some methodology to identify gaps, determine research priorities, and/or display gaps or priorities in health research. All study designs were eligible, including those that used qualitative or quantitative methods, methodology, or guideline reports. We focused our inclusion criteria to capture reports scoping within the domain of health, reporting on and/or describing methods for identifying, prioritizing and/or displaying research gaps. We excluded publications that did not explicitly describe how they aimed to identify, prioritize, and/or display research gaps. For addition information see Appendix C.

2.3. Abstract and full-text screening and selection of articles

Abstract and full-text screening was performed by two authors. The first reviewer (LN) performed the entire screening of 1,938 abstracts, and a second reviewer (VN), screened 10% (194/1,938) of all abstracts. Agreement on selection of abstracts was 174/194 (90%). In total, 237 articles were selected for full-text screening. LN performed the entire screening and VN screened 10% (24/237) of articles. Among the 24 articles that were double screened, agreement was reached on 20 (85%). Title and abstract screening involved use of the software package Covidence for conducting systematic reviews. Full-text screening involved using EndNote to manage and retrieve full texts.

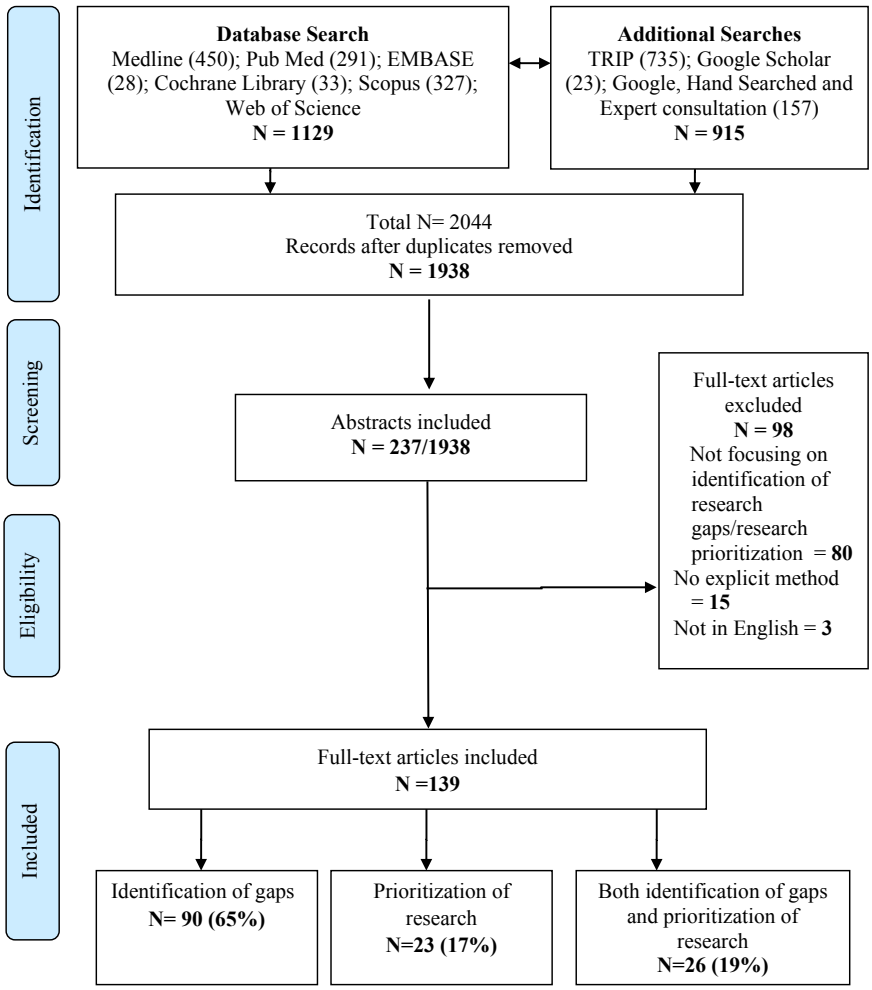


Fig. 1. Scoping review flow diagram.

Box 1 Definitions of research gaps as reported by the scoping review

Missing information

Research/evidence/knowledge gaps

- Evidence is missing from a body of research on a particular topic that could otherwise potentially answer the questions of decision-makers (clinicians, other practitioner groups, administrators, policy-makers) [14]

Synthesis/unidentified gaps

- Little or no evidence from systematic reviews is available and could be a valuable resource to inform the evidence base in a particular area [15,16]
- Lack of up-to-date and conclusive systematic reviews at low risk of bias mapped to a clinical question [17]

Treatment uncertainty

- Lack of up-to-date, reliable systematic reviews of research evidence addressing the uncertainty about the effects of treatment, and/or up-to-date systematic reviews of research evidence show that uncertainty exists [18]

Absolute evidence gaps

- Little or no evidence from primary studies is available [15]

Knowledge gap (knowledge void gap)

- Desired research findings do not exist [10,19,20]

Practical knowledge gap (action–knowledge conflict gap)

- Professional behavior or practices deviate from research findings or are not covered by research [10,19,20]

Inadequate information

Research/evidence/knowledge gaps

- The evidence base inadequately addresses a key question [21]

Empirical gap (evaluation void gap)

- Research findings or propositions need to be evaluated or empirically verified [10,19,20]

Population gap

- Research regarding a population that is not adequately represented or under-researched in the evidence base or prior research (e.g., sex, race/ethnicity, age) [5]

Insufficient information

Research/evidence/knowledge gaps

- Not much information is available and/or there is a lot of uncertainty about the accuracy of the existing estimates/evidence [7]
- Additional research is needed, from policy-makers perspectives, to address the evidence gap in the available primary research [14]

Methodological gap (method and research design gap)

- A variation in research methods is required to generate new insights or to avoid distorted findings [10,19,20]

Theoretical gap (theory-application void gap)

- Theory should be applied to certain research issues to generate new insights; theory is lacking, so a gap exists [10,19,20]

Discrepancies in both abstract and full-text screening were resolved in a meeting with senior researchers.

2.4. Data charting and synthesis

Data charting involved the use of Google Forms developed by LN with expert consultation, guidance from senior researchers and reviewing a previous methodological study [12]; the form was calibrated by LN and VN. See Appendix D for the complete data extraction form. All data collection and analyses were conducted by LN, and VN extracted 10% (14/139 of all full-text articles). As an additional data cleaning step, two senior researchers then verified and discussed the 14 articles extracted by the second reviewer, to ensure data accuracy. A 95% agreement was achieved; disagreements were mainly on interpretation of methods used to identify gaps and/or determine research priorities. Disagreements were resolved in a meeting with

senior researchers. We extracted the following data from articles: type of article, main objective of the study, main study methodology, definition of research gaps, and specific methods to identify research gaps, determine research priorities and display research gaps or priorities [12]. The synthesis included quantitative analysis (i.e., frequency analysis) and qualitative analysis (i.e., thematic analysis) of the components of the methods to identify, prioritize, and/or display gaps in health research and conceptual definitions of gaps in health research [13].

3. Results

3.1. Results of the search

The literature search retrieved 2,044 citations, and after duplicates were removed, 1,938 remained. Overall, 247 references were considered potentially eligible. After full-text assessment, 98 articles were excluded, and 139 were included. Fig. 1 shows the flow chart of articles through the scoping review.

3.2. Summary of study designs

Among the 139 included articles, 90 (65%) aimed to identify gaps, 23 (17%) aimed to determine research priorities, and 26 (19%) focused on both identifying gaps and determining research priorities.

3.3. Definitions of research gaps reported in articles

We explored the definitions as reported in the included studies. We identified a total of 12 different definitions, some of which overlapped, as presented in Box 1. Three cross-cutting themes were identified: definitions related to missing information, inadequate information, and insufficient information.

3.4. Specific methods for identifying research gaps

We then classified the methods used to identify research gaps. The most frequent methods in the review were aimed at gap identification (including both identification and prioritization) and involved secondary research, including knowledge synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80, 73%) (Table 1). Overall, 24/116 (21%) articles described the use of both primary and secondary research and (12/116, 10%) only primary research. We found seven specific methods for identifying research gaps that we describe along with the purpose of the method (Table 2).

Table 1. Study designs used

Study design	All articles (*N* = 139)	Identification of research gaps (*N* = 90)	Research prioritization (*N* = 23)	Both identification and prioritization (*N* = 26)
Primary research	25 (18%)	8 (9%)	13 (56%)	4 (15%)
Qualitative study	3 (12%)	1 (13%)	2 (15%)	0
Quantitative survey	2 (8%)	1 (12%)	2 (15%)	1 (25%)
Both qualitative study and quantitative survey	20 (80%)	6 (75%)	9 (69%)	3 (75%)
Secondary research	85 (61%)	77 (86%)	5 (22%)	3 (12%)
Knowledge synthesis				
Systematic review[a]	36 (42%)	33 (43%)	1 (20%)	2 (67%)
Scoping review	25 (29%)	23 (30%)	2 (40%)	0
Evidence mapping	4 (4%)	3 (4%)	0	1 (33%)
Mapping study	2 (2%)	2 (3%)	0	0
Literature review	4 (4%)	4 (5%)	0	0
Umbrella review	4 (4%)	4 (5%)	0	0
Other (integrative review, critical interpretive synthesis)	8 (9%)	6 (8%)	2 (40%)	0
Bibliometric analysis	2 (1%)	2 (3%)	0	0
Both primary and secondary research	29 (21%)	5 (6%)	5 (22%)	19 (73%)
Review of evidence and quantitative study	6 (21%)	0	1 (20%)	5 (26%)
Review of evidence and qualitative study	3 (10%)	2 (40%)	0	1 (5%)
Review of evidence and both quantitative and qualitative study	20 (69%)	3 (20%)	4 (80%)	13 (68%)

[a] Including methods used in Health Technology Assessments.

Table 2. Overview of specific methods to identify research gaps

Methods to identify research gaps	Definition	Purpose
Primary research methods		
Quantitative survey [17]	A scientific procedure for collecting information and making quantitative inferences about a predefined population	Determine evidence gaps by using a Likert-type response scale and scoring from 0 (not important at all) to 10 (highly important)
Academic crowd- sourcing [7]	An emerging paradigm that is based on harnessing the power of the crowd to solve problems [7]	Aims to reach a wider range of people, which may sometimes be required to solve a problem correctly and efficiently including identifying research gaps [7]
Needs assessment [22,23]	A systematic process for determining and addressing needs, or ''gaps'' between current conditions and desired conditions or "wants'' by using various techniques including primary or secondary research methods (e.g., reviewing evidence, guidelines, and conducting interviews) [19]	Clarify problems and identify appropriate interventions or solutions [19]
Secondary research methods		
Knowledge synthesis		
Systematic review [14,24–27][a]	Efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored [28]	Summarize all pertinent studies on a specific question; improve the understanding of inconsistencies in diverse evidence and identify gaps in research evidence to define future research agendas [28]
Scoping review [12,29,30]		
Evidence mapping [15,31,32]		
Mapping study [33–35]		
Umbrella review [6]		
Integrative review [36]		
Critical interpretive synthesis [37]		
Bibliometric study [38,39]	The quantitative study of bibliographic material used to examine the knowledge structure and development of research fields based on analysis of related publications [40]	Provide a general picture of a research field that can be classified by papers, authors, and journals [40]
Both primary and secondary research methods		
Priority setting [17,18,41–51]		
James Lind Alliance priority setting partnership (JLA PSP), Cochrane Priority Setting (*consists of four steps: the first two aim at gap identification and the last two aim at research prioritization*)	JLA PSP methods were designed to allow clinicians, patients, and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that could be answered by research by gathering research questions, checking existing research evidence, interim prioritization, and a final consensus meeting to reach agreement on the top 10 research priorities [41]	Raise awareness of research questions that are of direct relevance and potential benefit to patients and the clinicians who treat them, to lead to changes in how research funding is granted [41]
Global evidence-mapping methods [31,52]	Maps available research and provides an overview of a broad range of research questions and identifies evidence gaps [31]	Characterize the breadth, depth, methodology of relevant evidence and make this readily accessible [53]; identify research gaps

Italics represents an additional information on method.
[a] Including methods used in Health Technology Assessments.

3.5. Specific methods for determining research priorities

Among the 49 studies aiming at research prioritization, the most frequent method involved both primary and secondary research, accounting for 24 (49%) studies, followed by primary research 17 (35%), and secondary research 8 (16%) (Table 1). We identified five specific methods for determining research priorities (Table 3).

3.6. Specific methods for displaying research gaps and research priorities

We identified 14 unique methods used to display research gaps and/or research priorities and give some

Table 3. Overview of specific methods to determine research priorities

Methods to determine research priorities	Description	Summary of steps if specified
Primary research		
Delphi survey [51,54–56]	A group facilitation technique that seeks to obtain consensus on the opinions of relevant stakeholders by a series of structured questionnaires (commonly referred to as rounds). The questionnaires are completed anonymously by the experts (commonly referred to as panelists, participants, or respondents) [41]	Involves a series of questionnaires that are completed anonymously by experts. A process of group communication without the group ever meeting face to face. The responses from each set of questionnaires are analyzed, summarized, and then sent back to the participants until a large degree of consensus is reached in the area of interest.
Quantitative survey [17]	Adapted to determine participant research priorities by using forced raking of research questions and Likert-type scale for responses	Developing and testing questionnaires to address research questions Forced ranking of research questions Likert-type scale
Secondary research		
Knowledge synthesis	Efficient scientific approach for identifying and summarizing evidence that allows for assessing the generalizability and consistency of research findings and exploring data inconsistencies [28]	Summarize all pertinent studies on a specific question; can improve the understanding of inconsistencies in diverse evidence, and identify gaps in research evidence to define future research agendas [28]
Systematic review [25]ª		
Scoping review [29,30]		
Both primary and secondary research		
Priority setting [1,17,18,42–44,46,47,49,50,54,57–65] *Example: JLA PSP methods*	Designed to enable clinicians, patients, and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that could be answered by research [18]	Survey to identify treatment uncertainties Review of existing systematic reviews to explore existing evidence and address treatment uncertainties Interim prioritization to identify the priorities of relevant individuals and stakeholder groups Focus groups to discuss the research priorities based on missing or inadequate evidence A final consensus meeting to reach agreement on the top 10 research priorities [18]
Global evidence-mapping method [31,52]	Maps available research and provides an overview of a broad range of research questions and identifies evidence gaps [31]	Question development involving expert consultation, preliminary literature search, mapping workshop, online survey, and development of clinical question Question prioritization Evidence search and selections

Italics represents an example of method.
[a] Including methods used in Health Technology Assessments.

examples of these methods (Table 4). We provide some illustrations of nontraditional methods. An illustration of all methods can also be found in Appendix E.

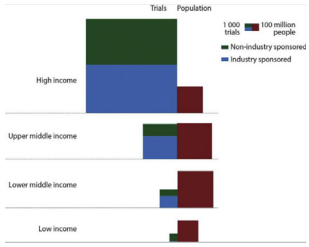
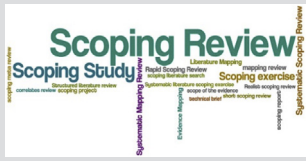
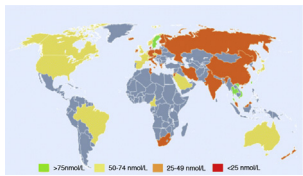
4. Discussion

Our findings demonstrate that the term ‘‘research gap’’ significantly differs across research contexts, and there is no common definition. It also reveals no clear methodological guidance on which methods should be used to identify research gaps or determine research priorities. This situation leads to a wide variety in methodology, for difficulties in comparing results across studies.

Also, many studies aimed at identifying gaps relied on secondary research, primarily systematic reviews. Systematic reviews are considered the gold standard in providing the highest level of evidence for the relative efficacy and safety of interventions [83] and summarizing the overall quality and results of research. A study on identifying and prioritizing research gaps corroborated that systematic reviews are the standard for evaluating the existing state of scientific knowledge regarding a specific clinical or policy question [79]. Robinson et al. [5] also developed a framework for using systematic reviews to identify research gaps. Although these two studies show that systematic reviews can identify research gaps, most systematic reviews address a highly focused question related to the existing evidence and thus present difficulties for explicitly identifying research gaps in a general area [5,79,84].

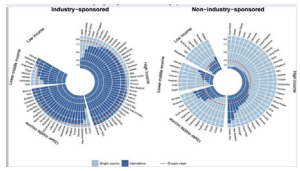
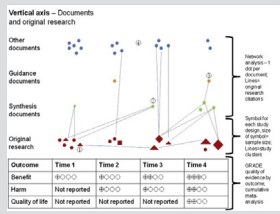
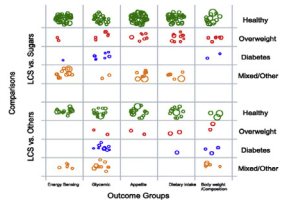
Other secondary research methods identified in this review were overviews of reviews, also known as umbrella reviews, scoping reviews, and evidence mapping.

Table 4. Overview of methods to display gaps and research priorities

Format	The information on research gaps and priorities displayed	Ways of displaying research
Table [1,14,17,18,23,30,31, 41,45,47–49,52,58,66,67]	List of clinical questions, gaps, and research priorities Ranking quality of evidence Study designs to address research questions Scoring of each research gap List of research questions Prioritization of research questions Metric of ranking information	Table format[a]
Box plot [17]	List of research questions Ranking of research questions	Box plot format[a]
Bar graph/horizontal bar graph [17,33,61,68–74]	List of research priorities Frequency of questions prioritized Number of studies and categories studied Frequency of research questions Quality of evidence metric	Bar graph format[a]
Scatter plot [75]	Numeric values of desired research and current research	Scatter plot format[a]
Funnel plot [76]	Number of studies included in the review Effect sizes of studies on the x-axis	Funnel plot format[a]
Pie chart [70,77]	Proportional size of health problems being investigated by trials registered in a registry platform	Pie chart format[a]
Mind maps [78]	Diagram used to represent concepts, ideas, or tasks linked to and arranged radially around a central key word or idea Primary branches represent the major ideas or themes around the central topic, and secondary branches tend to include more concrete illustrative examples	Mind map format[a]
Tree map chart [33]	Number of clinical trials, population, and income group Comparison study of clinical registry data vs. global health research data from the Global Burden of Disease	 [33]
Word cloud [12]	Frequency of words Frequency of words between two groups	 [12]
Geographic map [24,79]	Studies mapped around the world using colors on a predefined health outcome. Different shapes and sizes also used for additional information on a map.	 [24]

(*Continued*)

Table 4. Continued

Format	The information on research gaps and priorities displayed	Ways of displaying research
Dot plot [80]	Number of studies Quality of evidence Different colors to show different study designs	 [80]
Radial bar plot/polar histogram [33]	Proportion of trials in several countries using one color per country among two groups over a period of time Comparison of proportion of trials in several countries using one color per country among two groups over a period of time	 [33]
Schematic representation [38]	Horizontal axis represents time, and vertical axis represents different documents	 [38]
Bubble plot/chart [15,81,82]	Bubbles represent studies; size indicates the relative number of studies and color the study design Number of studies by intervention type and health status Compares three sets of values	 [82]

[a] Examples available on Appendix E.

Overviews of reviews focus on a much broader area, compiling evidence from multiple reviews into one accessible and usable document and highlighting other reviews within the specified topic area [85,86]. Moreover, scoping reviews and evidence mapping are designed to describe existing evidence in a broader content area [82,87,88]. They descriptively summarize results, which can be presented in a user-friendly format, often a visual figure called an evidence gap map [15], or a searchable database, to improve research planning, strategic research prioritization, and evidence-informed policies.

A mix of primary and secondary research was most frequently used to determine research prioritization, namely, priority setting (e.g., JLA PSP methods). These methods involve the participation of patients, caregivers, and health and social care professionals in identifying research questions, then prioritizing them by using a combination of primary and secondary research [1,17,18,42–44,46,47,49,50,54,57–65]. The main method for determining research prioritization with primary research was the Delphi survey, which is a practical and productive approach to obtaining opinions from a wide number of relevant experts for identifying potential priority topic areas for research [51,54–56].

To display research gaps, half of the methods still used traditional ways to present findings (e.g., summary table and bar charts), and the other half used more advanced ways to display information (e.g., tree map charts, radial

bar plots, and bubble plots).The nontraditional methods used more colors and diagrams in presenting research gaps or priorities. For example, bubble plots use different shapes, sizes, and colors to display information and can be used to present up to three different variables in one diagram. These characteristics could be further explored to determine the appropriate method to be used along with specified methods to identify research gaps and/or determine research priorities.

Finally, our study confirmed that the various methods identified consist of both emerging and established approaches. Nonetheless, these methods can provide rich contextual details for establishing methodological guidance. We propose more work to improve the understanding of the methods and investigate ways to give the public, patients, clinicians, health researchers, decision-makers, and funders more opportunities to know what methodologies are available and can be used.

Our scoping review had some limitations. How the term ‘‘research gap’’ is used and defined varied widely among different authors and articles, and our exploration of methods for identifying research gaps, determining research priorities and displaying research gaps and priorities relied on definitions used by authors of included studies. Therefore, we may have missed some methods because of lack of clear definitions. Also, we included only documents written in English, relying on key articles to identify the steps involved in each method and as presented by the authors of the included articles. As such, we may have missed important methods that were published in other languages. In addition, the time restriction to the last 10 years provided a comprehensive list of recent methods used rather than an exhaustive list of all methods used.

Finally, we anticipate that our results will be of interest to knowledge users, including patients, public, clinicians, researchers, decision-makers, funders, key organizations such as JLA, the UK Database of Uncertainties about the Effects of Treatments, Joanna Briggs Institute, Campbell Collaboration, Africa Evidence Network, Cochrane Priority Setting Methods Group and Evidence-based Research Network, and finally methodologists focused on identifying and displaying gaps, and determining priorities in health research.

5. Conclusion

This study provides an overview of different methods used for and/or reporting on identifying gaps, determining research priorities and displaying both gaps and research priorities. The findings can be adopted to inform the development of standardized methods to identify, prioritize, and display gaps. They can inform further research and evidence-based decision-making by providing descriptions of different methods that can be adopted in identifying research gaps. These methods will also guide the development of a qualitative study to explore key stakeholders’ perceived needs in identifying, communicating, and displaying gaps in research.

CRediT authorship contribution statement

Linda Nyanchoka: Conceptualization, Writing - review & editing, Formal analysis, Writing - original draft, Data curation. **Catrin Tudur-Smith:** Conceptualization, Writing - review & editing, Formal analysis. **Van Nguyen Thu:** Formal analysis, Writing - review & editing, Data curation. **Valentia Iversen:** Writing - review & editing. **Andrea C. Tricco:** Writing - review & editing. **Raphaël Porcher:** Conceptualization, Writing - review & editing, Formal analysis.

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Supplementary data

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References

[1] Ioannidis JPA. Why most clinical research is not useful. PLoS Med 2016;13(6). e1002049.
[2] Chalmers I, Glasziou P. Avoidable waste in the production and reporting of research evidence. Lancet 2009;374(9683):86−9.
[3] Young C, Horton R. Putting clinical trials into context. Lancet 2005; 366(9480):107−8.
[4] Framework for identifying research gaps. Hamilton, ON: McMaster University. http://www.nccmt.ca/resources/search/118.
[5] Robinson KA, Saldanha IJ, McKoy NA. Development of a framework to identify research gaps from systematic reviews. J Clin Epidemiol 2011;64:1325−30.
[6] Nasser M. Setting priorities for conducting and updating systematic reviews. UK: Peninsula Schools of Medicine and Dentistry, University of Plymouth; 2018.
[7] Rudan I, Campbell H, Marusic A, Sridhar D, Nair H, Adeloye D, et al. Assembling GHERG: could "academic crowd-sourcing" address gaps in global health estimates? J Glob Health 2015;5(1): 010101.

[8] Viergever RF, Olifson S, Ghaffar A, Terry RF. A checklist for health research priority setting: nine common themes of good practice. Health Res Policy Syst 2010;8:36.
[9] Arksey H, O'Malley L. Scoping studies: towards a methodological framework. Int J Soc Res Methodol 2005;8(1):19−32.
[10] Peters MD, Godfrey CM, Khalil H, McInerney P, Parker D, Soares CB. Guidance for conducting systematic scoping reviews. Int J Evid Based Healthc 2015;13(3):141−6.
[11] McGowan J, Sampson M, Salzwedel DM, Cogo E, Foerster V, Lefebvre C. PRESS peer review of electronic search strategies: 2015 guideline statement. J Clin Epidemiol 2016;75:40−6.
[12] Tricco AC, Lillie E, Zarin W, O'Brien K, Colquhoun H, Kastner M, et al. A scoping review on the conduct and reporting of scoping reviews. BMC Med Res Methodol 2016;16:15.
[13] Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. Prisma extension for scoping reviews (prisma-scr): checklist and explanation. Ann Intern Med 2018;169:467−73.
[14] Scott NA, Carmen M, Christa H, Jacques M. Using health technology assessment to identify research gaps: an unexploited resource for increasing the value of clinical research. Healthc Policy 2008; 3(3):e109−27.
[15] Birte S, Martina V, Ami B, Jennifer S, Marie G. Evidence & gap maps: a tool for promoting evidence informed policy and strategic research agendas. J Clin Epidemiol 2016;79:120−9.
[16] Brearley SG, Stamataki Z, Addington-Hall J, Foster C, Hodges L, Jarrett N, et al. The physical and practical problems experienced by cancer survivors: a rapid review and synthesis of the literature. Eur J Oncol Nurs 2011;15(3):204−12.
[17] Yu T, Li T, Lee KJ, Friedman DS, Dickersin K, Puhan MA. Setting priorities for comparative effectiveness research on management of primary angle closure: a survey of Asia-Pacific clinicians. J Glaucoma 2015;24:348−55.
[18] van Middendorp JJ, Allison HC, Ahuja S, Bracher D, Dyson C, Fairbank J, et al. Top ten research priorities for spinal cord injury: the methodology and results of a British priority setting partnership. Spinal Cord 2016;54(5):341−6.
[19] Fulgham SM, Shaughnessy MF. O & a with ed tech leaders: interview with roger kaufman. Educ Technol 2008;48(5):49.
[20] Müller-Bloch C, Kranz J. A framework for rigorously identifying research gaps in qualitative literature reviews. In: Proceedings of the 36th International Conference on Information Systems, Fort Worth, Texas, USA. 2015.
[21] Hiten DP, Emmanuel I, Phillip MP, Stephen MS, Michael HJ, Ritu S, et al. A systematic review of research gaps in the evaluation and management of localized renal masses. Urology 2016;98:14.
[22] Kitson A, Straus SE. The knowledge-to-action cycle: identifying the gaps. Can Med Assoc J 2010;182(2):E73−7.
[23] Thompson A, Brennan K, Cox A, Gee J, Harcourt D, Harris A, et al. Evaluation of the current knowledge limitations in breast cancer research: a gap analysis. Breast Cancer Res 2008;10(2):R26.
[24] Wahl DA, Cooper C, Ebeling PR, Eggersdorfer M, Hilger J, Hoffmann K, et al. A global representation of vitamin D status in healthy populations. Arch Osteoporos 2012;7:155−72.
[25] Chang SM, Carey TS, Kato EU, Guise JM, Sanders GD. Identifying research needs for improving health care. Ann Intern Med 2012; 157:439−45.
[26] Mickenautsch S. Research gaps identified during systematic reviews of clinical trials: glass-ionomer cements. BMC Oral Health 2012; 12(1):18.
[27] Tunis SR, Turkelson C. Using health Technology assessment to identify gaps in evidence and inform study design for comparative effectiveness research. J Clin Oncol 2012;30:4256−61.
[28] Mulrow CD. Rationale for systematic reviews. BMJ 1994;309:597−9.
[29] Levac D, Colquhoun H, O'Brien KK. Scoping studies: advancing the methodology. Implement Sci 2010;5:69.
[30] Schwartz SR, Baral S. Fertility-related research needs among women at the margins. Reprod Health Matters 2015;23(45):30−46.
[31] Clavisi O, Bragge P, Tavender E, Turner T, Gruen RL. Effective stakeholder participation in setting research priorities using a global evidence mapping approach. J Clin Epidemiol 2013;66:496−502.e492.
[32] Schmucker C, Motschall E, Antes G, Meerpohl JJ. [Methods of evidence mapping. A systematic review]. Bundesgesundheitsblatt Gesundheitsforschung Gesundheitsschutz 2013;56(10):1390.
[33] Atal I, Trinquart L, Porcher R, Ravaud P. Differential globalization of industry- and non-industry-sponsored clinical trials. PLoS One 2015;10:e0145122.
[34] Ahmad N, Boutron I, Dechartres A, Durieux P, Ravaud P. Geographical representativeness of published and ongoing randomized controlled trials. The example of: tobacco consumption and HIV infection. PLoS One 2011;6:e16878.
[35] Ndounga Diakou LA, Ntoumi F, Ravaud P, Boutron I. Published randomized trials performed in Sub-Saharan Africa focus on high-burden diseases but are frequently funded and led by high-income countries. J Clin Epidemiol 2017;82:29−36.e26.
[36] Marion LM, Fiona C, Susanne K, Renee S, Jenny M, Toni D. Patient, family-centred care interventions within the adult ICU setting: an integrative review. Aust Crit Care 2016;29(4):179.
[37] Johnson M, Tod AM, Brummell S, Collins K. Prognostic communication in cancer: a critical interpretive synthesis of the literature. Eur J Oncol Nurs 2015;19(5):554−67.
[38] Kho ME, Brouwers MC. The systematic review and bibliometric network analysis (SeBriNA) is a new method to contextualize evidence. Part 1: description. J Clin Epidemiol 2012;65:1010−5.
[39] Mbuagbaw L, Kredo T, Welch V, Mursleen S, Ross S, Zani B, et al. Critical EPICOT items were absent in cochrane human immunodeficiency virus systematic reviews: a bibliometric analysis. J Clin Epidemiol 2016;74:66−72.
[40] Merigó JM, Yang J-B. A bibliometric analysis of operations research and management science. Omega 2017;73(Supplement C):37−48.
[41] Boney O, Bell M, Bell N, Conquest A, Cumbers M, Drake S, et al. Identifying research priorities in anaesthesia and perioperative care: final report of the joint national Institute of academic anaesthesia/-james lind alliance research priority setting partnership. BMJ Open 2015;5(12):e010006.
[42] Buckley BS, Grant AM, Glazener CM. Case study: a patient-clinician collaboration that identified and prioritized evidence gaps and stimulated research development. J Clin Epidemiol 2013;66: 483−9.
[43] Buckley BS, Grant AM, Tincello DG, Wagg A, Firkins L. Reaching a consensus and ranking research priorities in urinary incontinence. Nurs Times 2010;106(24):36−7.
[44] Heazell AE, Whitworth MK, Whitcombe J, Glover SW, Bevan C, Brewin J, et al. Research priorities for stillbirth: process overview and results from UK stillbirth priority setting partnership. Ultrasound Obstet Gynecol 2015;46(6):641−7.
[45] Knight SR, Metcalfe L, O'Donoghue K, Ball ST, Beale A, Beale W, et al. Defining priorities for future research: results of the UK kidney transplant priority setting partnership. PLoS One 2016;11: e0162136.
[46] Welsh E, Stovold E, Karner C, Cates C. Cochrane Airways Group reviews were prioritized for updating using a pragmatic approach. J Clin Epidemiol 2015;68:341−6.
[47] Rees SE, Chadha R, Donovan LE, Guitard AL, Koppula S, Laupacis A, et al. Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus. Can J Diabetes 2017;41:156−63.
[48] Jennifer MG, Evan RM, Kristine MS, Douglas CM, Remy RC, Matthew JC, et al. Prioritization of patient-centered comparative effectiveness research for osteoarthritis. Ann Intern Med 2014; 160:836.
[49] Ingram JR, Abbott R, Ghazavi M, Alexandroff AB, McPhee M, Burton T, et al. The hidradenitis suppurativa priority setting partnership. Br J Dermatol 2014;171:1422−7.

[50] Gadsby R, Snow R, Daly AC, Crowe S, Matyka K, Hall B, et al. Setting research priorities for Type 1 diabetes. Diabet Med 2012; 29:1321−6.
[51] Li T, Vedula SS, Scherer R, Dickersin K. What comparative effectiveness research is needed? A framework for using guidelines and systematic reviews to identify evidence gaps and research priorities. Ann Intern Med 2012;156:367−77.
[52] Jaramillo A, Welch VA, Ueffing E, Gruen RL, Bragge P, Lyddiatt A, et al. Prevention and self-management interventions are top priorities for osteoarthritis systematic reviews. J Clin Epidemiol 2013; 66:503−510.e504.
[53] Katz DL, Williams AL, Girard C, Goodman J, Comerford B, Behrman A, et al. The evidence base for complementary and alternative medicine: methods of evidence mapping with application to CAM. Altern Ther Health Med 2003;9(4):22−30.
[54] Yoshida S. Approaches, tools and methods used for setting priorities in health research in the 21(st) century. J Glob Health 2016;6(1): 010507.
[55] Evelina C, Ludovic R, Amy C, Stephanie S, Xavier B. Cochrane systematic reviews are useful to map research gaps for decreasing maternal mortality. J Clin Epidemiol 2013;66:105.
[56] Mitchell RB, Hussey HM, Setzen G, Jacobs IN, Nussenbaum B, Dawson C, et al. Clinical consensus statement: tracheostomy care. Otolaryngol Head Neck Surg 2013;148(1):6−20.
[57] Wald HL, Leykum LK, Mattison ML, Vasilevskis EE, Meltzer DO. A patient-centered research agenda for the care of the acutely ill older patient. J Hosp Med 2015;10(5):318−27.
[58] Lophatananon A, Tyndale-Biscoe S, Malcolm E, Rippon HJ, Holmes K, Firkins LA, et al. The James Lind Alliance approach to priority setting for prostate cancer research: an integrative methodology based on patient and clinician participation. BJU Int 2011; 108:1040−3.
[59] Pollock A, St George B, Fenton M, Firkins L. Top 10 research priorities relating to life after stroke–consensus from stroke survivors, caregivers, and health professionals. Int J Stroke 2014;9(3):313−20.
[60] Meremikwu M, Udoh E, Nwagbara B, Effa E, Oringanje C, Edet B, et al. Priority setting for systematic review of health care interventions in Nigeria. Health Policy 2011;99(3):244−9.
[61] Mitnick CD, Rodriguez CA, Hatton ML, Brigden G, Cobelens F, Grobusch MP, et al. Programmatic management of drug-resistant tuberculosis: an updated research agenda. PLoS One 2016;11:e0155968.
[62] Pollock A, St George B, Fenton M, Crowe S, Firkins L. Development of a new model to engage patients and clinicians in setting research priorities. J Health Serv Res Policy 2014;19:12−8.
[63] van Furth EF, van der Meer A, Cowan K. Top 10 research priorities for eating disorders. Lancet Psychiatry 2016;3(8):706−7.
[64] Chapman E, Reveiz L, Sangalang S, Manu C, Bonfill X, Munoz S, et al. A survey study identified global research priorities for decreasing maternal mortality. J Clin Epidemiol 2014;67:314−24.
[65] Gierisch JM, Myers ER, Schmit KM, Crowley MJ, McCrory DC, Chatterjee R, et al. Prioritization of research addressing management strategies for ductal carcinoma in situ. Ann Intern Med 2014;160:484−91.
[66] Knight R, Small W, Pakula B, Thomson K, Shoveller J. A scoping study to identify opportunities to advance the ethical implementation and scale-up of HIV treatment as prevention: priorities for empirical research. BMC Med Ethics 2014;15:54.
[67] Sun C, Dohrn J, Omoni G, Malata A, Klopper H, Larson E. Clinical nursing and midwifery research: grey literature in African countries. Int Nurs Rev 2016;63(1):104−10.
[68] Wan YL, Beverley-Stevenson R, Carlisle D, Clarke S, Edmondson RJ, Glover S, et al. Working together to shape the endometrial cancer research agenda: the top ten unanswered research questions. Gynecol Oncol 2016;143(2):287−93.
[69] DeFrank JT, Barclay C, Sheridan S, Brewer NT, Gilliam M, Moon AM, et al. The psychological harms of screening: the evidence we have versus the evidence we need. J Gen Intern Med 2015;30:242−8.
[70] Viergever RF, Terry RF, Karam G. Use of data from registered clinical trials to identify gaps in health research and development. Bull World Health Organ 2013;91(6):416−425C.
[71] Kumar MB, Wesche S, McGuire C. Trends in Metis-related health research (1980-2009): identification of research gaps. Can J Public Health 2012;103(1):23−8.
[72] De Luca Canto G, Pacheco-Pereira C, Aydinoz S, Major PW, Flores-Mir C, Gozal D. Biomarkers associated with obstructive sleep apnea and morbidities: a scoping review. Sleep Med 2015; 16(3):347−57.
[73] Tavender EJ, Bosch M, Fiander M, Knott JC, Gruen RL, O'Connor D. Implementation research in emergency medicine: a systematic scoping review. Emerg Med J 2016;33(9):652−9.
[74] Singh Ospina N, Rodriguez-Gutierrez R, Brito JP, Young WF Jr, Montori VM. Is the endocrine research pipeline broken? A systematic evaluation of the Endocrine Society clinical practice guidelines and trial registration. BMC Med 2015;13:187.
[75] Azeredo TB, Luiza VL, Oliveira MA, Emmerick IC, Bigdeli M. Stakeholders' perspectives on access-to-medicines policy and research priorities in Latin America and the Caribbean: face-to-face and web-based interviews. Health Res Policy Syst 2014;12:31.
[76] Umscheid CA. A primer on performing systematic reviews and meta-analyses. Clin Infect Dis 2013;57:725−34.
[77] Atal I, Zeitoun J-D, Névéol A, Ravaud P, Porcher R, Trinquart L. Automatic classification of registered clinical trials towards the global burden of diseases taxonomy of diseases and injuries. BMC Bioinformatics 2016;17(1):392.
[78] van den Eertwegh V, van Dulmen S, van Dalen J, Scherpbier AJ, van der Vleuten CP. Learning in context: identifying gaps in research on the transfer of medical communication skills to the clinical workplace. Patient Educ Couns 2013;90(2):184−92.
[79] Carey T, Yon A, Beadles C, Wines R. Prioritizing future research through examination of research gaps in systematic reviews. USA: Prepared for the Patient-Centered Outcomes Research Institute; 2012.
[80] Bhavisha V, Emma J, Iris G, Cova B, Elena S. Availability of evidence on cataract in low/middle-income settings: a review of reviews using evidence gap maps approach. Br J Ophthalmol 2016; 100:1455−60.
[81] Miake-Lye IM, Hempel S, Shanman R, Shekelle PG. What is an evidence map? A systematic review of published evidence maps and their definitions, methods, and products. Syst Rev 2016;5:28.
[82] Wang DD, Shams-White M, Bright OJ, Parrott JS, Chung M. Creating a literature database of low-calorie sweeteners and health studies: evidence mapping. BMC Med Res Methodol 2016;16:1.
[83] Impellizzeri FM, Bizzini M. Systematic review and meta-analysis: a primer. Int J Sports Phys Ther 2012;7(5):493−503.
[84] Tricco AC, Zarin W, Ghassemi M, Nincic V, Lillie E, Page MJ, et al. Same family, different species: methodological conduct and quality varies according to purpose for five types of knowledge synthesis. J Clin Epidemiol 2018;96:133−42.
[85] Nikolakopoulou A, Mavridis D, Furukawa TA, Cipriani A, Tricco AC, Straus SE, et al. Living network meta-analysis compared with pairwise meta-analysis in comparative effectiveness research: empirical study. BMJ 2018;360:k585.
[86] Grant MJ, Booth A. A typology of reviews: an analysis of 14 review types and associated methodologies. Health Info Libr J 2009;26:91−108.
[87] Althuis MD, Weed DL. Evidence mapping: methodologic foundations and application to intervention and observational research on sugar-sweetened beverages and health outcomes. Am J Clin Nutr 2013;98:755−68.
[88] Hetrick SE, Parker AG, Callahan P, Purcell R. Evidence mapping: illustrating an emerging methodology to improve evidence-based practice in youth mental health. J Eval Clin Pract 2010;16:1025−30.

Chapter 4: Key Stakeholders' Perspectives and Experiences with Defining, Identifying and Displaying Gaps in Health Research

4.1. Background

In the previous chapter, I described a scoping review of 139 scientific articles to identify research gaps, which can help inform the design and conduct of health research, practice and policies by providing a better understanding of the current body of evidence. The review showed that the term "research gap" has a variety of definitions, and its meaning can differ depending on the researcher and research context. Twelve different definitions related to gaps in health research (e.g., population, theoretical and methodology gaps), each describing research gaps differently [104]. This finding shows the ambiguity of the term "research gaps" and the different practices they may be related to.

As a basis for further exploring and understanding "research gaps", we started from the definition given by the NCCMT in Canada based on the work of Robinson et al., whereby a research gap is defined as a topic or area for which missing or insufficient information limits the ability to reach a conclusion for a question [2]. Given the different meanings and definitions of research gaps found in the scoping review [104], we consider it important to further explore definitions rather than just adopt or modify the NCCMT definition. Clearly defining the type of research gap can help determine how to better identify, characterize, prioritize and address research gaps.

Different methods for identifying research gaps have been reported; for example, scoping reviews and umbrella reviews are emerging methods for mapping and summarizing evidence. These methods have an explicit aim of identifying research gaps in a broad area as compared with systematic reviews, which focus on answering a specific research question [8, 21, 29, 39, 105].

Robinson et al. developed a framework using systematic reviews to identify research gaps [2]: they classified the reasons for the existence of research gaps and used the PICOS process to characterize them. Scoping, umbrella and systematic reviews are reported to specifically identify research gaps, but other methods are being used, and further exploring these methods can optimize their definition, methodological scrutiny, and practice [106-114]. Furthermore, the aforementioned methods focus on the use of secondary research methods to identify research gaps. However, a recent scoping review showed that other methods have been used to identify gaps, including primary and both primary and secondary research methods [104]. The scoping review showed a lack of consensus on what constitutes the best methodological approaches to identify research gaps, determine research priorities, and display research gaps or priorities [8, 21, 104]. Therefore, to better understand the different methods and ongoing practices, we aimed to conduct a qualitative study to further explore more in-depth key stakeholder experiences in describing research gaps and the methods used to identify and display gaps in health research.

Healthcare decisions for individual patients, public health policies, and clinical guidelines should be informed by the best available research evidence while taking into consideration research gaps. Investigating experiences with practices/methods used to identify research gaps can inform explicit methodological approaches in identifying and describing research gaps. This investigation can enhance practices of different stakeholder groups (i.e., health professionals, commissioners, researchers, patients/the public and decision-makers) when addressing areas of uncertainty within the research problem and topic area[115]. The aim of the study was to investigate the experience of key stakeholders (i.e., researchers, funders, clinicians, clinical guideline developers, public health professionals, commissioners, patients/the public and policy-makers) with defining research gaps and practices/methods used to identify and display research gaps. The final step was an

integration and overview combining findings from the scoping review and qualitative study to provide a comprehensive overview of methods used to identify and display research gaps. These study findings will be used to inform the methodological guidance on identifying research gaps.

The specific objectives of the study were to 1) investigate key stakeholders' knowledge, perceptions and experiences with defining research gaps and 2) characterize methods/practices used for identifying and displaying gaps in health research.

4.2. Method

4.2.1. Reflexivity

Reflexivity pertains to the "analytic attention to the researcher's role in qualitative research". It is both a concept and a process. As a concept, it refers to a certain level of consciousness[116, 117]. Prior to this PhD, I was a Public Health Advisor at the Norwegian National of Public Health in Oslo, Norway, focused on knowledge production for the health sector and providing knowledge about the health status of the population, influencing factors and how the status can be improved. During my years as a Public Health Advisor, I recognized the need for evidence to inform research planning, implementation and evaluation. Evidence was also requested to inform health policy and practices at short notice. At the time, in the department I was working in, knowledge production requests were not systematically produced, and with a very short timeline to complete such assignments, most did not have the rigour and systematic approach needed to adequately produce comprehensive evidence reports. Additionally, the reports were targeted to not only researchers and policy-makers but also the public, yet very little attention was given on how to involve the public in the evidence synthesis and knowledge production process. Therefore, I was motivated to further investigate how we can make the research we conduct, implement and evaluate more relevant to the public, particularly what research we prioritize and what research is needed. This

experience led me to start my PhD on meta research and more specifically on the topic of methods to identify research gaps. My past experience influenced my choice of research project and the conceptualization of this qualitative study.

4.2.2. Study design

4.2.3.1. Qualitative study design

I conducted an exploratory qualitative study using semi-structured interviews. This method was selected to provide in-depth understanding into key stakeholders' perspectives, experiences, and practices with defining, identifying and displaying research gaps. This method also ensures that we explore key stakeholders' understandings and practices related to identifying research gaps through a variety of lenses from different stakeholder groups. This, in turn, provides multiple facets of research gap definitions and methodological practices to identify and display gaps [118].

4.2.3.2. Sampling and recruitment

We used purposive sampling to ensure that the perspectives of all identified stakeholder groups were represented. Purposive sampling is widely used in qualitative research for identifying and selecting information-rich cases [23, 24]. The study sample included the following stakeholder groups: researchers, funders, healthcare providers, patients/the public and policy-makers. The stakeholder groups were determined from the findings of a previously conducted scoping review [104] and organized in three main categories focusing on the use of evidence to inform health policy, health practice, and health research (Table 5). Detailed information and examples of organizations were given in the previously published study protocol.

Table 5. Key informants

Categories	Key informants	Examples	Expected number of interviews
Health policy	Policy-makers	Ministry of health officials	2–4
Health practice	Clinicians	Healthcare professionals (doctors, nurses)	2–4
	Clinical guideline developers	UK National Institute for Health and Care Excellence	2–4
	Public health professionals, Commissioners	National public health bodies	2–4
	Public/patients	Patient forums/groups	2–4
Health research	Researchers	Research institutes/universities Knowledge synthesis research groups Belgian Health Care Knowledge Centre (KCE) Africa Evidence Network Student Forums	2–4
	Funding bodies	UK National Institute for Health Research European Union	2–4

Study participants were recruited via contacts and organizations identified in the scoping review, relevant scientific publications, existing professional networks (e.g., H2020 International Training Network MiRoR), and contacts from conference attendance (e.g., Evidence Live and Cochrane Colloquium).

The estimation of the sample size for qualitative studies depends on the point at which data saturation is reached (i.e., the point when new data does not add to a better understanding of the studied phenomenon but rather repeats what was previously expressed [119]). Considering that the point of saturation cannot be specified in advance, we planned to conduct between 14 and 28 interviews, owing to usual points of data saturation reported in qualitative studies [120]. The point of data saturation was determined based on the seven parameters identified by Hennink et al., [121, 122]: the study purpose, population, sampling strategy, data quality, type of codes, code book and

saturation goal, and focus retrieved from the study. These parameters were discussed throughout the study primarily between the lead researcher (LN) and senior researcher (Darko Hren, DH).

4.2.3.3. Data collection and recording

I used semi-structured interviews for this study. The main reason for selecting semi-structured interviews was to allow specific areas to be addressed while giving the interviewees the opportunity to reflect on their experiences and perspectives related to defining, identifying, and presenting research gaps that are relevant to them and that may not have been explored or anticipated by the researcher(s) [123].

The guide was developed by focusing on exploring key stakeholder perspectives and experiences with the following key areas:

1) Participant background information
2) Definitions of research gaps
3) Knowledge, perceptions and experiences on methods/practices used to identify and display gaps in health research to inform further health policy, practice and research

These three categories were developed with information from the scoping review to guide the questions. The interview topic guide was piloted before data collection. It was also adapted according to key stakeholder groups to ensure that it is meaningful to their background and to gather more relevant information based on their experiences and knowledge [124].

The semi-structured interview guide contained two levels of questions: main themes and follow-up questions. The main themes covered the general content of the research gaps aimed to encourage participants to speak freely about their perceptions, experiences, and practices. Follow-up questions

were used as prompts and probes aimed at following respondents' answers and investigating the raised issues more in-depth. The interview guide covered the main topics of the study, providing a focused structure for the discussion during the interviews [125].

I conducted in-person, telephone and teleconference interviews. In-person interviews were conducted with participants residing or reachable in London, UK, and other participants were interviewed via telephone or teleconference (for the interview guide for both the in-person and teleconference interviews see supplementary appendix 1).

All interviews were digitally recorded, transcribed verbatim and anonymised. The lead researcher (LN) transcribed two interviews to help inform the analytical process, and the other audio files were transcribed by a professional transcription agency licensed from the University of Liverpool.

4.2.3.4. Data analysis

I used analytical categories to describe and explain definitions, experiences and practices reported among the groups of participants. All data relevant to each category (defining research gaps, experiences with methods/practices used to identify and display gaps in health research) was identified and examined to ensure that each data item was checked accordingly.

Our approach was based on the thematic analysis outlined by Braun and Clarke [126]. The steps included 1) transcription and checking transcripts with recordings for accuracy; 2) open coding from interview responses performed by two researchers independently (LN and DH); 3) agreement of initial codes discussed among the researchers and an initial codebook developed; 4) the code structure used for analysing the remaining responses with openness that included new codes and

refined existing ones; and 5) themes and subthemes identified from the final code structure and their relationships presented [126].

The initial coding framework for our analysis started from broad categories identified in the previous scoping review, upon which the interviews were structured. Within these broad categories (i.e., defining research gaps, experiences on methods/practices used to identify and display gaps in health research), analytic categories were inductively derived from the data. In this sense, our approach includes both top-down and bottom-up development of analytic categories and themes. QSR International's NVivo 12 qualitative data analysis software was used for data management and analysis.

4.2.3.5. Securing study quality

To further ensure rigour and trustworthiness, the study was guided by Guba and Lincoln's concepts for defining and investigating quality in qualitative research that can be considered parallel to quantitative research concepts of validity and reliability [121, 127, 128]. The concepts include credibility, transferability, dependability, confirmability, audit trails and reflectivity. They are interrelated, and thinking through them from the onset and incorporating them in a study improves the study rigour.

Credibility is defined as the confidence that can be placed in the truth of the research findings, and is considered the most important criterion [129-131]. To ensure credibility of our study, we used peer debriefing, which entailed the qualitative lead researcher (LN) seeking support from the senior researcher (DH) to provide scholarly guidance. The feedback helped improve the quality of the inquiry findings [131]. Transferability refers to the extent to which findings of qualitative research

can be transferred to other contexts and are useful to people in other settings [118, 131-133]. We addressed this by capturing a rich, description of the key stakeholders' context [131, 133]. Dependability is related to whether the research questions are clear and logically connected to the research purpose and design [132]. We achieved dependability by first drafting a study protocol to guide our study and future studies with a similar purpose[104]. Confirmability is related to objectivity or neutrality for establishing that the data and interpretations of the findings are not figments of the inquirer's imagination but are clearly derived from the data, that data collection and interpretations of the study are clearly deliberated from the data and not misinterpreted [132]. We addressed confirmability by documenting the justification of methodological and analytical choices to illustrate how the data were derived in relation to the study objectives and transparently describing the research steps taken from the start of the project to the development and reporting of the findings. Records of the research path were kept throughout the study, and de-briefing sessions were held between the main researcher (LN) and senior researcher (DH). Finally, reflexivity included examining one's own conceptual lens, explicit and implicit assumptions, preconceptions and values and how these affect research decisions in all phases of qualitative studies. Reflexivity was achieved by ensuring transparency of the study process by maintaining clear documentation.

4.2.3.6. Ethical consideration and confidentiality

Informed consent was obtained in accordance with the University of Liverpool Ethics Committee board requirements. Verbal consent was sought for phone interviews and written consent for in-person interviews. Confidentiality and data protection were ensured in accordance with the University of Liverpool Ethics Committee board. All participant information was anonymized, and hard-copy data will be stored in a locked unit. Soft-copy material was stored in a password-

protected file. Upon publication of the study results, all study material will be stored and disposed of according to the rules and regulations of the University of Liverpool. The research obtained ethical approval from the University of Liverpool, UK.

4.3. Results

Among 30 key stakeholders contacted, 20 agreed to participate in the study. Hence, we conducted 20 interviews with 20 participants involved in using evidence for informing health policy, practice or future research (Table 6).

Table 6. Participant characteristics (n=20)

Category	No. (% of total)
Researcher	9 (45%)
Methodologist	5 (25%)
Data visualization	3 (15%)
PhD student	1 (5%)
Health practitioner	6 (40%)
Healthcare provider	5 (33%)
Public health professional	1 (7%)
Oversight bodies	3 (15%)
Health policy-maker	2 (10%)
Funding body	1 (5%)
Patients/public	2 (10%)

4.3.1. Definitions of gaps in health research

I first explored what participants reported as gaps in health research. Given the nature of our interest, participants' answers could be grouped under a single theme "Definitions of Gaps in Health Research". However, the focus of the definitions differed, and within this main theme, we

identified 5 subthemes (i.e., gaps in information, knowledge/evidence related gaps, quality of evidence, uncertainties and patient related gaps) (Figure 3 and Table 7). The terms ranged from lack of information, inadequate information, insufficient information, and evidence gaps to treatment uncertainty, among others. Patient/public participants defined research gaps in a much more literal manner than other participants did; for example, *"The gap is to get more patients involved in doing … clinical trials; have [someone] at the beginning introduce me, [educate me], [provide] awareness [because] I didn't know what [a clinical trial] was. I [didn't] know what they're talking about" (PPI01, patient/public person) and "Get me involved in co-production. That is the gap that is missing in clinical research" (PPI01, patient/public person).*

One participant related research gaps to quality of evidence by use of GRADE, an approach for rating the quality of evidence and grading the strength of recommendations in healthcare[12]. GRADE presents the use of a prominent framework for evaluating the certainty of evidence that can inform the research gap and characterize it [26]. Another participant emphasized the importance of public and community involvement in gap identification to ensure that it takes into account their perspectives and contributions to the research ecosystem: *"Existing knowledge but not documented is of key importance in understanding the current body of knowledge on a particular topic area…. Evidence gaps need to be defined not only by [the] research community but also according to the key stakeholders including community members. Community knowledge is of key importance to inform the evidence base. Further evaluation on research findings to characterize the nature of research gaps can be carried out by evaluating community perspectives and local evidence to confirm scientific evidence*." (*R01, health research PhD student*). The most common description research participants provided of research gaps, was a research gap as the absence of scientific information to answer a research question.

The study showed variability in participant responses as to how to define gaps in health research; this variability was mainly observed in individual responses for the three main categories (research, practice, and policy and funding). Most participants described research gaps as missing, inadequate or giving insufficient information about a particular issue. This finding was similar to the adapted classification from a previous study that developed a framework by identifying research gaps in systematic reviews and the scoping review findings [2, 7]. In summary, this study showed that research gaps need to be defined by researchers and confirmed by patients and the public to ensure societal relevance and importance. We also found that clearly defining research gaps can give information on the most appropriate methodological approach to adopt in identifying and displaying gaps.

Figure 3. Reported descriptions of gaps in health research

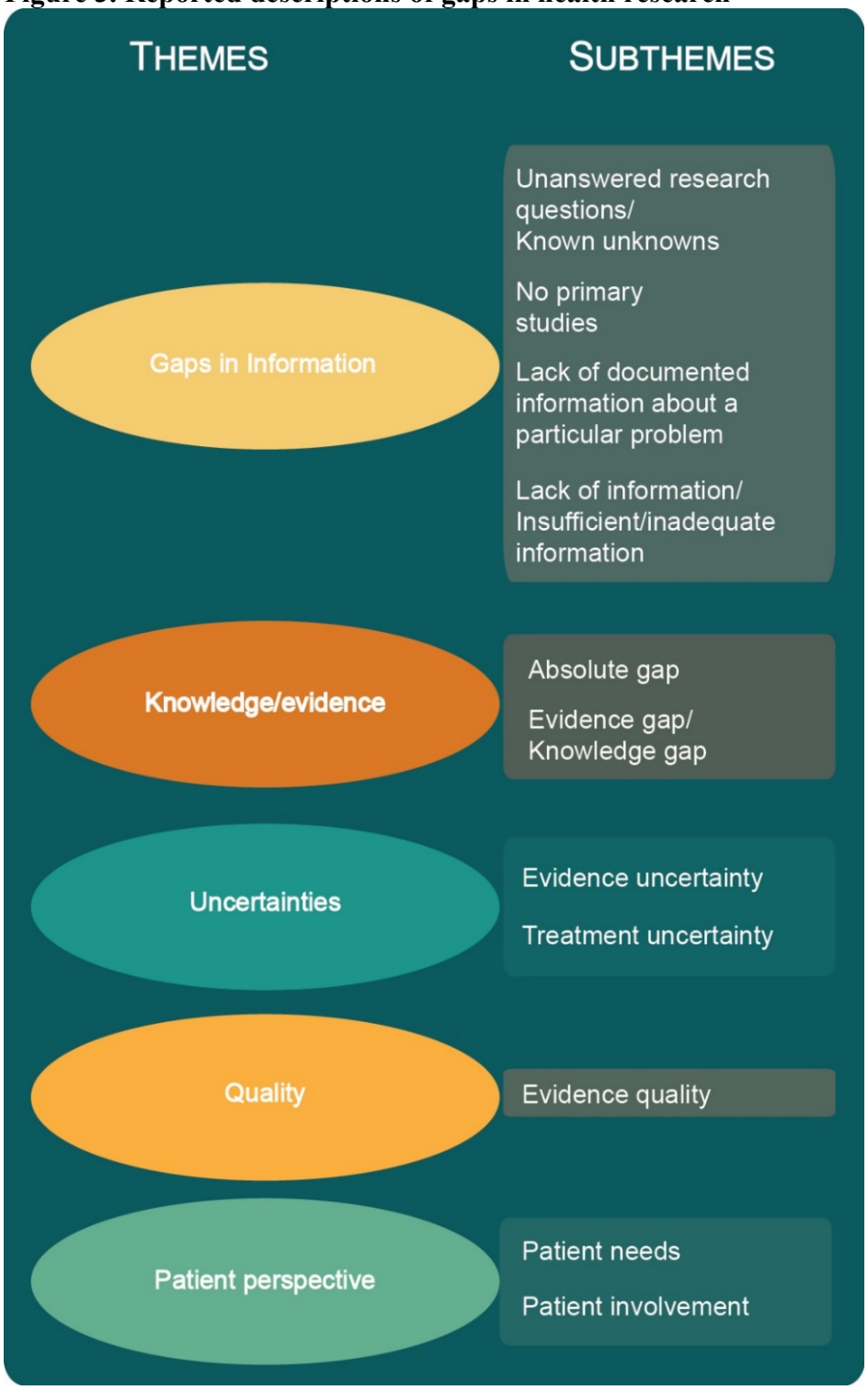

Table 7. Reported descriptions of gaps in health research

Theme	Sub-themes	Codes	Key quotation (quoted source)
Definitions of gaps in health research	Gaps in Information	Lack of information/ insufficient information	*Research gap is basically a lack of information I would say for me. It can be lack of information or lack of sufficient information about a particular topic, or an area, or lack of documented information about a particular problem, issue. (R01, health research student)*
			An area where there is missing or … insufficient information. And because of this … you cannot reach a conclusion for a question. So it is, it is a field, it is an area, a question an issue to which you don't have an appropriate answer because there is missing … information or the research that still needs to be done in that particular area. (F01, funding body)
		Lack of documented information about a particular problem	*I feel like it is more about lack of documented information rather than lack of information per se. Because these people already know what the issues are…. but because it hasn't been documented in research, people assume that it is lack of information (R01, health researcher)*
		Known unknowns	*Known unknowns that there are answers to these, but often these are information gaps, and so there often are … questions that perhaps the medical world knows the answer to but patients don't*

		necessarily know the answer to. (*P01, health research methodologist)*
	No primary studies	*…a review that looked at that area, but found no studies, that is also a gap. (R07, data visualization in health research)*
	Unanswered research questions	*… questions have not yet been answered by research and it is those questions that are of most importance to the people who the research will actually make a difference to. (R04, health research methodologist)*
Knowledge/evidence	Knowledge gap	*…a gap in knowledge. If, there is not enough research done in one area then we can't really answer questions in an evidence-based way. (R02, health research methodologist)*
	Evidence gap	*…we leave it to the priority setting partnership itself to define what they call an evidence gap. So we let them define it. But often it is about … if there is ongoing research in that area. (R04 & R05, health research methodologists)*
	Absolute gap	*….there is no evidence whatsoever. (R04 & R05, health research methodologists)*
Uncertainties	Treatment uncertainty	*…treatment uncertainties... and uncertainty defined as whether or not an area had a systematic review. (R04 & R05, health research methodologists)*
	Evidence uncertainty	*Evidence that was uncertain, maybe lower quality, not directly applicable, not very large body of evidence, not a large number of studies etc. … So*

		you might have uncertain evidence and be less able to make a strong recommendation. (PO1, health research methodologist)
Quality	Evidence quality	*Evidence that was uncertain maybe lower quality, not directly applicable, not very large body of evidence, not a large number of studies etc. Like the things that you might consider in a grade profile, … then they are articulating gaps in certainty and strength of evidence. (PO1, health research methodologist)*
Patient perspective	Patient involvement	*The gap is to get more patients involved in doing like clinical trials, have some at the beginning introduce me, education, awareness because I didn't know what a clinical trial was, I don't know what they're talking about. (PPI01, patient/public)* *Get me involved co-production that is the gap that is missing in clinical research. (PPI01, patient/public)*
	Patient needs	*….patient relevant research and how to identify their needs, for example, with burn wound patients … all the [studies] were focused on the stimulation of the healing of the burn wound, whereas the patients' real problem was the itching of the healed burn wound and none of the researchers was actually working on a solution for the itching. So there was a gap between the research community and the real*

patient needs, so they were studying the wrong thing…. therefore, a research gap is only a research gap if it is confirmed by patients as a research gap. … It should not be an artificial gap created by some stakeholders. (HP03, health practitioner)

4.3.2. Methods to identify gaps in health research

Among the methods to identify gaps in health research, participants reported a wide range of examples that demonstrate the applicable methods being used by different key stakeholders. The findings provided a comprehensive list of methods that adds to initial list of methods identified in the scoping review.

The participants referred to a number of different methods used to identify gaps in health research (Figure 4 and Table 8). The methods were also characterized by the different research methodologies used (i.e., primary, secondary, both). Participants also expressed the difficulty in identifying research gaps; for example, *"It is really difficult to identify research gaps. Lots of people you know will try and use the discussion section from research, [whereas] other authors have asked for further research, but in my experience that has not been a very useful method because sometimes authors will write that you know without really seeing or understanding that there has been something similar done in that field." (R02, health research methodologist).* The variety of identified methods reflected the state of the field in the sense of the wide array of methods currently used with no clear consensus or guidance.

Figure 4. Methods used to identify gaps in health research

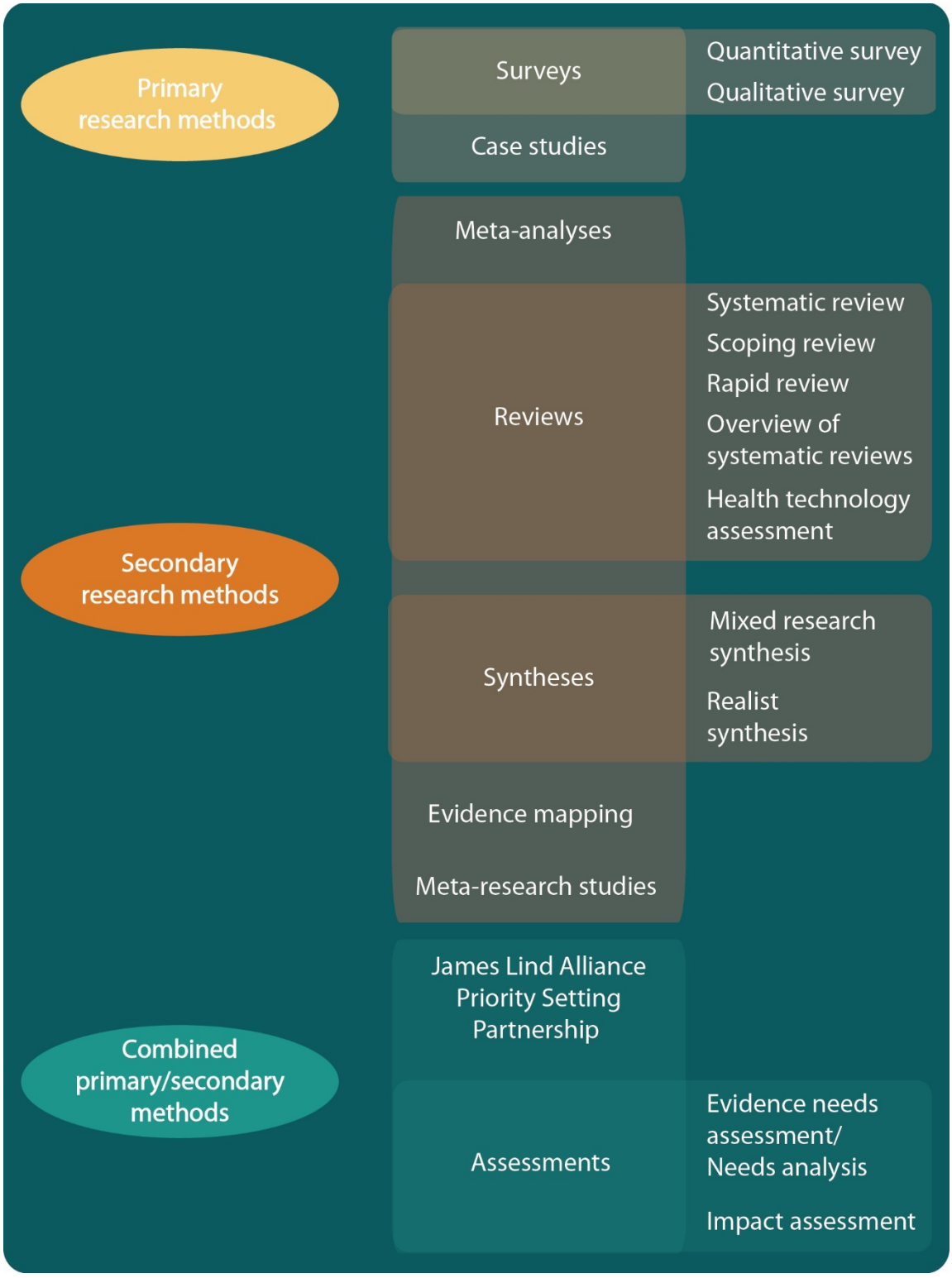

Table 8. Reported methods to identify gaps in health research

Methods to identify gaps according to interviewees	Description of method according to literature	Key quotation (quoted source)
Quantitative and qualitative survey	"A scientific procedure for collecting information and making quantitative inferences about a predefined population. Surveys allow selection of a relatively large sample of people from a predetermined population, followed by collection of data from those individuals, and may be exploratory, descriptive, or explanatory. The key considerations for research using surveys are that as with other methods, developing and refining a central research question are important, followed by careful consideration of the population being sought" [134]	*A very large data gathering exercise, of qualitative data. So it is like a mix of a quantitative survey but with qualitative data and so someone has to manage and process all that information. A good range of responses from all the different stakeholders.* *An overall limitation is that you can get a huge amount of responses back and someone or a group have to go through each of those responses* (*R04 & R05, health research methodologists)*
James Lind Alliance Priority Setting Partnership (JLA PSP)	"JLA PSP methods were designed to allow clinicians, patients, and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that	*Priority Setting Partnerships are run by a steering group, which is a small group that represents patients, carers, and clinicians and they really… make the decisions together, about how the James*

	could be answered by research by gathering research questions, checking existing research evidence, interim prioritization, and a final consensus meeting to reach agreement on the top 10 research priorities" [104, 135]	*Lind Alliance methodology will be applied to their condition, or setting, whichever topic area they are looking at. (R04 & R05, health research methodologists)*
Evidence needs assessment/need analysis **(Searching systematic reviews, conducting community-based survey and stakeholder focus group)**	"It is a systematic method of identifying unmet health and healthcare needs of a population and making changes to meet these unmet needs. It involves an epidemiological and qualitative approach to determining priorities which incorporates clinical and cost effectiveness and patients' perspectives. This approach must balance clinical, ethical, and economic considerations of need—that is, what should be done, what can be done, and what can be afforded" [136]	*There is a need analysis, there is a kind of review as to what we could commission to fill those needs, other methodologies that we used ... a systematic review on a specific topic's needs, because there is more ... literature that is published around that theme now ... recognising that a lot of it was grey literature, but nevertheless it can be informative. ... we also did a community-based survey. We did a stakeholder focus group, ... so if you were to look at some of these methods and how they were conducted.* (*HP02, health practitioner)* *The evidence needs assessment is based on expert consultations, saying what your evidence needs are, what research question do you want answered and so*

		they got the list ….so it is a big document. (*HP02, health practitioner)*
Impact assessment	"Health impact assessment is a means of evidence-based policy making for improvement in health. It is a combination of methods whose aim is to assess the health consequences to a population of a policy, project, or programme that does not necessarily have health as its primary objective" [137, 138]	*…so when you want to launch in a certain area, we may do impact assessment studies and this is a longer procedure, and they are free to all to come so everybody can contribute with comments but mostly people who are working at the certain area they do contribute with this. (F01, health research funder)*

Comparative effectiveness trials	"Comparative effectiveness research is the generation and synthesis of evidence that compares the benefits and harms of alternative methods to prevent, diagnose, treat, and monitor a clinical condition or to improve the delivery of care. Evidence is generated through research that uses various study designs (e.g., observational, clinical trial) and synthesized through systematic reviews" [139]	*That is right and the focus really is comparative effectiveness trials, so it is comparing interventions, it can be medicines, it can be medical devices it can be diet, it can be psychotherapy, whatever, radiotherapy. And it can be different technologies in the two arms that are on the market and that are being used, and so we can compare in fact two strategies that are accepted and are on the market today and are being used, but no one knows which one is the better strategy for the patient and which one is then also the better cost-effective option. (HP04, health practitioner)*
Case report/case series	"Case reports and case series or case study research are descriptive studies that are prepared for illustrating novel, unusual, or atypical features identified in patients in medical practice, and they potentially generate new research questions. They are empirical inquiries or investigations of a patient or a group of patients in a	*So, finally I mean you start by trying it yourself, and do your small case report, or case series … if we do a nice methodology we can actually come up with the evidence for this. So, it is a usual struggle and at the end you end up probably um… evaluating if there is no harm and trying to yourself, without the appropriate evidence as background. (HP04, health practitioner)*

	natural, real-world clinical setting" [140]	
Qualitative, quantitative meta-analysis	"Qualitative meta-analysis is an attempt to conduct a rigorous secondary qualitative analysis of primary qualitative findings. Its purpose—to provide a more comprehensive description of a phenomenon and an assessment of the influence of the method of investigation on findings—is discussed. The distinctive features of conducting meta-analysis approaches are presented. Several considerations important for conducting qualitative meta-analysis are also discussed" [141] "Meta-analysis is a quantitative, formal, epidemiological study design used to systematically assess previous research studies to derive conclusions about that body of research" [142]	*Especially the term meta-analysis, I am now getting [32.45] but I really dislike the way how the term meta-analysis is used because it is just in the end it is just a statistical technique that can be used when you are synthesising quantitative studies. But it does not necessarily have to be used when synthesising quantitative and definitely is not the same thing as doing a whole systematic review; it is just a systematic review is just an overarching term, in which you can choose to synthesise quantitative studies, qualitative studies or quantitative and qualitative studies, and when you are synthesising quantitative studies it is one way of analysing the data. But it is nothing else than that but this term meta-analysis is being misused I think but…* *(R09, health research methodologist)*

Systematic reviews	"Efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored" [49]	*It is really difficult to identify research gaps; lots of people you know will try and use the discussion section from research [in which] other authors have asked for further research, but in my experience that has not been a very useful method because sometimes authors will write that you know without really seeing or understanding that there has been something similar done in that field.(R09, health research methodologist)*
Health technology assessment	"Any process of examining and reporting properties of a medical technology used in health care, such as safety, efficacy, feasibility, and indications for use, cost, and cost effectiveness, as well as social, economic, and ethical consequences, whether intended or unintended" [143]	*...health technology assessments. These broad evaluations of existing evidence to inform decision making. Producing health technology assessments largely for NICE but contracting directly by the National Institute for Health Research HTA programme. (P01, health policy and guideline developer)*
Overview of systematic reviews	"Also known as overview; umbrella review; meta-review; (systematic) review of (systematic) reviews; synthesis of systematic reviews; and summary of systematic reviews. The	*An overview of systematic reviews of methods. So what has actually been used, what is the evidence for other kinds of interventions. (R08, health research methodologist)*

	common feature of the methods associated with all of these terms is the fundamental process of synthesising evidence which is derived, often exclusively, from systematic reviews" [144, 145]	
Rapid review	"A rapid review is a type of knowledge synthesis in which components of the systematic review process are simplified or omitted to produce information in a short period of time" [146, 147]	*...a cut-down systematic review.* *...Well clinicians do rapid reviews every day, but they don't do it in a formalised academic sense. (HP01, health practitioner)*
Scoping review	"It aims to map the existing literature in a field of interest in terms of the volume, nature, and characteristics of the primary research" [21]	*I would recognise research gaps through [a] systematic scope of the literature. (R02, health research methodologist)*
Evidence map	"A systematic search of a broad field to identify gaps in knowledge and/or future research needs that presents results in a user-friendly format, often a visual figure or graph, or a searchable database" [95]	*I do believe evidence maps should be a systematic product, so I do believe they should use systematic review principles, there should be PICOS clearly defining what the question is, the scope of the map. There should be a clear explicit extensive search strategy with clear inclusion, exclusion criteria. There should be systematic search with double screening, double coding and*

		everything all those systematic review principles should apply. (R07, data visualisation in health research)
Meta research studies	"Meta-research is the study of research itself: its methods, reporting, reproducibility, evaluation, and incentives" [148].	*Meta-research studies or research-on-research studies that have delved, are dealing with this particular topic of how to how do you justify your study, how do you use earlier studies in the systematic way, how do you design a new study and how do you replace new results in the context, in a systematic way, of earlier similar trials so you are not you know can be accused of being biased and only presenting what fits your own opinion or your own perspective. But you have to deal, or you have to relate yourself to all that's published regarding a specific topic. (R08, health research methodologist)*
Mixed studies reviews/ Mixed method integrated synthesis	"Mixed research synthesis entails the 'mixing' of the differences characterizing efforts to integrate qualitative research findings with the differences characterizing efforts to integrate quantitative research findings" [149]	*Use of mixed studies reviews of course, to identify evidence gaps … think about one that provides some nice display of the results which is very intuitive for those in practice (R09, health research methodologist)*

Realist synthesis	"Realist synthesis is an increasingly popular approach to the review and synthesis of evidence, which focuses on understanding the mechanisms by which an intervention works (or not)" [150]	*A realist synthesis can establish contextual factors that influence the mechanism of an intervention so, say if you were to evaluate a healthcare intervention then the realist synthesis can, because of its theory building element, can provide you with all the small things that happen in the chain of evidence that eventually lead to the output of the intervention being effective or not*. (*R09, health research methodologist*)

4.3.3. Methods to display gaps in health research

The participants mainly expressed the importance of using data visualisation in research; there was a common understanding on the usefulness of data visualisation as a whole, particularly with the growth of technology and the need to capitalize on it. The main challenges expressed were how to identify an appropriate visualisation to present the research and how to effectively present data. Similar to the methods used to identify gaps in health research, participants referred to a number of different methods used to display gaps in health research (i.e., forest plots, diagrams/illustrations, evidence maps, mega maps, 3IE gap maps and info graphics) (Figure 5 and Table 9). Participant perspectives varied; one of the interviewees pointed out, "*I think with the growth of technology, it is very important to use sophisticated methods to better communicate evidence for policy making and decision making. I think the availability of evidence is not enough on its own and finding different methods to communicate is important, not only the analysis and*

findings but also sharing it in different platforms online for a greater audience" (P02, health policy and guideline developer). Another participant highlighted that one of the key benefits of visually presenting research is being able to immediately see what information is available and missing. Participants mainly expressed the importance of data visualisation in communicating research and the difficulty in finding the right tool to use to present research findings. Below we present participants' general experiences with data visualisation in health research.

4.3.4. Summary

Finally, few respondents were able to define research gaps, unless contextualizing them within a specific study or area, or methods of identification. Fully understanding research gaps in health research and adequately addressing them is difficult. In this study, I highlighted four key items on the topic: 1) clearly defining research gaps provides a context to understand better what the gaps are and what they are caused by; 2) a clear definition of research gaps can inform the methods used to identify research gaps, similar to how a clear research question can inform the research study methodology;3) upon adopting the most appropriate methods to identify research gaps, finding the right visualisation to communicate them effectively is important; and 4) public involvement, when applicable, is needed to verify that gaps are important and relevant to the public.

Figure 5. Methods used to display gaps in health research

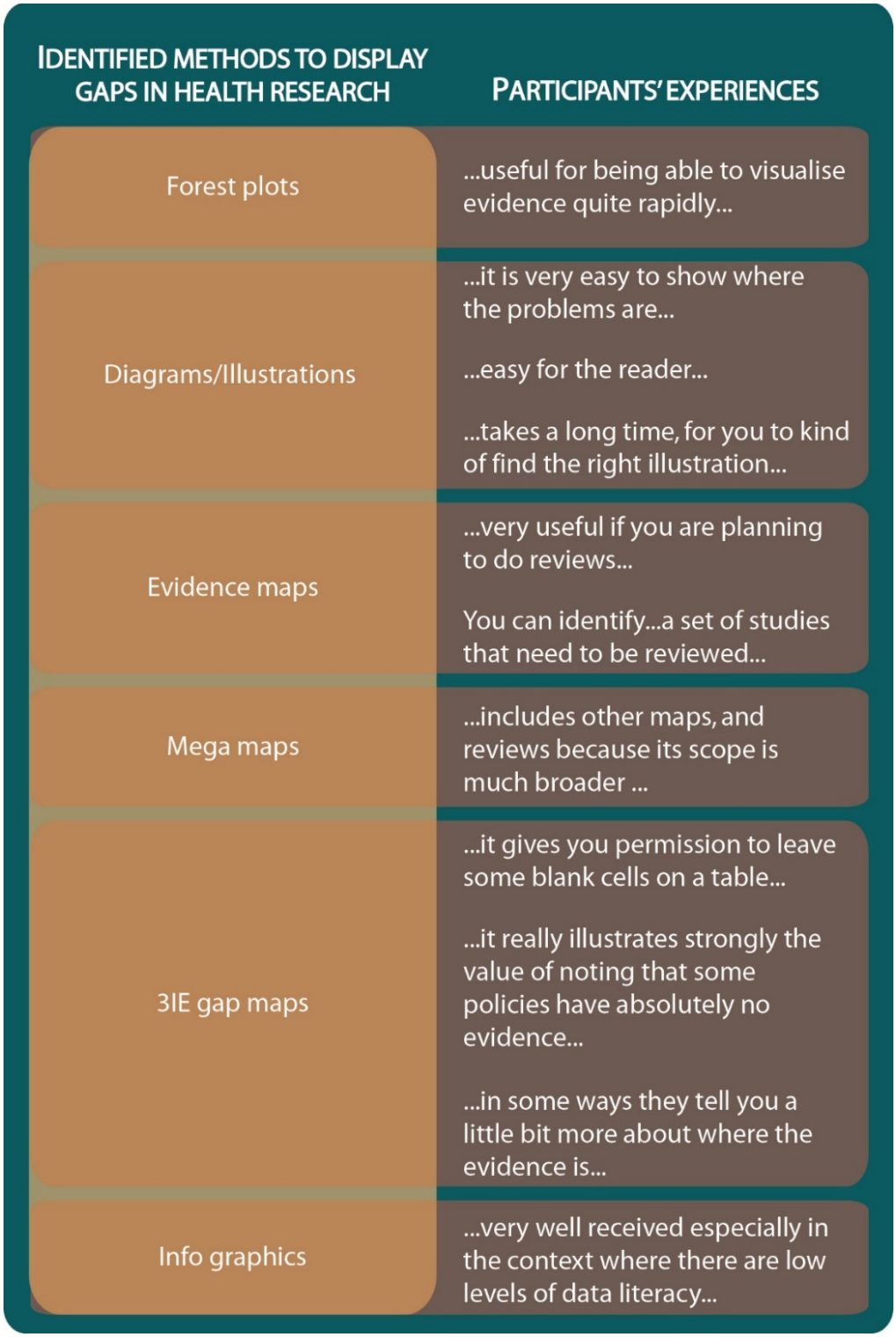

Table 9. Reported methods to display gaps in health research

Code	Sub-codes	Quotations
Experiences with methods to display gaps	**Forest plots**	*… meta-analysis forest plots I think are useful for being able to visualise evidence quite rapidly. (P01, health policy and guideline developer)*
	Diagrams/Illustrations	*When I am presenting my findings, I use diagrams because it is very easy to show where the problems are, so normally I just use like illustrations. (R01, health research student)* *Yes there is plenty of illustrations … extensive illustrations to make it really easy for the reader to which catches the eye... (HP03, health practitioner)* *illustrations are good, but I feel like it takes a long time, for you to kind of find the right illustration for me think it takes a bit of peer review, you have to talk to some people before you can actually use that. (R01, health research student)*
	Evidence maps	*So I would see maps very much as being a tool towards people who are planning research, so they could be research commissioners, they could be researchers themselves, to identify what evidence is there. They are very useful if you are planning to do reviews, you can identify here is a set of studies that need to be reviewed they have already been searched and screened so we just take those studies and use those ones. (R07, data visualization in health research)*

Mega maps	*...then we have got the mega map, so the mega map just includes other maps, and reviews because its scope is much broader. (R07, data visualization in health research)*
3IE gap maps	*And one important thing about the gap map 3IE approach is at least it gives you permission to leave some blank cells on a table. And it really illustrates strongly the value of noting that some policies have absolutely no evidence. Some of their gap maps can use size of shape, a dot or a circle, to illustrate how many studies or you know numbers of people or strength of evidence in some ways they tell you a little bit more about where the evidence is. (P01, health policy and guideline developer)*
Info graphics	*I would make the effort to make that information more accessible using info graphics, which are very well received especially in [a] context where there are low levels of data literacy. (HP02, health practitioner)*

4.4. Discussion

This study provides insight into issues related to defining, identifying and displaying research gaps in health from the perspective of key stakeholders. The findings indicate several definitions of gaps in health research and methods used to identify and display research gaps. Our study confirmed the ambiguity in defining research gaps and methodological approaches to identify research gaps[2, 151] and display research gaps [104]. The methods used to identify research gaps were closely linked to the definition of research gaps. For example, the JLA method of gap identification and setting priorities for research begins by clearly defining what the alliance refers to as evidence uncertainty; that is, there is no up-to-date, reliable systematic review of research evidence

addressing the uncertainty or showing that uncertainty [135]. This step further informs the rest of the methodology used and is critical in identifying the treatment uncertainties and determining the future research priorities. This method combines both primary and secondary approaches and not only identifies research gaps but also verifies them across different relevant stakeholders including researchers, patients, their carers and clinicians, to ensure the relevance and potential benefit to them[135]. This verification is important, given that some research gaps may be of key interest to researchers but have little relevance and importance to patients or the public, who should be the main beneficiaries of research to improve their health and well-being.

The overall methods to identify research gaps involved primary, secondary or both approaches (Figure 2). Most of the participants mentioned the use of secondary research methods; this is in accordance with the research that has been conducted on research gaps, which has also primarily focused on the use of secondary research and developed frameworks for identifying research gaps [2, 7-9, 104]. The most commonly adopted framework involves identifying research gaps from systematic reviews using the PICOS framework (Population, Intervention, Comparison, Outcome, Setting) to characterize a research gap[2]. The other framework involves identifying research gaps in qualitative literature reviews[9]. Additionally, the GRADE approach for rating the quality of evidence and grading the strength of recommendations in healthcare[12] presents the use of a prominent framework for evaluating the certainty of evidence that can inform the research gap and characterize it [26]. Moreover, scoping reviews are commonly used, and the definition includes aiming to identify research gaps by mapping the current body of evidence. These examples focus on the use of secondary research methods, but we lack studies that specifically explore the use of primary or both primary and secondary methods to identify research gaps, yet these methods

equally exist and are being used. Additional exploration of applicable methods for identifying gaps can improve their usefulness and relevance in health research.

Participants mainly expressed the importance of data visualisation in communicating research; no specific methods or formats to present gaps were expressed. Thus, the use of data visualisation is desirable among different stakeholders, particularly researchers, when communicating research, although we found few examples of experiences with developing and using data visualisation. The participants mainly expressed the difficulty in finding the right tool to use to present research findings.

In summary, this study showed that research gaps need to be defined by researchers and confirmed by different research stakeholders such as patients and the public to ensure societal relevance and importance[152]. We also found that clearly defining research gaps can give information on the most appropriate methodological approach to adopt in identifying and displaying gaps, for example, for exploring research gaps in a specific or broad area. For a specific area, a systematic review can be considered and within a broad area, an umbrella review. The study also showed that the use of both primary and secondary methods (JLA method) to identify gaps is the most robust method for gap identification. The main reported advantage of this method is that it identifies gaps (treatment uncertainties) and involves different stakeholders, including patients and the public, to confirm and prioritize gaps. The main disadvantage is that it is labor-intensive (requires a team of different specialists) and expensive (administrative support, meeting rooms, catering, among others) as compared with secondary methods (evidence synthesis) or primary methods (survey).

To advance efforts in identifying research gaps, further work and different study designs are needed to take this work to the next step, to find consensus on definitions and different practices for methods in identifying research gaps. Subsequently, also assessing the best methods according to different stakeholders will be informative and important.

4.5. Ethics and dissemination

Informed consent was obtained in accordance with the University of Liverpool Ethics Committee board requirements. Verbal consent was sought for phone interviews and written consent for in-person interviews. Confidentiality and data protection were ensured in accordance with the University of Liverpool Ethics Committee board. All participant information was anonymized and hard-copy data stored in a locked unit. Soft-copy material was stored in a password-protected file. Upon completion of the study and publication of the study results, all study material will be stored and disposed of according to the rules and regulations of the University of Liverpool. The study protocol will be stored in the data repository Zenodo. The research obtained ethical approval from the University of Liverpool, UK. At the end of this research project, the results will be presented at conferences and relevant meetings (e.g., H2020 International Training Network MiRoR). They will also be published in a peer-reviewed journal and as part of a doctoral thesis of the PhD fellow (LN) as well as in professional and lay magazines and presented in workshops at professional events for stakeholder groups and as online materials with good practice examples.

4.6. Strengths and limitations

This study was the first to look into key stakeholder experiences in the topic area. It provides an overview of how research gaps are defined and the different approaches used to identify research gaps in practice. Several limitations need to be considered when interpreting the results. One is

that patients or the public were not involved in the design or analysis of this study. Including patients/public perspectives would have benefited the study design by being able to improve the importance and relevance of the findings for this population. However, we involved them as study participants and will disseminate the study findings that pertain to them by using a patient/public online platform, peopleinresearch.org.

One of the main challenges in this study was that because the topic area is still very vague and unclear, the recruitment and interview process was challenging. Therefore, this study was primarily limited to what participants were familiar with and not necessarily representative of the full scope of the status of health researchers, health practitioners, oversight bodies and patients/the public. A more generalizable understanding of this topic area would require a larger sample of participants and methodology such as a Delphi survey and/or a priority-setting partnership with representatives using evidence to inform policy, practice and research. This study would also have benefited from widening the scope of the stakeholder categories (use of evidence to inform health policy, health practice, and health research)[104]. This would have enriched our study findings and provided a wider view of stakeholder experiences outside our categories. Another limitation of this study is not including patients/the public in designing the study. Including patient/public perspectives would have benefited the study design by being able to improve the importance and relevance of the findings for this population.

One of the main strengths of the study is improving the definition of research gaps and subsequently improving the accurate reporting of research gaps, to elucidate the characteristics, which can help in evidence-based decisions. For example, a decision based on a research gap contributing to lack of primary research on a specific health problem can differ from one based on a research gap related

to lack of secondary research summarizing the research. Hence, all these factors regarding research gaps need to be highlighted if they are known and made explicit when disseminating and communicating research. Additionally, providing more information on what the gap represents may inform users of evidence of more specific information about the research gap and how it can be addressed more accurately.

Also, only 20 key stakeholders were recruited for the qualitative interviews; hence, our results should be considered hypothesis-forming and not representative of all stakeholders. However, the purpose of the qualitative interviews was to identify general themes with respect to how different stakeholders experienced defining research gaps and methods to identify and display research gaps. The optimal follow-up research design to compliment and strengthen this study would be to quantify the frequency of different opinions by use of a survey with a representative sampling scheme.

4.7. Conclusion

This qualitative study went beyond the scientific literature in describing, identifying and displaying gaps in health research and directly talked to people about their understanding and practices. Given the nature of this topic that is not fully explored, understanding real practices to complement the existing literature can better inform the development of methodological guidance. We anticipate that this study will advance efforts in research and practice in this area.

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Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study protocol

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ABSTRACT

Introduction Identifying research gaps can inform the design and conduct of health research, practice and policies by informing the current body of evidence. Audiences including researchers, clinical guideline developers, clinicians, policymakers, research regulatory bodies, funders and patients/the public can also benefit from understanding the status of research and research gaps to make informed choices. This study aims to explore how key informants define research gaps and characterise methods/practices used to identify and display gaps in health research to inform future research practice and policies.

Methods and analysis This is an exploratory qualitative study using semi-structured in-depth interviews. The participants will be recruited by purposive sampling from initiatives and organisations previously identified in a scoping review on methods to identify, prioritise and display gaps in health research. We anticipate performing up to 28 interviews with the different key informant groups who are involved in using evidence to inform health policy, practice and research. Interviews will be thematically analysed as outlined by Braun and Clarke. The qualitative data-analysis software NVivo V.12 Pro will be used to aid data management and analysis.

Discussion This is the protocol for a follow-up study that aims to complement and enrich the findings of the scoping review on methods to identify, prioritise and display gaps in health research. The overall project aims to develop methodological guidance for describing, identifying and displaying gaps in health research.

Ethics and dissemination The research obtained ethical approval from the University of Liverpool, UK. The findings will be disseminated via conferences, meetings (organised by the Methods in Research on Research project), peer-reviewed publications and lay magazines because the study participants will include the public/patients.

Strengths and limitations of this study

- The qualitative nature of this study provides an in-depth understanding of key informants' perspectives and experiences in describing, identifying and displaying gaps in health research.
- This study is embedded in a larger study aiming to develop methodological guidance to identify and display gaps in health research.
- This study would have benefited from including patient/public perspectives in designing the study to be able to improve the importance and relevance of the findings for this population.

BACKGROUND

Identifying research gaps can help inform the design and conduct of health research, practice and policies by providing a better understanding of the current body of evidence. The term 'research gap' is not well defined, and its meaning can differ depending on the researcher and research context. A recent scoping review on methods used to identify, prioritise and display gaps in health research reported 12 different definitions related to gaps in health research (eg, population, theoretical and methodology gaps), each describing research gaps differently.[1] This finding shows the ambiguity of the term 'research gaps' and the different practices it may be related to.

As a basis for further exploring and understanding 'research gaps', we start from the definition given by the National Collaborating Centre for Methods and Tools (NCCMT) in Canada based on the work of Robinson *et al*, whereby a research gap is defined as a topic or area for which missing or insufficient information limits the ability to reach a conclusion for a question.[2] Given the different meanings and definitions of research gaps found in the scoping review,[1] we consider it important to further explore definitions rather than just adopt or modify the NCCMT definition. Clearly defining the type of research gap can help determine how to better identify, characterise, prioritise and address research gaps.

Different methods for identifying research gaps have been reported; for example, scoping reviews and umbrella reviews are emerging methods for mapping and

summarising evidence. These methods have an explicit aim of identifying research gaps in a broad area as compared with systematic reviews that focus on answering a specific research question.[3–7] Robinson *et al* developed a framework using systematic reviews to identify research gaps[2] in which they classified the reasons for the existence of research gaps and used the population, intervention, comparison, outcome and setting process to characterise them. Scoping, umbrella and systematic reviews are reported to specifically identify research gaps, but other methods are being used, and further exploring these methods can optimise their definition, methodological scrutiny and practice.[8–18] Furthermore, the aforementioned methods focus on the use of secondary research methods to identify research gaps. However, a recent scoping review showed that other methods have been used to identify gaps, including primary and both primary and secondary research methods.[1] The scoping review showed a lack of consensus on what constitutes the best methodological approaches to identify research gaps, determine research priorities and display research gaps or priorities.[1 5 7] Therefore, to better understand the different methods and ongoing practices, we aimed to conduct a qualitative study to further explore more in-depth key stakeholder experiences in describing research gaps and the methods used to identify and display gaps in health research.

This study is part of larger ongoing efforts to avoid waste in producing and reporting research evidence, with a focus on the identification of research gaps.[19] Healthcare decisions for individual patients, public health policies and clinical guidelines should be informed by the best available research evidence while taking into consideration research gaps. Investigating experiences with practices/methods used to identify research gaps can inform explicit methodological approaches in identifying and describing research gaps. This investigation can enhance practices of different stakeholder groups (ie, health professionals, commissioners, researchers, patients/the public and decision-makers) when addressing areas of uncertainty within the research problem and topic area.[20] Initiatives such as the James Lind Alliance, UK Database of Uncertainties about the Effects of Treatments, Cochrane Agenda and Priority Setting Methods Group and Evidence-based Research Network are some examples of existing efforts to identify and prioritise research gaps in health.[1]

This study is nested in a larger project aimed at developing methodological guidance for identifying gaps in health research. The first step in the project was a scoping review describing methods used to identify, prioritise and display gaps in health research in scientific literature. The scoping review mapped evidence on different definitions reported for the term 'research gap' as well as methods used to identify research gaps and determine research priorities and display research gaps or research priorities.[1] The second step is the qualitative study described in this protocol. The aim of the study is to investigate the experience of key stakeholders (ie, researchers, funders, clinicians, clinical guideline developers, public health professionals, commissioners, patients/the public and policymakers) with defining research gaps and practices/methods used to identify and display research gaps. The final step will be an integration and overview combining findings from the scoping review and qualitative study to provide a comprehensive overview of methods used to identify and display research gaps. These study findings will be used to inform the methodological guidance on identifying research gaps.

The specific objectives of the study are to (1) investigate key stakeholders' knowledge, perceptions and experiences with defining research gaps and (2) characterise methods/practices used for identifying and displaying gaps in health research.

METHODS AND ANALYSIS

Qualitative study design

This study is an exploratory qualitative study using semi-structured interviews. This method will provide in-depth insight into key stakeholders' perspectives, experiences, and practices with defining, identifying and displaying research gaps. Investigating perspectives of different key stakeholders will ensure that the issue is not explored through one lens but rather a variety of lenses. This will allow for revealing and better understanding multiple facets of research gaps including definitions and methodological approaches/practices to identify and display gaps.[21]

Study sample and recruitment

The study sample will include the following stakeholder groups (ie, researchers, funders, clinicians, clinical guideline developers, public health professionals, commissioners, patients/the public and policymakers). The stakeholder groups will be organised in three main categories focusing on the use of evidence to inform health policy, health practice and health research. These categories (policy, practice and research) are determined from the scoping review findings.[1] More information and examples of organisations are given in table 1. Study participants will be recruited via contacts and organisations identified in the scoping review, relevant scientific publications, existing professional networks (eg, Horizon 2020 (H2020) Project Methods in Research on Research (MiRoR)) and contacts from conference attendance (eg, Evidence Live and Cochrane Colloquium).

This study will also include patients or members of the public as key informants, which will allow for better understanding participants' perceived needs and priorities in identifying research gaps to make informed health decisions. Patients/the public will be recruited and identified via patient support groups online, community centres and public involvement websites such as the peopleinresearch.org platform that involves the public in health research.

Table 1 Key informants

Categories	Key informants	Examples	Expected number of interviews
Health policy	Policymakers	Ministry of health officials	2–4
Health practice	Clinicians	Healthcare professionals (doctors, nurses)	2–4
	Clinical guideline developers	UK National Institute for Health and Care Excellence	2–4
	Public health professionals, commissioners	National public health bodies	2–4
	Public/patients	Patient forums/groups	2–4
Health research	Researchers	Research institutes/universities Knowledge synthesis research groups Belgian Health Care Knowledge Centre Africa Evidence Network Student forums	2–4
	Funding bodies	UK National Institute for Health Research European Union	2–4

We will use purposive sampling to ensure that the perspectives of all identified stakeholder groups are represented. Purposeful sampling is widely used in qualitative research for identifying and selecting information-rich cases, and in this study, further elaboration of the term research gap is needed to better understand the context of the research gaps and methods/practices used to identify and display the research gaps.[22 23]

We anticipate performing about 14–28 interviews. This number of interviews will provide for data saturation (ie, the point when new data do not add to a better understanding of the studied phenomenon but rather repeat what was previously expressed[24]) and also obtain a scope of responses from each stakeholder group. This estimation of interview participants is based on a study involving 60 interviews that showed saturation with 12 interviews, with broader themes apparent after only 6 interviews.[25] The authors noted that factors such as heterogeneity of the sample affect how many interviews are required but concluded that to understand common perceptions and experiences among a group of relatively homogeneous individuals, 12 interviews should suffice.[25] Another study, after examining 25 in-depth interviews, found code saturation after interviews, with the range of thematic issues identified; the authors proposed 16–24 interviews to reach saturation (ie, a richly textured understanding of issues[26]). Therefore, we aim to gather 14–28 interviews for our three main categories (health policy, practice and research).

Saturation will be guided by the seven parameters identified by Hennink *et al*,[26 27] including the study purpose, population, sampling strategy, data quality, type of codes, code book and saturation goal, and focus retrieved from the study. Each of these parameters will be considered throughout the study.

Data collection and recording

Semi-structured interviews will be used for this study. The main reason for selecting semi-structured interviews is to allow for specific areas to be addressed while giving the interviewees the opportunity to reflect on their experiences and perspectives related to defining, identifying and presenting research gaps that are relevant to them and that may not have been explored or anticipated by the researcher(s).[28]

We will conduct interviews in-person and using teleconference, according to the participant's availability and preference. In-person interviews will be conducted primarily with participants residing or reachable in London, UK, and other participants will be interviewed via teleconference (see online supplementary appendix 1 for the interview guide for both the in-person and teleconference interviews). The interviews will be recorded on a digital recorder for face-to-face interviews and electronically for teleconference interviews.

The guide was developed by focusing on exploring key stakeholder perspectives and experiences with the following key areas:

1. Participant background information.
2. Definitions of research gaps.
3. Knowledge, perceptions and experiences on methods/practices used to identify and display gaps in health research to inform further health policy, practice and research.

These three domains were developed with information from the scoping review to guide the questions. The interview topic guide will be piloted before data collection. It will also be adapted according to key stakeholder groups to ensure that it is meaningful to their background and to gather more relevant information based on their experiences and knowledge.[29]

The semi-structured interview guide contains two levels of questions: main themes and follow-up questions. The main themes cover the general content of the research gaps aimed to encourage participants to speak freely about their perceptions, experiences and practices. Follow-up questions are prompts and probes aimed at following respondents' answers and investigating the raised issues more in-depth. The interview guide covers the main topics of the study, providing a focused structure for the discussion during the interviews. However, it does not need to be strictly followed — the main focus is on providing a setting that encourages respondents to share their perceptions and experiences with research gaps as thoroughly as possible within the constraints of our study aims.[30]

All interviews will be transcribed verbatim and anonymised. The lead researcher (LN) will transcribe two interviews to help inform the analytical process, and the other audio files will be transcribed by a professional transcription agency licensed from the University of Liverpool.

Data analysis

We will use analytical categories to describe and explain definitions, experiences and practices reported among the groups of participants. All data relevant to each category (describing research gaps, experience with identifying and displaying research gaps) will be identified and examined to ensure that each data item is checked accordingly.

Our approach is based on the thematic analysis outlined by Braun and Clarke.[31] The steps include the following: (1) transcription and checking transcripts with recordings for accuracy; (2) open coding from interview responses to be performed by two researchers independently (LN and DH); (3) agreement of initial codes to be discussed among the researchers and an initial codebook developed; (4) the code structure to be used for analysing the remaining responses with openness to including new codes and refining existing ones; and (5) themes and subthemes to be identified from the final code structure and their relationships presented.[31]

The initial coding framework for our analysis will start from broad categories identified in the previous scoping review, on which the interviews were structured. Within these broad categories (ie, describing research gaps, experience with identifying and displaying research gaps), analytic categories will be inductively derived from the data. In this sense, our approach includes both top-down and bottom-up development of analytic categories and themes.

Trustworthiness during thematic data analysis will be ensured by storing raw data systematically, documenting detailed notes about the development and hierarchies of concepts and themes, establishing consensus on themes, providing detailed descriptions of context and describing the process of coding and analysis.[8 9] NVivo V.12 Pro, a qualitative data analysis software, will be used for data management and analysis.

Ensuring study quality

To further ensure rigour and trustworthiness, this study will be guided by Guba and Lincoln's concepts for defining and investigating quality in qualitative research that can be considered parallel to quantitative research concepts of validity and reliability.[27 32 33] The concepts include credibility, transferability, dependability, confirmability, audit trails and reflectivity. They are inter-related, and thinking through them from the onset and incorporating them in a study will improve the study rigour.

Credibility is defined as the confidence that can be placed in the truth of the research findings[34–36]; it is considered the most important criterion to ensure rigour and trustworthiness. To ensure credibility of our study, we will use peer debriefing, which will entail the qualitative lead researcher (LN) seeking support from the senior researcher (DH) to provide scholarly guidance. The feedback will help improve the quality of the inquiry findings.[36] Transferability refers to the extent to which findings of qualitative research can be transferred to other contexts and are useful to people in other settings.[21 36–38] We aim to address transferability by reporting a rich, detailed description of the key stakeholders' context and location.[36 38] Dependability is related to whether the research questions are clear and logically connected to the research purpose and design.[37] We aim to achieve dependability by first drafting this protocol to guide our study and future studies with a similar purpose. Confirmability has been related to objectivity or neutrality for establishing that the data and interpretations of the findings are not figments of the inquirer's imagination but are clearly derived from the data, that data collection and interpretations of the study are clearly deliberated from the data and not misinterpreted.[37] We aim to address confirmability by documenting the justification of methodological and analytical choices to illustrate how the data were derived in relation to the study objectives and transparently describing the research steps taken from the start of the project to the development and reporting of the findings. Records of the research path will be kept throughout the study, and debriefing sessions will be held between the main researcher (LN) and senior researcher (DH). Finally, reflexivity includes examining one's own conceptual lens, explicit and implicit assumptions, preconceptions and values and how these affect research decisions in all phases of qualitative studies. Reflexivity will be achieved by ensuring transparency of the study process by maintaining clear documentation.

Patient or public involvement

There is no patient or public involvement in the design or analysis of this study. However, we plan to involve patients/the public in findings that pertain to them and in disseminating study findings. This will be achieved by using patient/public online platforms such as peopleinresearch.org.

DISCUSSION

This study will provide insights into issues related to defining research gaps and methods used to identify and display gaps in health research from perspectives of key stakeholders involved in the process. This is a follow-up study of a wider project; the first study was a scoping review exploring methods used to identify and display research gaps reported in scientific publications.[1] The scoping review showed variation and ambiguity in how research gaps are described as well as the methods used to identify and prioritise research gaps. Several of the articles described the development of a framework or tool for identifying and prioritising research gaps and applying it to a specific topic area as an example for application.[1 2 7 39] There were no evaluations of reproducibility of the method/frameworks identified in the scoping review.[1 7] Furthermore, despite articles highlighting the existence of research gaps in their studies, very few specifically described the gaps and the causes or the method of identification, so fully understanding the relevance and importance of the research gap to adequately address it is difficult. Our scoping review also primarily found the use of secondary research methods such as systematic reviews and scoping reviews as the most commonly used methods to identify gaps; although other methods were identified, they were inadequately described. The scoping review also showed that besides researchers, different audiences including clinicians, policymakers, funders and patients or the public can benefit from understanding gaps and methods/practices on how to identify and display gaps in health research. This qualitative study aims to go beyond the scientific literature in describing, identifying and displaying gaps in health research and directly talk to people about their understanding and practices. Given the nature of this topic that is not fully explored, there is a need to investigate real practices to be able to develop methodological guidance, taking into consideration the existing literature and ongoing practices.

This study has some limitations; one is not including patients/the public in designing the study. Including patients/public perspectives would have benefited the study design by being able to improve the importance and relevance of the findings for this population. One of the main strengths of the study is improving the definition of research gaps and subsequently improving the accurate reporting of research gaps to clearly elucidate the characteristics, which can help in making evidence-based decisions. For example, making a decision based on a research gap contributing to lack of primary research on a specific health problem can differ from a research gap related to lack of secondary research summarising the research. Hence, all these factors regarding research gaps need to be highlighted if they are known and made explicit when disseminating and communicating research. In addition, providing more information on what the gap represents may inform users of evidence on more specific information about the research gap and how it can be addressed more accurately. We anticipate that this study will advance efforts in research and practice on this topic area.

ETHICS AND DISSEMINATION

Informed consent will be obtained in accordance with the University of Liverpool Ethics Committee board requirements. Verbal consent will be sought for phone interviews and written consent for in-person interviews. Confidentiality and data protection will be ensured in accordance with the University of Liverpool Ethics Committee board. All participant information will be anonymised, and hard-copy data will be stored in a locked unit. Soft-copy material will be stored in a password-protected file. On completion of the study and publication of the study results, all study material will be stored and disposed of according to the rules and regulations of the University of Liverpool. The study protocol will be stored in the data repository Zenodo.

At the end of this research project, the results will be presented at conferences and relevant meetings (eg, H2020 Project MiRoR). They will also be published in a peer-reviewed journal and as part of a doctoral thesis of the PhD fellow (LN) as well as in professional and lay magazines and presented in workshops at professional events for stakeholder groups and as online materials with good practice examples.

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Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1. Nyanchoka L, Tudur-Smith C, Thu VN, *et al*. A scoping review describes methods used to identify, prioritize and display gaps in health research. *J Clin Epidemiol* 2019;109:99–110.
2. Robinson KA, Saldanha IJ, Mckoy NA. Development of a framework to identify research gaps from systematic reviews. *J Clin Epidemiol* 2011;64:1325–30.
3. Snilstveit B, Vojtkova M, Bhavsar A, Birte S, Martina V, Ami B, *et al*. Evidence & Gap Maps: A tool for promoting evidence informed policy and strategic research agendas. *J Clin Epidemiol* 2016;79:120.
4. Pham MT, Rajić A, Greig JD, *et al*. A scoping review of scoping reviews: advancing the approach and enhancing the consistency. *Research Synthesis Methods* 2014;5:371–85.
5. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. *Int J Soc Res Methodol* 2005;8:19–32.

6. Levac D, Colquhoun H, O'Brien KK. Scoping studies: advancing the methodology. *Implementation Science* 2010;5.
7. Carey T, Yon A, Beadles C, *et al*. *Prioritizing future research through examination of research gaps in systematic reviews*. Prepared for the Patient-Centered Outcomes Research Institute, 2012.
8. Whittemore R, Chao A, Jang M, *et al*. Methods for knowledge synthesis: an overview. *Heart & Lung* 2014;43:453–61.
9. Kastner M, Antony J, Soobiah C, *et al*. Conceptual recommendations for selecting the most appropriate knowledge synthesis method to answer research questions related to complex evidence. *J Clin Epidemiol* 2016;73:43–9.
10. Manson H. Systematic reviews are not enough: policymakers need a greater variety of synthesized evidence. *J Clin Epidemiol* 2016;73:11–14.
11. Perrier L, Lightfoot D, Kealey MR, *et al*. Knowledge synthesis research: a bibliometric analysis. *J Clin Epidemiol* 2016;73:50–7.
12. Pluye P, Hong QN, Bush PL, *et al*. Opening-up the definition of systematic literature review: the plurality of worldviews, methodologies and methods for reviews and syntheses. *J Clin Epidemiol* 2016;73:2–5.
13. Sales A. Generating and using evidence: reflections from the perspective of implementation. *J Clin Epidemiol* 2016;73:6–7.
14. Straus SE, Kastner M, Soobiah C, *et al*. Introduction: engaging researchers on developing, using, and improving knowledge synthesis methods: a series of articles describing the results of a scoping review on emerging knowledge synthesis methods. *J Clin Epidemiol* 2016;73:15–18.
15. Tricco AC, Antony J, Soobiah C, *et al*. Knowledge synthesis methods for generating or refining theory: a scoping review reveals that little guidance is available. *J Clin Epidemiol* 2016;73:36–42.
16. Tricco AC, Antony J, Soobiah C, *et al*. Knowledge synthesis methods for integrating qualitative and quantitative data: a scoping review reveals poor operationalization of the methodological steps. *J Clin Epidemiol* 2016;73:29–35.
17. Tricco AC, Soobiah C, Antony J, *et al*. A scoping review identifies multiple emerging knowledge synthesis methods, but few studies operationalize the method. *J Clin Epidemiol* 2016;73:19–28.
18. Wong G. Knowledge synthesis approaches–spoilt for choice? *J Clin Epidemiol* 2016;73:8–10.
19. Chalmers I, Glasziou P. Avoidable waste in the production and reporting of research evidence. *The Lancet* 2009;374:86–9.
20. Bouma GD. *The research process*. USA: Oxford University Press, 2004.
21. Yin RK. *Case study research: design and methods (applied social research methods)*. London and Singapore: Sage, 2009.
22. Setia M. Methodology series module 5: sampling strategies. *Indian J Dermatol* 2016;61:505–9.
23. Patton MQ. *Qualitative evaluation and research methods*. SAGE Publications, 1990.
24. Saunders B, Sim J, Kingstone T, *et al*. Saturation in qualitative research: exploring its conceptualization and operationalization. *Qual Quant* 2018;52.
25. Guest G, Bunce A. How many interviews are enough?: an experiment with data saturation and variability. *Field Methods* 2006;18:59–82.
26. Hennink MM, Kaiser BN, Marconi VC. Code saturation versus meaning saturation: how many interviews are enough? *Qual Health Res* 2017;27:591–608.
27. Shenton A. *Strategies for ensuring Trustworthiness in qualitative research projects*. 22, 2004.
28. Britten N. Qualitative interviews in medical research. *BMJ* 1995;311:251–3.
29. Kvale S, Brinkmann S. *Interviews: learning the craft of qualitative research interviewing*. Sage, 2009.
30. Gill P, Stewart K, Treasure E, *et al*. Methods of data collection in qualitative research: interviews and focus groups. *Br Dent J* 2008;204:291–5.
31. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.
32. Guba EG, Lincoln YS. *Naturalistic inquiry*, 1985.
33. Glonti K, Hren D. Editors' perspectives on the peer-review process in biomedical journals: protocol for a qualitative study. *BMJ Open* 2018;8:e020568.
34. Macnee CL, McCabe S. *Understanding nursing research: using research in evidence-based practice*. Lippincott Williams & Wilkins, 2008.
35. Holloway I, Wheeler S. Ensuring trustworthiness and quality. In: Holloway I, Wheeler S, eds. *Research in nursing*. 2nd edn. India: Blackwell Publishing, 2002: 250–63.
36. AnneyVN. Ensuring the quality of the findings of qualitative research: looking at trustworthiness criteria. *JETERAPS*;5:272–81.
37. Tobin GA, Begley CM. Methodological rigour within a qualitative framework. *J Adv Nurs* 2004;48:388–96.
38. Connelly LM. Trustworthiness in qualitative research. *Medsurg Nursing* 2016;25:435–7.
39. Robinson KA, Saldanha IJ, McKoy NA. Identification of research gaps from evidence-based guidelines: a pilot study in cystic fibrosis. *Int J Technol Assess Health Care* 2011;27:247–52.

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Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study

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ABSTRACT

Introduction Mapping the current body of evidence including what is missing helps provide a better understanding of what research is available, ongoing and needed and should be prioritised. Identifying research gaps can inform the design and conduct of health research by providing additional context information about the body of evidence in a given topic area. Despite the commonly used term 'research gap' in scientific literature, little is written on how to find a 'research gap' in the first place. Moreover, there is no clear methodological guidance to identify and display gaps.

Objective This study aimed to explore how key stakeholders define research gaps and characterise methods/practices used to identify and display gaps in health research to further advance efforts in this area.

Design This was an exploratory qualitative study using semistructured in-depth interviews. The study sample included the following stakeholder groups: researchers, funders, healthcare providers, patients/public and policymakers. Interview transcripts were subjected to thematic analysis.

Results Among the 20 interviews conducted (20 participants), a variety of research gap definitions were expressed (ie, five main themes, including gaps in information, knowledge/evidence gaps, uncertainties, quality and patient perspective). We identified three main themes for methods used to identify gaps (primary, secondary and both primary and secondary) and finally six main themes for the methods to display gaps (forest plots, diagrams/illustrations, evidence maps, mega maps, 3IE gap maps and info graphics).

Conclusion This study provides insights into issues related to defining research gaps and methods used to identify and display gaps in health research from the perspectives of key stakeholders involved in the process. Findings will be used to inform methodological guidance on identifying research gaps.

Strengths and limitations of this study

- This study used qualitative methodology that provided an in-depth understanding of key stakeholders' perspectives and experiences in identifying, describing and displaying gaps in health research.
- The study benefited from having a variety of different stakeholders participating in semistructured interviews, which provided a wider scope of perspectives and experiences in identifying, describing and displaying gaps in health research.
- This study could have benefited from involving patient/public perspectives to inform the design of the study to improve the importance and relevance of the findings for this population.

BACKGROUND

Identifying research gaps can help inform the design and conduct of health research, practice and policies by providing a better understanding of the current body of evidence. Healthcare decisions for individual patients, public health policies and clinical guidelines should be informed by the best available research while taking into account research gaps.

The identification of research gaps has no well-defined process, although research gaps serve as the basis in developing a new research question and informing future research, healthcare delivery and health policies. In addition, research gaps in healthcare do not necessarily align directly with research needs. Hence, research gaps are critical in that knowledge gaps substantially inhibit the decision-making ability of stakeholders such as patients, healthcare providers and policymakers, thus creating a need to fill the knowledge gap.[1]

Moreover, identifying and characterising research gaps often highlight multiple competing gaps that are worthwhile to be explored.[1] Initiatives such as the James Lind Alliance (JLA), UK Database of Uncertainties about the Effects of Treatments, Cochrane Agenda and Priority Setting Methods Group, and Evidence-based Research Network are

some examples of existing efforts to identify and prioritise research gaps in health.[2]

The term 'research gap' is not well defined, and its meaning can differ depending on the researcher and research context. A recent scoping review of methods used to identify, prioritise and display gaps in health research reported 12 different definitions related to gaps in health research, each describing research gaps differently.[2] This finding shows the ambiguity of the term 'research gap' and the different practices it may relate to.

As a basis for further exploring and understanding 'research gaps', we start from the definition given by the National Collaborating Centre for Methods and Tools in Canada based on the work by Robinson *et al*, whereby a research gap is defined as a topic or area for which missing or insufficient information limits the ability to reach a conclusion for a question.[3] Given the different meanings and definitions of research gaps identified in the scoping review,[2] we considered it important to further explore key stakeholders' perspectives to better understand the topic area. Clearly defining the type of research gap can help determine how to better identify, characterise, prioritise and address research gaps.

Different methods for identifying research gaps reported include scoping reviews and umbrella reviews for mapping and summarising evidence. These methods have an explicit aim of identifying research gaps in a broad area compared with systematic reviews, which focus on answering a specific research question.[4–8] Furthermore, investigating experiences with practices/methods used to identify research gaps can inform explicit methodological approaches in identifying and describing research gaps. This investigation can enhance practices of different stakeholder groups (ie, health professionals, health commissioners, researchers, patients/public and decision-makers) when addressing areas of uncertainty within the research problem and topic area.[9]

The specific objectives of the study were to (1) investigate key stakeholders' knowledge and perceptions of and experiences with defining research gaps and (2) characterise methods/practices used to identify and display gaps in health research.

METHODS AND ANALYSIS

Qualitative study design

We conducted an exploratory qualitative study using semi-structured interviews. This method was selected to provide an in-depth understanding of key stakeholders' perspectives, experiences and practices in defining, identifying and displaying research gaps. This method also ensured that we explored key stakeholders' understanding and practices related to identifying research gaps through a variety of lenses from different stakeholder groups. In turn, this process provided multiple facets of research gap definitions and methodological practices to identify and display gaps.[10]

Table 1 Participant characteristics (n=20)

Category	No. (% of total)
Researcher	9 (45)
Methodologist	5 (25)
Data visualisation	3 (15)
PhD student	1 (5)
Health practitioner	6 (30)
Healthcare provider	5 (25)
Public health professional	1 (5)
Oversight bodies	3 (15)
Health policy-maker	2 (10)
Funding body	1 (5)
Patients/public	2 (10)

Study sample and recruitment

We used purposive sampling to ensure that the perspectives of all identified stakeholder groups were represented. Purposive sampling is widely used in qualitative research to identify and select information-rich cases. The study sample included the following stakeholder groups: researchers, funders, healthcare providers, patients/public and policy-makers. The stakeholder groups were determined according to the findings of a previously conducted scoping review[2] and organised into three main categories focusing on the use of evidence to inform health policy, health practice and health research (table 1). A detailed description of participant categories was given in the previously published study protocol.[11] Study participants were recruited via contacts and organisations identified in the scoping review, relevant scientific publications, existing professional networks (eg, H2020 International Training Network 'Methods in Research on Research') and contacts from conference attendance (eg, Evidence Live and Cochrane Colloquium).

The main inclusion criteria for the study were as follows:

1. Adults aged ≥18 years (researchers, funders, healthcare providers, patients/public and policy-makers).
2. Experience with the use of evidence to inform health decisions/choices, policy, practice or research.
3. Ability to converse in English.
4. Consent for research.

The sample size for qualitative studies usually depends on the point when data saturation is reached (ie, the point when new data do not add to a better understanding of the studied phenomenon but rather repeat what was previously expressed[12]). Considering that the point of saturation cannot be specified in advance, we planned to conduct between 14 and 28 interviews, owing to usual points of data saturation reported in qualitative studies.[11] The point of data saturation was determined based on the seven parameters identified by Hennink *et al*,[13 14] including the study purpose, population, sampling strategy, data quality, type of codes, code book and saturation goal, and focus retrieved from the study. These parameters were

discussed throughout the study primarily between the lead researcher (LN) and the senior researcher (DH).

Data collection and recording

Semistructured interviews were used for this study. The main reason for selecting semi-structured interviews was to allow for specific areas to be addressed while giving the interviewees the opportunity to reflect on their experiences and perspectives related to defining, identifying and presenting research gaps that are relevant to them and that may not have been explored or anticipated by the researcher(s).[15]

The guide was developed by focusing on exploring key stakeholders' perspectives and experiences with the following key areas:

1. Participant background information.
2. Definitions of research gaps.
3. Knowledge and perceptions of and experiences with methods/practices used to identify and display gaps in health research to inform further health policy, practice and research.

These three categories were developed with information from the scoping review to guide the questions. The interview topic guide was piloted before data collection. It was also adapted according to key stakeholder groups to ensure that it was meaningful to their background and to gather more relevant information based on their experiences and knowledge.[16]

The semistructured interview guide contained two levels of questions: main themes and follow-up questions. The main themes covered the general content of the research gaps aimed at encouraging participants to speak freely about their perceptions, experiences and practices. The follow-up questions were used as prompts and probes aiming to follow respondents' answers and to investigate the issues raised more in depth. The interview guide covered the main topics of the study, providing a focused structure for the discussion during the interview.[17]

We conducted in-person, telephone and teleconference interviews. In-person interviews were conducted with participants residing or reachable in London, UK, and other participants were interviewed via telephone or teleconference (for the interview guide for both in-person and teleconference interviews, see online supplemental appendix).

All interviews were digitally recorded, transcribed verbatim and anonymised. The lead researcher (LN) transcribed two interviews to help inform the analytical process, and the other audio files were transcribed by a professional transcription agency licensed from the University of Liverpool.

Data analysis

We used analytical categories to describe and explain definitions, experiences and practices reported among the groups of participants. All data relevant to each category (defining research gaps, experiences with methods/practices used to identify and display gaps in health research) were identified and examined to ensure that each data item was checked accordingly.

Our approach was based on the thematic analysis outlined by Braun and Clarke.[18] The steps included the following: (1) transcription and checking transcripts with recordings for accuracy, (2) open coding from interview responses performed by two researchers independently (LN and DH), (3) agreement of initial codes discussed among the researchers and an initial codebook developed, (4) developing the code structure used for analysing the remaining responses with openness that included new codes and refined existing ones and (5) themes and subthemes identified from the final code structure and their relationships presented.[18]

The initial coding framework for our analysis started from broad categories identified in the previous scoping review with which the interviews were structured. Within these broad categories (ie, defining research gaps, experiences with methods/practices used to identify and display gaps in health research), analytical categories were inductively derived from the data. In this sense, our approach includes both top-down and bottom-up development of analytical categories and themes.

QSR International's NVivo V.12 qualitative data analysis software was used for data management and analysis.

Ensuring study quality

To further ensure rigour and trustworthiness, the study was guided by Lincoln and Guba 's concepts for defining and investigating quality in qualitative research that can be considered parallel to quantitative research concepts of validity and reliability.[13 19 20] The concepts include credibility, transferability, dependability, confirmability, audit trails and reflexivity. They are interrelated, and thinking through them from the onset and incorporating them into a study improve the study's rigour.

The main researcher's (LN) past experience as a Public Health Advisor at a National Institute of Public Health in Europe influenced the conceptualisation and conduct of this study, including the interviews. Her previous role focused on knowledge production for the health sector and providing knowledge about the health status of the population, influencing factors and how the status can be improved. She recognised the need for evidence to inform research planning, implementation and evaluation. Therefore, the design and conduct of this study were informed by her previous role and influenced the development of the interview guide, and interpretation and reporting of study findings. Throughout the different steps of the study, she consulted a senior researcher (DH) to discuss all matters related to the study design, conduct and reporting.

Patient and public involvement

Patients and the public were not involved in the design or analysis of this study. However, we involved them as study participants and will disseminate the study findings that

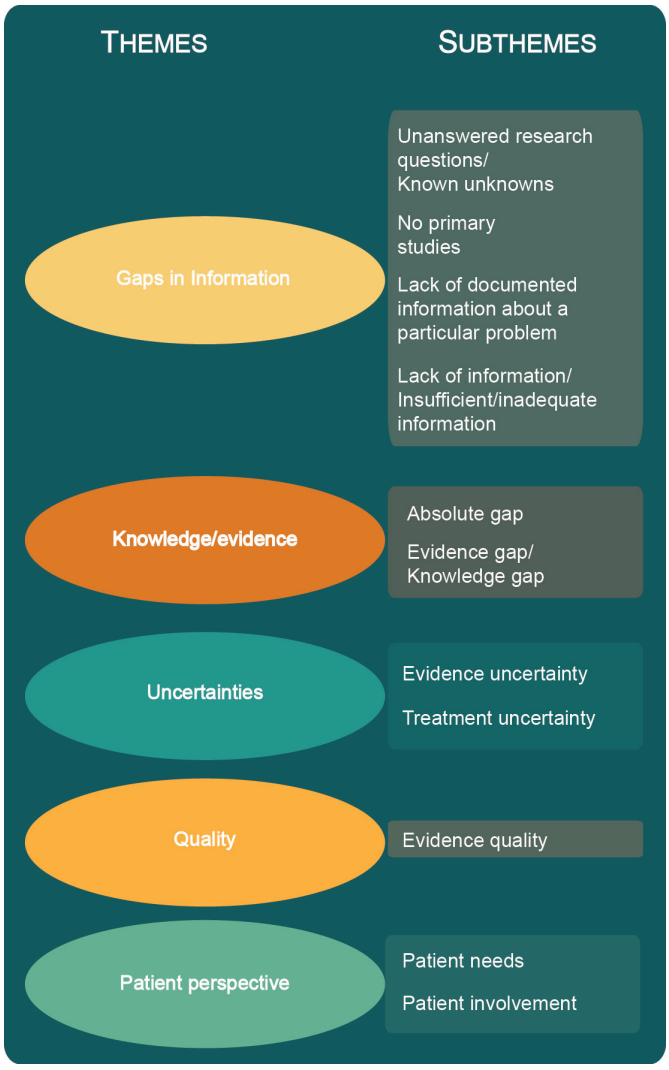

Figure 1 Reported descriptions of gaps in health research.

pertain to them using a patient/public online platform, peopleinresearch.org.

RESULTS

Among the 30 key stakeholders contacted, 20 agreed to participate in the study. Hence, we conducted 20 interviews with 20 participants involved in using evidence for informing health policy, practice or future research (table 1).

Definitions of gaps in health research

We first explored what participants reported as gaps in health research. Given the nature of our interest, all participants' answers were grouped under a single theme 'Definitions of Gaps in Health Research'. However, the focus of the definitions differed, and within this main theme, we identified five subthemes related to gaps in health research described by the participants (ie, gaps in information, knowledge/evidence-related gaps, quality of evidence, uncertainties and patient-related gaps; summarised in figure 1). The discrepancies and similarities of terms used are further illustrated in the online supplemental appendix. Terms ranged from lack of information/insufficient information, known unknowns/unanswered research questions and evidence uncertainty to treatment uncertainty, among others.

We identified some similarities among the participants on how they defined research gaps, for example, researchers and oversight bodies mainly defined gaps in health research as a lack of information/insufficient information, known unknowns and no primary studies (more information can be found in online supplemental appendix). Patient/public participants defined research gaps in a much more literal manner, for example, 'The gap is to get more patients involved in doing … clinical trials; have [someone] at the beginning introduce me, [educate me], [provide] awareness [because] I didn't know what [a clinical trial] was. I [didn't] know what they're talking about' (patient/public person, PPI01) and 'Get me involved in co-production. That is the gap that is missing in clinical research' (patient/public person, PPI01). The most common description research participants provided was the absence of scientific information to answer a research question, for example,

> An area where there is missing or … insufficient information. And because of this … you cannot reach a conclusion for a question. So … it is a field, it is an area, a question an issue to which you don't have an appropriate answer because there is missing … information or the research that still needs to be done in that particular area. (Funding body, F01)

One participant related research gaps to quality of evidence by use of Grades of Recommendation, Assessment, Development and Evaluation (GRADE), an approach for rating the quality of evidence and grading the strength of recommendations in healthcare.[21] Another participant emphasised the importance of public and community involvement in gap identification to ensure that it takes into account their perspectives and contributions to the research ecosystem:

> existing knowledge but not documented is of key importance in understanding the current body of knowledge on a particular topic area …. Evidence gaps need to be defined not only by [the] research community but also according to the key stakeholders including community members. Community knowledge is of key importance to inform the evidence base. Further evaluation on research findings to characterise the nature of research gaps can be carried out by evaluating community perspectives and local evidence to confirm scientific evidence. (Health research PhD student, R01)

We identified variability in participant responses on how to define gaps in health research; this variability was mainly observed in individual responses for the three main categories (research, practice, and policy and funding).

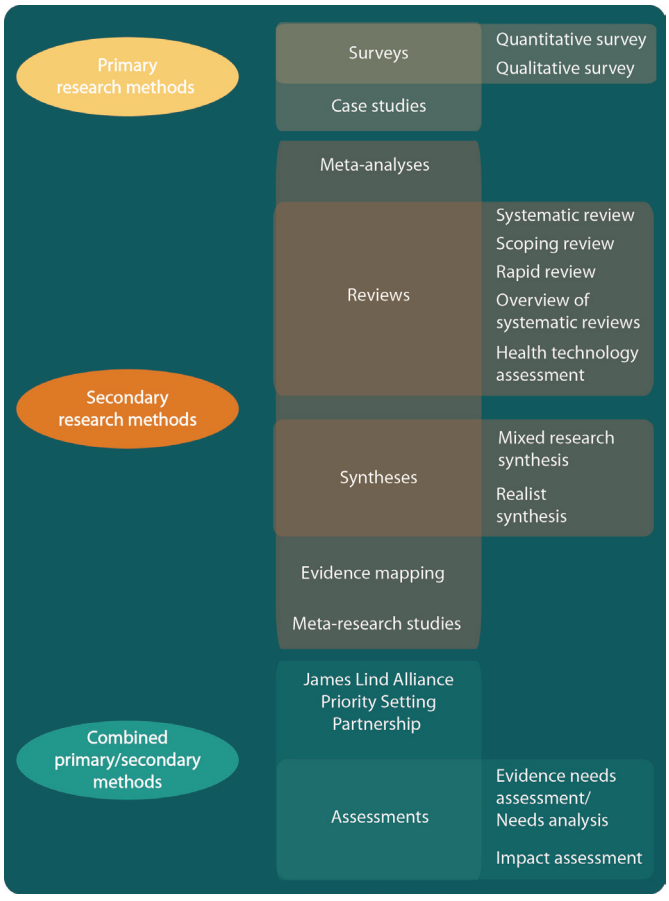

Figure 2 Methods used to identify gaps in health research.

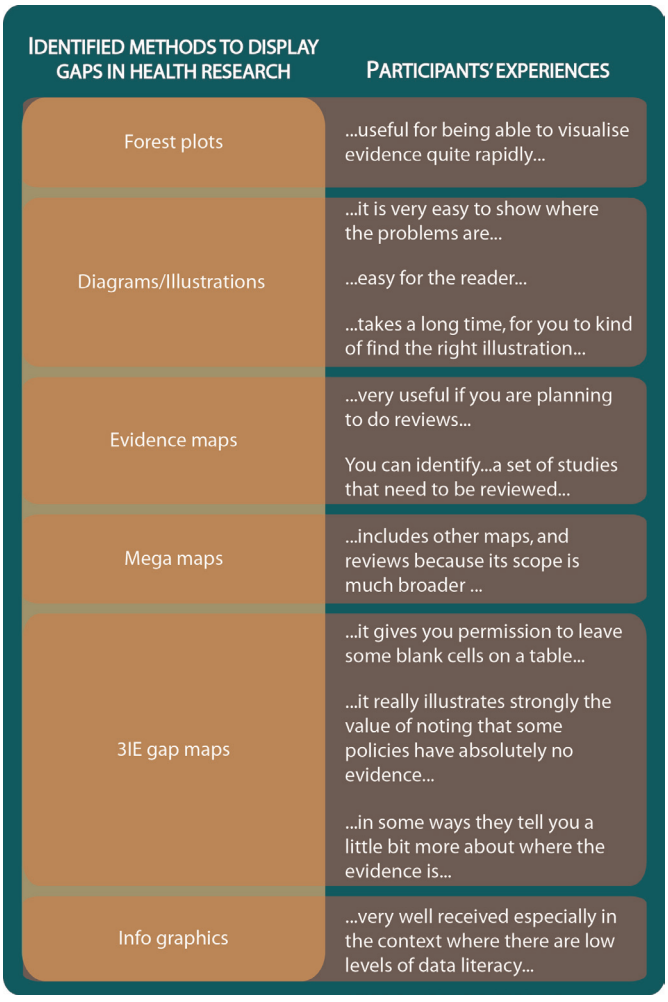

Figure 3 Methods used to display gaps in health research.

Methods to identify gaps in health research

Participants reported a range of applicable methods to identify gaps in health research (eg, surveys, reviews, syntheses, priority-setting partnerships and assessments) as shown in figure 2. The methods were also characterised by the different research methodologies used (ie, primary, secondary and both). Participants also expressed their difficulty in identifying research gaps, for example,

> It is really difficult to identify research gaps. Lots of people you know will try and use the discussion section from research, [whereas] other authors have asked for further research, but in my experience that has not been a very useful method because sometimes authors will write that you know without really seeing or understanding that there has been something similar done in that field. (Health research methodologist, R02)

The variety of identified methods reflected the state of the field in the sense of the wide array of methods currently used, in line with the variety of specific goals of studies on research gaps (figure 1). The difficulty in identifying research gaps raised by participants, together with the plurality of definition of gaps and range of methodologies, may, however, also reflect a possible lack of consensus and guidance on what method would be best suited for a given objective.

Methods to display gaps in health research

Participants referred to a number of different methods used to display gaps in health research (ie, forest plots, diagrams/illustrations, evidence maps, mega maps, 3IE gap maps and info graphics) (figure 3). Participant perspectives varied; one of the interviewees pointed out,

> I think with the growth of technology, it is very important to use sophisticated methods to better communicate evidence for policy-making and decision-making. I think the availability of evidence is not enough on its own and finding different methods to communicate is important, not only the analysis and findings but also sharing it in different platforms online for a greater audience. (Health policy and guideline developer, P02)

Another participant highlighted that one of the key benefits of visually presenting research is being able to immediately see what information is available and missing.

The participants mainly expressed the importance of using data visualisation in research; there was a common understanding on the use of data visualisation as a whole, particularly with the growth of technology and the need to capitalise on it. The main challenges expressed were

how to identify an appropriate visualisation to present the research and also how to effectively present data. We summarise these general experiences with data visualisation in health research in figure 3 and the online supplemental appendix.

DISCUSSION

This study provides insight into issues related to defining, identifying and displaying research gaps in health from the perspectives of key stakeholders. The findings indicate several definitions of gaps in health research and methods used to identify and display research gaps.

Our study confirmed the ambiguity in defining research gaps and methodological approaches to identify[3 22] and display research gaps.[2] The methods used to identify research gaps were closely linked to the definition of research gaps. For example, the JLA method of gap identification and setting priorities for research begins by clearly defining what the alliance refers to as evidence uncertainty, that is, there is no up-to-date, reliable systematic review of research evidence addressing the uncertainty or showing that uncertainty.[23] This step further informs the rest of the methodology used and is critical in identifying treatment uncertainties and determining future research priorities. This method combines both primary and secondary approaches and not only identifies research gaps but also verifies them across different relevant stakeholders, including researchers, patients, their carers and clinicians, to ensure the relevance and potential benefit to them.[23] This verification is important, given that some research gaps may be of key interest to researchers but have little relevance and importance to patients or the public, who should be the main beneficiaries of research to improve their health and well-being.

The overall method to identify research gaps involved primary, secondary or both approaches (figure 2). Most of the participants mentioned the use of secondary research methods; this is in accordance with the research that has been conducted on research gaps, which has also primarily focused on the use of secondary research and developed frameworks for identifying research gaps.[2 3 8 24 25] The most commonly adopted framework involves identifying research gaps from systematic reviews using the Population, Intervention, Comparison and Outcome framework to characterise a research gap.[3] The other framework involves identifying research gaps in qualitative literature reviews.[25] In addition, the GRADE approach for rating the quality of evidence and grading the strength of recommendations in healthcare[21] presents the use of a prominent framework for evaluating the certainty of evidence that can inform the research gap and characterise it.[26] Moreover, scoping reviews are commonly used, and the definition includes aiming to identify research gaps by mapping the current body of evidence. These examples focus on the use of secondary research methods, but we lack studies that specifically explore the use of primary or both primary and secondary methods to identify research gaps, yet these methods equally exist and are being used. Additional exploration of applicable methods for identifying gaps can improve their usefulness and relevance in health research.

In summary, this study showed that research gaps need to be defined by researchers and confirmed by different research stakeholders such as patients and the public to ensure societal relevance and importance.[1] We also found that clearly defining research gaps can provide information on the most appropriate methodological approach to adopt in identifying and displaying gaps, for example, for exploring research gaps in a specific or broad area. For a specific area, a systematic review can be considered, and within a broad area, an umbrella review can be considered. The study also showed that the use of both primary and secondary methods (JLA method) to identify gaps is the most robust method for gap identification. The main reported advantage of this method is that it identifies gaps (treatment uncertainties) and involves different stakeholders, including patients and the public, to confirm and prioritise gaps. The main disadvantage is that it is labor-intensive (requires a team of different specialists) and expensive (administrative support, meeting rooms and catering, among others) compared with secondary methods (evidence synthesis) or primary methods (survey).

Participants mainly expressed the importance of data visualisation in communicating research; no specific methods or formats to present gaps were expressed. Thus, the use of data visualisation is desirable among different stakeholders, particularly researchers, when communicating research, although we found few examples of experiences with developing and using data visualisation. The participants mainly expressed their difficulty in finding the right tool to use to present research findings.

Finally, although scientific articles often refer to the existence of research gaps in studies, few respondents were able to define research gaps, unless contextualising them within a specific study or area, or methods of identification. Fully understanding research gaps in health research and adequately addressing them are difficult. In this study, we highlighted three key items on the topic: (1) clearly defining research gaps provides a context to understand better what the gaps are and what they are caused by; (2) a clear definition of research gaps can inform the methods used to identify research gaps, similar to how a clear research question can inform the research study methodology; and (3) on adopting the most appropriate methods to identify research gaps, finding the right visualisation to communicate them effectively is important. Last but not least, public involvement, when applicable, is needed to verify that gaps are important and relevant to the public.

To conclude, our study found that various methods can be used to identify gaps (ie, primary, secondary and both primary and secondary). Of all the methods used to identify gaps, secondary methods are the most common, specifically systematic reviews, which are considered the gold standard in that they address a highly focused question related to the existing evidence and thus present difficulties for explicitly identifying research gaps in a general area.[3 8 27] Other secondary

research methods reported were overviews of reviews, also known as umbrella reviews, scoping reviews and evidence mapping. Overviews of reviews focus on a much broader area, compiling evidence from multiple reviews into one accessible and usable document and highlighting other reviews within the specified topic area.[28 29] Given the resource requirements of formal evidence reviews, topic prioritisation is needed to best allocate resources to those areas deemed the most relevant for the health system. Regardless of the topic, the prioritisation process is likely to be stakeholder-dependent. Priorities for evidence synthesis will vary depending on the mission of the healthcare system and the local needs of the healthcare stakeholders.[1] Hence, using both primary and secondary methods is the most robust because it involves the participation of patients, caregivers and healthcare and social care professionals in identifying research questions and then prioritising them using a combination of primary and secondary research.[30–49]

To advance efforts in identifying research gaps, further work and different study designs are needed to take this work to the next step, to find consensus on definitions and different practices for methods in identifying research gaps. Subsequently, also assessing the best methods according to different stakeholders will be informative and important.

One of the main challenges of this study was that because the topic area is still very vague and unclear, the recruitment and interview process was challenging. Therefore, this study was primarily limited to what participants were familiar with and not necessarily representative of the full scope of the status of health researchers, health practitioners, oversight bodies and patients/public. A more generalisable understanding of this topic area would require a larger sample of participants and methodology, such as a Delphi survey, and/or a priority-setting partnership with representatives using evidence to inform policy, practice and research. This study would also have benefited from widening the scope of the stakeholder categories (use of evidence to inform health policy, health practice and health research).[2] This would have enriched our study findings and provided a wider view of stakeholder experiences outside our categories. Another limitation of this study is not including patients/public in designing the study. Including patient/public perspectives would have benefited the study design by being able to improve the importance and relevance of the findings for this population.

One of the main strengths of the study is improving the definition of research gaps and subsequently improving the accurate reporting of research gaps to elucidate the characteristics, which can help in evidence-based decisions. For example, a decision based on a research gap contributing to lack of primary research on a specific health problem can differ from the one based on a research gap related to lack of secondary research summarising the research. Hence, all these factors regarding research gaps need to be highlighted if they are known and made explicit when disseminating and communicating research. In addition, providing more information on what the gap represents may inform users of evidence of more specific information about the research gap and how it can be addressed more accurately.

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Competing interests None declared.

Patient consent for publication Not required.

Ethics approval Informed consent was obtained in accordance with the University of Liverpool Ethics Committee board requirements. Verbal consent was sought for phone interviews and written consent for in-person interviews. Confidentiality and data protection will be ensured in accordance with the University of Liverpool Ethics Committee board. All participant information will be anonymised, and hard-copy data will be stored in a locked unit. Soft-copy material will be stored in a password-protected file. On completion of the study and publication of the study results, all study material will be stored and disposed of according to the rules and regulations of the University of Liverpool. The study protocol was stored in the data repository Zenodo. The research obtained ethical approval from the University of Liverpool, UK. This research project is part of a doctoral thesis of the PhD fellow (LN).

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REFERENCES

1 Hempel S, Gore K, Belsher B. Identifying research gaps and prioritizing psychological health evidence synthesis needs. *Med Care* 2019;57 Suppl 10 Suppl 3:S259–64.
2 Nyanchoka L, Tudur-Smith C, Thu VN, *et al*. A scoping review describes methods used to identify, prioritize and display gaps in health research. *J Clin Epidemiol* 2019;109:99–110.
3 Robinson KA, Saldanha IJ, McKoy NA. Development of a framework to identify research gaps from systematic reviews. *J Clin Epidemiol* 2011;64:1325–30.
4 Snilstveit B, Vojtkova M, Bhavsar A, *et al*. Evidence & Gap Maps: A tool for promoting evidence informed policy and strategic research agendas. *J Clin Epidemiol* 2016;79:120–9.
5 Pham MT, Rajić A, Greig JD, *et al*. A scoping review of scoping reviews: advancing the approach and enhancing the consistency. *Res Synth Methods* 2014;5:371–85.
6 Arksey H, O'Malley L. Scoping studies: towards a methodological framework. *Int J Soc Res Methodol* 2005;8:19–32.
7 Levac D, Colquhoun H, O'Brien KK. Scoping studies: advancing the methodology. *Implement Sci* 2010;5:69.

8 Carey T, Yon A, Beadles C, *et al*. *Prioritizing future research through examination of research gaps in systematic reviews*. Prepared for the Patient-Centered Outcomes Research Institute, 2012.
9 Bouma GD, Ling R. *The research process*. USA: Oxford University Press, 2004.
10 Yin RK. *Case study research: design and methods (applied social research methods*. London and Singapore: Sage, 2009.
11 Nyanchoka L, Tudur-Smith C, Porcher R, *et al*. Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study protocol. *BMJ Open* 2019;9:e027926.
12 Saunders B, Sim J, Kingstone T, *et al*. Saturation in qualitative research: exploring its conceptualization and operationalization. *Qual Quant* 2018;52:1893–907.
13 Shenton AK. Strategies for ensuring trustworthiness in qualitative research projects. *EFI* 2004;22:63–75.
14 Hennink MM, Kaiser BN, Marconi VC. Code saturation versus meaning saturation: how many interviews are enough? *Qual Health Res* 2017;27:591–608.
15 Britten N. Qualitative interviews in medical research. *BMJ* 1995;311:251–3.
16 Kvale S, Brinkmann S. *Interviews: learning the craft of qualitative research interviewing*. Sage, 2009.
17 Gill P, Stewart K, Treasure E, *et al*. Methods of data collection in qualitative research: interviews and focus groups. *Br Dent J* 2008;204:291–5.
18 Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006;3:77–101.
19 Glonti K, Hren D. Editors' perspectives on the peer-review process in biomedical journals: protocol for a qualitative study. *BMJ Open* 2018;8:e020568.
20 Lincoln YS, Guba EG. *Naturalistic inquiry*. Beverly Hills: Sage, 1985: 289–331.
21 Guyatt GH, Oxman AD, Schünemann HJ, *et al*. Grade guidelines: a new series of articles in the Journal of clinical epidemiology. *J Clin Epidemiol* 2011;64:380–2.
22 Rudan I, Campbell H, Marušić A, *et al*. Assembling GHERG: Could 'academic crowd–sourcing' address gaps in global health estimates? *J Glob Health* 2015;5.
23 Boney O, Bell M, Bell N, *et al*. Identifying research priorities in anaesthesia and perioperative care: final report of the joint National Institute of academic Anaesthesia/James Lind alliance research priority setting partnership. *BMJ Open* 2015;5:e010006.
24 Robinson KA, Saldanha IJ, McKoy NA. Identification of research gaps from evidence-based guidelines: a pilot study in cystic fibrosis. *Int J Technol Assess Health Care* 2011;27:247–52.
25 Müller-Bloch C, Kranz J. *A framework for Rigorously identifying research gaps in qualitative literature reviews*. Proceedings/ International Conference on Information Systems, 2015.
26 Scott NA, Moga C, Harstall C, Carmen M, Christa H, *et al*. Using health technology assessment to identify research gaps: an unexploited resource for increasing the value of clinical research. *Healthc Policy* 2008;3:109.
27 Tricco AC, Zarin W, Ghassemi M, *et al*. Same family, different species: methodological conduct and quality varies according to purpose for five types of knowledge synthesis. *J Clin Epidemiol* 2018;96:133–42.
28 Pollock M, Fernandes RM, Becker LA, *et al*. What guidance is available for researchers conducting overviews of reviews of healthcare interventions? A scoping review and qualitative metasummary. *Syst Rev* 2016;5:190.
29 Grant MJ, Booth A. A typology of reviews: an analysis of 14 review types and associated methodologies. *Health Info Libr J* 2009;26:91–108.
30 Yoshida S. Approaches, tools and methods used for setting priorities in health research in the 21(st) century. *J Glob Health* 2016;6:010507.
31 Welsh E, Stovold E, Karner C, *et al*. Cochrane airways group reviews were prioritized for updating using a pragmatic approach. *J Clin Epidemiol* 2015;68:341–6.
32 Wald HL, Leykum LK, Mattison MLP, *et al*. A patient-centered research agenda for the care of the acutely ill older patient. *J Hosp Med* 2015;10:318–27.
33 Ingram JR, Abbott R, Ghazavi M, *et al*. The hidradenitis suppurativa priority setting partnership. *Br J Dermatol* 2014;171:1422–7.
34 Lophatananon A, Tyndale-Biscoe S, Malcolm E, *et al*. The James Lind alliance approach to priority setting for prostate cancer research: an integrative methodology based on patient and clinician participation. *BJU Int* 2011;108:1040–3.
35 Gadsby R, Snow R, Daly AC, *et al*. Setting research priorities for type 1 diabetes. *Diabet Med* 2012;29:1321–6.
36 Heazell AEP, Whitworth MK, Whitcombe J, *et al*. Research priorities for stillbirth: process overview and results from UK stillbirth priority setting partnership. *Ultrasound Obstet Gynecol* 2015;46:641–7.
37 Pollock A, St George B, Fenton M, *et al*. Top 10 research priorities relating to life after stroke--consensus from stroke survivors, caregivers, and health professionals. *Int J Stroke* 2014;9:313–20.
38 Rees SE, Chadha R, Donovan LE, *et al*. Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus. *Can J Diabetes* 2017;41:156–63.
39 van Middendorp JJ, Allison HC, Ahuja S, *et al*. Top ten research priorities for spinal cord injury: the methodology and results of a British priority setting partnership. *Spinal Cord* 2016;54:341–6.
40 Meremikwu M, Udoh E, Nwagbara B, *et al*. Priority setting for systematic review of health care interventions in Nigeria. *Health Policy* 2011;99:244–9.
41 Buckley BS, Grant AM, Glazener CMA. Case study: a patient-clinician collaboration that identified and prioritized evidence gaps and stimulated research development. *J Clin Epidemiol* 2013;66:483–9.
42 Buckley BS, Grant AM, Tincello DG, *et al*. Reaching a consensus and ranking research priorities in urinary incontinence. *Nurs Times* 2010;106:36–7.
43 Mitnick CD, Rodriguez CA, Hatton ML, *et al*. Programmatic management of drug-resistant tuberculosis: an updated research agenda. *PLoS One* 2016;11:e0155968.
44 Pollock A, St George B, Fenton M, *et al*. Development of a new model to engage patients and clinicians in setting research priorities. *J Health Serv Res Policy* 2014;19:12–18.
45 van Furth EF, van der Meer A, Cowan K. Top 10 research priorities for eating disorders. *Lancet Psychiatry* 2016;3:706–7.
46 Chapman E, Reveiz L, Sangalang S, *et al*. A survey study identified global research priorities for decreasing maternal mortality. *J Clin Epidemiol* 2014;67:314–24.
47 Gierisch JM, Myers ER, Schmit KM, *et al*. Prioritization of research addressing management strategies for ductal carcinoma in situ. *Ann Intern Med* 2014;160:484–91.
48 Yu T, Li T, Lee KJ, *et al*. Setting priorities for comparative effectiveness research on management of primary angle closure: a survey of Asia-Pacific clinicians. *J Glaucoma* 2015;24:348–55.
49 Al-Khatib SM, Gierisch JM, Crowley MJ, *et al*. Future research prioritization: implantable cardioverter-defibrillator therapy in older patients. *J Gen Intern Med* 2015;30:1812–20.

Chapter 5: Developing Methodological Guidance

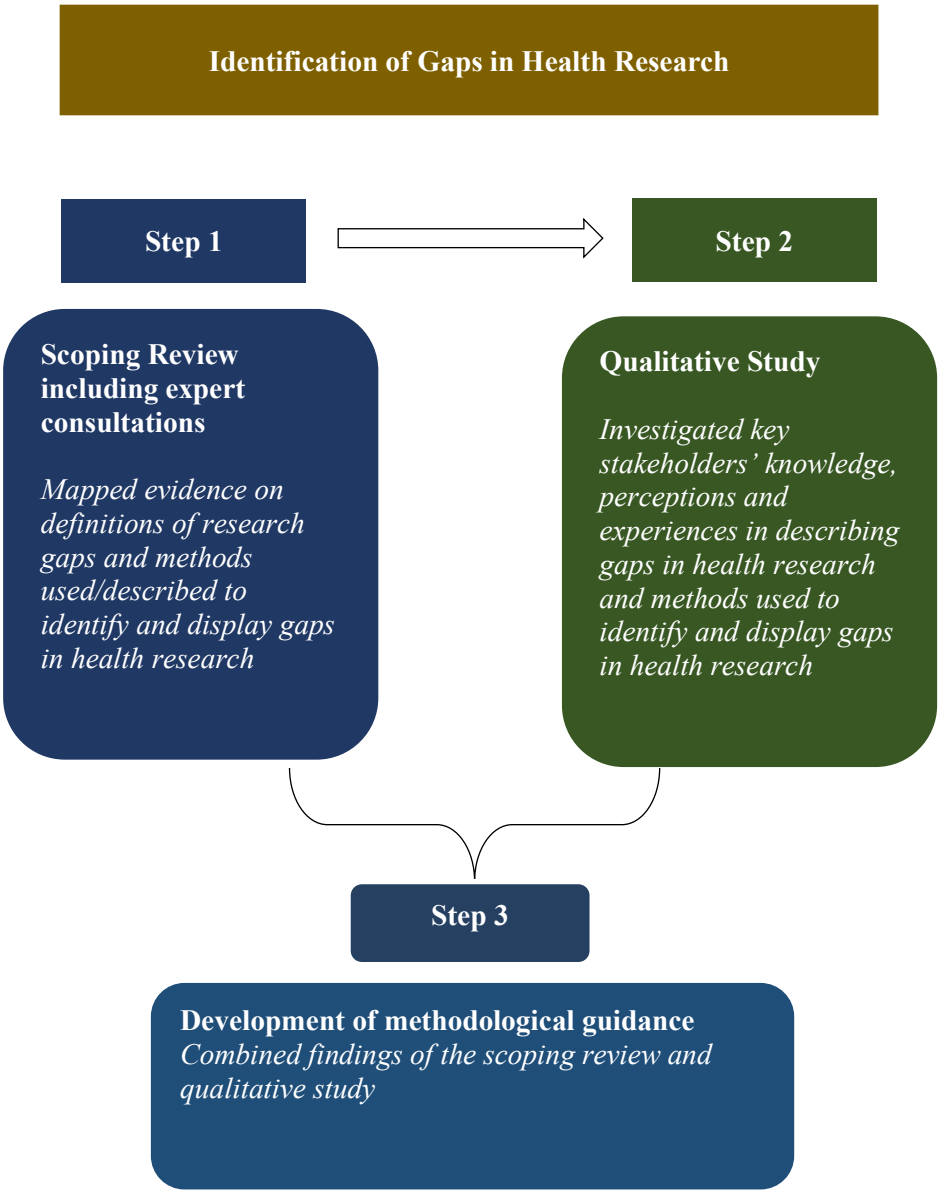
5.1. Background

The overall PhD project aimed to develop methodological guidance to identify gaps in health research. The first step in the process was a scoping review describing methods used to identify, prioritize and display gaps in health research in scientific literature. The scoping review mapped evidence on different definitions reported for the term "research gap" as well as methods used to identify research gaps and determine research priorities and display research gaps or research priorities [104]. The second step was to conduct a qualitative study using semi-structured in-depth interviews aimed to investigate the experience of key stakeholders (i.e., researchers, funders, clinicians, clinical guideline developers, public health professionals, commissioners, patients/the public and policy-makers) with defining research gaps and practices/methods used to identify and display research gaps[120]. The final step is this chapter, which involved combining findings from the scoping review and qualitative study findings on defining and identifying and displaying research gaps to inform the development of methodological guidance to identify gaps in health research.

5.2. Method

The steps undertaken to inform the methodological guidance involved synthesizing the study findings of the scoping review and qualitative study, further illustrated on (Figure 6).

Figure 6. Overall PhD project design used to develop methodological guidance



5.3. Results

In this section, I present the combination of our two study findings (i.e., the scoping review and qualitative study) on 1) defining research gaps, 2) methods used to identify research gaps, and 3) methods to display research gaps.

5.3.1. Combined findings on key definitions of types of research gaps

This section presents terms related to gaps in health research and definitions linked to these terms from the scoping review and qualitative study.

Table 10. Identified definitions of types of research gaps

Terms related to research gaps	Description	Key characteristics
Research/evidence/knowledge gaps	- Evidence is missing from a body of research on a particular topic that could otherwise potentially answer the questions of decision-makers (clinicians, other practitioner groups, administrators, policy-makers) [26] - The evidence base inadequately addresses a key question [27] - Not much information is available and/or there is a lot of uncertainty about the accuracy of the existing estimates/evidence [4] - Additional research is needed, from policy-makers perspectives, to	- Missing evidence - Inadequate evidence - Insufficient evidence - Contradictory evidence - Additional research needed - Uncertain evidence/estimates

	address the evidence gap in the available primary research [26]	
Synthesis/unidentified gaps	- Little or no evidence from systematic reviews is available and could be a valuable resource to inform the evidence base in a particular area [29, 30] - Lack of up-to-date and conclusive systematic reviews at low risk of bias mapped to a clinical question [31]	- Little or no evidence from reviews is available and could be a valuable resource to inform the evidence base in a particular area - Lack of up-to-date and conclusive systematic reviews
Treatment uncertainty	- Lack of up-to-date, reliable systematic reviews of research evidence addressing the uncertainty about the effects of treatment, and/or up-to-date systematic reviews of research evidence show that uncertainty exists [32]	- Lack of up-to-date, reliable systematic reviews addressing the uncertainty about the effects of treatment - Lack of up-to-date systematic reviews showing that uncertainty exists
Evidence uncertainty	- *Evidence that was uncertain, maybe lower quality, not directly applicable, not very large body of evidence, not a large number of studies etc. So you might have uncertain evidence and be less able to make a strong*	- lower quality, - not directly applicable, - Not a very large body of evidence - Unable to make a strong recommendation from the evidence

	recommendation... (PO1, health research methodologist)	
Practical knowledge gap (action–knowledge conflict gap)	- Professional behavior or practices deviate from research findings or are not covered by research [9, 22, 28]	- Professional behavior or practices deviate from research findings or are not covered by research
Empirical gap (evaluation void gap)	- Research findings need to be evaluated or empirically verified [9, 22, 28]	- Research findings need to be evaluated or empirically verified
Population gap	- Research regarding a population that is not adequately represented or under-researched in the evidence base or prior research (e.g., sex, race/ethnicity, age) [2]	- Research regarding a population that is not adequately represented or under-researched in the evidence base or prior research (e.g., sex, race/ethnicity, age)
Methodological gap (method and research design gap)	- A variation in research methods is required to generate new insights or to avoid distorted findings [9, 22, 28]	- A variation in research methods is required to generate new insights or to avoid distorted findings
Theoretical gap (theory–application void gap)	- Theory should be applied to certain research issues to generate new insights; theory is lacking, so a gap exists [9, 22, 28]	- Theory should be applied to certain research issues to generate new insights; theory is lacking

Absolute gap	- Few or no impact evaluations exist [29]	- There is no evidence whatsoever
Evidence quality	- Evidence that was uncertain, maybe lower quality, not directly applicable, not very large body of evidence, not a large number of studies etc.	- Evidence that was uncertain - Maybe lower quality - Not directly applicable - Not a very large body of evidence - Not a large number of studies - Articulating gaps in certainty and strength of evidence
Known unknowns	- Known unknowns are questions that perhaps the medical world knows the answer to but patients don't necessarily know the answer to.	- Information gaps
Lack of documented information	- Lack of documented information vs lack of information per se.	- Lack of documented information
Patient/public-related gap	- Lack of patient involvement in health research	- No patient involvement in design, conduct and reporting of study
Artificial gap	- *There is a gap between the research community and the real patient needs; therefore, the research gap needs to be confirmed by patients. It should not be an artificial gap*	- Arises when research gap is not representative of patient/public needs - Research community define gap, but it is not

	created by some stakeholders. (HP03, health practitioner)	confirmed by patients/public - A gap between the research community and the real patient needs - Patient needs

5.3.2. Combined findings of key methods to identify gaps in health research

Upon combining the scoping review and qualitative study we identified the methods to identify gaps in health research (i.e., primary [quantitative and qualitative survey, case report/case series, health impact assessment], secondary [evidence/knowledge synthesis, bibliometric study, meta research studies] and combined primary and secondary methods [evidence needs assessment, priority setting, global evidence-mapping methods, comparative effectiveness trials, academic crowd-sourcing]). Table 12 presents the specific methods to identify gaps in health research classified by primary, secondary and combined both methods.

Table 12. Identified key methods to identify gaps in health research

Primary methods	
Quantitative and qualitative survey [31]	"A scientific procedure for collecting information and making quantitative inferences about a predefined population. Surveys allow selection of a relatively large sample of people from a predetermined population, followed by collection of data from those individuals, and may be exploratory, descriptive, or explanatory." [134]
Case report/case series	"Case reports and case series or case study research are descriptive studies that are prepared for illustrating novel, unusual, or atypical features identified in patients in medical practice, and they potentially generate new research questions. They are empirical inquiries or investigations of a patient or a group of patients in a natural, real-world clinical setting" [140]

Health impact assessment	"A means of evidence-based policy making for improvement in health. It is a combination of methods whose aim is to assess the health consequences to a population of a policy, project, or programme that does not necessarily have health as its primary objective" [137, 138]
Secondary methods	
Evidence/knowledge synthesis	Efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored [49] Meta-analysis, systematic review [35-38], scoping review [39-41], rapid review, evidence mapping [29, 42, 43], mapping study [44-46], umbrella review [3], integrative review [47], mixed studies integrated review, critical interpretive synthesis [48], realist synthesis
Bibliometric study [50, 51]	The quantitative study of bibliographic material used to examine the knowledge structure and development of research fields based on analysis of related publications [52]
Meta research studies	"This is the study of research itself: its methods, reporting, reproducibility, evaluation, and incentives." [148]
Combined (primary and secondary methods)	
Evidence needs assessment/needs analysis [33, 34]	A systematic process for determining and addressing needs, or "gaps" between current conditions and desired conditions or "wants" by using various techniques including primary or secondary research methods (e.g., reviewing evidence, guidelines, and conducting interviews) [28]
Priority setting [31, 32, 53-63]	JLA PSP, Cochrane Priority Setting (*consists of four steps: the first two aim at gap identification and the last two at research prioritization*) JLA PSP methods were designed to allow clinicians, patients and caregivers to work together to identify and prioritize uncertainties about the effects of treatments that could be answered by research by gathering research questions, checking existing research evidence, interim prioritization and a final consensus meeting to reach agreement on the top 10 research priorities [53]
Global evidence-mapping methods [42, 64]	Maps available research and provides an overview of a broad range of research questions and identifies evidence gaps [42]
Comparative effectiveness trials	"Comparative effectiveness research is the generation and synthesis of evidence that compares the benefits and harms of alternative methods to

	prevent, diagnose, treat, and monitor a clinical condition or to improve the delivery of care. Evidence is generated through research that uses various study designs (e.g., observational, clinical trial) and synthesized through systematic reviews." [139]
Academic crowd-sourcing [4]	An emerging paradigm that is based on harnessing the power of the crowd to solve problems [4]

*including methods used in Health Technology Assessments

5.3.3. Combined findings of methods to display research gaps

Among the methods to display research gaps, the combined methods used to display research gaps include the following formats: table, box plot, bar graph/horizontal bar graph, scatter plot, funnel plot, pie chart, mind maps, tree map chart, word cloud, geographic map, dot plot, radial bar plot/polar histogram, schematic representation and bubble plot/chart. The list of formats identified were used for different purposes such as providing a list of research questions, research gaps and research priority, ranking quality of evidence; and scoring research gaps. The full list of the formats is presented in Table 13. Future research needs to determine the preferences of the formats among different key stakeholders including researchers, policy-makers and health practitioners.

Table 13. Identified methods to display research gaps

Format	Information on research gaps and priorities displayed
Table [19, 26, 31, 32, 40, 42, 57, 59-61, 64, 69, 71, 80-82]	List of clinical questions, gaps and research priorities Ranking quality of evidence Study designs to address research questions Scoring of each research gap List of research questions Prioritization of research questions Metric of ranking information
Box plot [31]	List of research questions Ranking of research questions
Bar graph/horizontal bar graph [31, 44, 75, 83-89]	List of research priorities Frequency of questions prioritized Number of studies and categories studied Frequency of research questions Quality of evidence metric
Scatter plot [90]	Numeric values of desired research and current research
Funnel plot [91]	Number of studies included in the review Effect sizes of studies on the x-axis
Pie chart [18, 85]	Proportional size of health problems being investigated by trials registered on a registry platform
Mind maps [92]	Diagram used to represent concepts, ideas or tasks linked to and arranged radially around a central key word or idea Primary branches represent the major ideas or themes around the central topic, and secondary branches tend to include more concrete illustrative examples
Tree map chart [44]	Number of clinical trials, population and income group Comparison study of clinical registry data versus global health research data from the Global Burden of Disease
Word cloud [93]	Frequency of words Frequency of words between two groups

Geographic map [8, 35]	Studies mapped around the world that uses colors on a pre-defined health outcome. Different shapes and sizes also used for additional information on a map.
Dot plot [94]	Number of studies Quality of evidence Different colors to show different study designs
Radial bar plot/polar histogram [44]	Proportion of trials in several countries with one color used per country among two groups over a period of time Comparison of proportion of trials in several countries with one color used per country among two groups over a period of time
Schematic representation [50]	Horizontal axis represents time, and vertical axis represents different documents
Bubble plot/chart [29, 95, 96]	Bubbles represent studies; size indicates the relative number of studies and color the study design Number of studies by intervention type and health status Compares three sets of values
* Examples available in **Appendix E**.	

5.3.4. Methodological guidance steps to identify research gaps

This next section presents the methodological steps to consider when identifying and displaying gaps in health research. It incorporates the findings previously presented from both the scoping review and qualitative study on defining gaps, identifying and displaying gaps. It also provides some key examples that authors can use to inform the process of gap identification. This guidance is particularly tailored to researchers, and to be adopted among other users, it needs to be first evaluated on usability and feasibility, then customized accordingly. We also provide some key questions (based on the methodological guidance) that researchers can ask when assessing gaps in health research for a specific topic. To demonstrate the use of the key questions, we applied this to a scientific article and illustrate the example in section 5.3.7.

5.3.5. A step-by-step guide to identifying research gaps

Table 14. Methodological guidance steps to identify research gaps

Step 1: Specify topic area and/or research question	This step involves clearly defining your research topic area and/research question in which you want to investigate research gaps
Step 2: Map evidence and identification of research gap(s)	This step involves mapping and clearly stating the existing research gap. This step is central and involves selecting a relevant and appropriate method to identify research gaps. *Specific examples of different methods to identify research gaps* ☐ **Primary methods** (quantitative and qualitative survey, case report/case series, health impact assessment), ☐ **Secondary methods** (evidence/knowledge synthesis, bibliometric study, meta research studies ☐ **Combined primary and secondary methods** (evidence needs assessment, priority setting, global evidence-mapping methods, comparative effectiveness trials, academic crowd-sourcing)
Step 3: Clearly describe the research gaps(s) identified	This is also an important step because it will provide a clear description of the research gap identified in step 2. *Specific examples of different terms that have been used to describe research gaps* ☐ **Research/evidence/knowledge gaps** - evidence is missing, inadequate or insufficient; additional research is needed, from policy-maker perspectives, to address the evidence gap in the available primary research ☐ **Synthesis/unidentified gaps** - little or no evidence from systematic reviews is available and could be a valuable resource to inform the evidence base in a particular area. Lack of up-to-date and conclusive systematic reviews at low risk of bias mapped to a clinical question ☐ **Treatment uncertainty** - lack of up-to-date, reliable systematic reviews of research evidence addressing the uncertainty about the effects of treatment, and/or up-to-date systematic reviews of research evidence show that uncertainty exists

	☐ **Evidence uncertainty** - evidence may be of lower quality or not directly applicable, not a very large body of evidence, not a large number of studies etc. ☐ **Practical knowledge gap (action–knowledge conflict gap)** - professional behavior or practices deviate from research findings or are not covered by research ☐ **Empirical gap (evaluation void gap)** - research findings need to be evaluated or empirically verified ☐ **Population gap** - research regarding a population that is not adequately represented or under-researched in the evidence base or prior research (e.g., sex, race/ethnicity, age) ☐ **Methodological gap (method and research design gap)** - a variation in research methods is required to generate new insights or to avoid distorted findings ☐ **Theoretical gap (theory–application void gap)** - theory should be applied to certain research issues to generate new insights; theory is lacking, so a gap exists
Step 4: Characterizing the research gaps	Use the PICOS framework to characterize research gaps related to interventions, screening tests, etc. The framework organizes research gaps as follows: ☐ **Population (P)**: information regarding the population that is not adequately represented in the evidence base (sex, race/ethnicity, age, etc.) ☐ **Intervention (I)**: information regarding the specific intervention that is inadequately included in the evidence base, the duration of the intervention, etc. ☐ **Comparison (C)**: lack of information regarding the comparison intervention or standard intervention ☐ **Outcomes (O)**: information regarding outcomes of interest, organized by type of outcome or timing of outcomes, to delineate where information is lacking ☐ **Setting (S):** information regarding the relevant settings for research gaps

<table>
<tr><td rowspan="2">Step 5: Presenting the research gaps</td><td colspan="3">Summarize the research gaps including clear descriptions, method of identification and further actions needed to address the research gaps. Additionally, the following data visualisations can be used to present the research gaps.</td></tr>
<tr><td>☐ Table
☐ Box plot
☐ Bar graph/horizontal bar graph
☐ Scatter plot
☐ Funnel plot</td><td>☐ Radial bar plot/polar histogram
☐ Schematic representation
☐ Bubble plot/chart
☐ Mind maps</td><td>☐ Word cloud
☐ Geographic map
☐ Dot plot
☐ Pie chart
☐ Tree map chart</td></tr>
</table>

5.3.6. Key questions to assess research gaps

A summary of the key questions to consider when reading the scientific article/information, with consideration of the research gaps, is as follows:

Box 2. Key questions to consider when assessing the nature of research gaps in a scientific article/information

- What is/are the key message(s) of the evidence provided in the text?
- What is/are the main source(s) of the evidence?
- What are the reported research gaps and research needed presented in the text?
- What is/are methods(s) used to display the research gaps?

5.3.7. Case study to illustrate how to apply the key questions to assess the nature of research gaps when reading a scientific article/information

This example applies the questions to a scientific article on COVID-19 article [153], showing how one can review the article and identify the research gaps. The questions are a tool to help better understand the body of evidence by highlighting the research gaps. Understanding the research gaps can improve public health and health practice efforts and action undertaken locally, nationally and globally and in this particular example, addressing a global pandemic. Subsequently, having clear research gaps can inform health practice, policy and future research.

The key questions presented in Box 1 were derived from synthesising the evidence of a scoping review and qualitative study of gaps in health research [104, 120]. The questions can be applied to guide the reader in the key messages from the evidence and research gaps reported by the author(s). The questions are derived from the methodological guidance in Table 14 to identify gaps in health research. Box 3 is a summary of the aforementioned key questions applied to the scientific article by Cascella et al. We show the application of the questions by using this article. This article was selected because it is a recent overview of the features, evaluation and treatment of COVID-19[153].

Box 3. Applying the key questions to consider when assessing the nature of research gaps in a scientific article/information

Article: Features, Evaluation and Treatment of Coronavirus (COVID-19)

Publication date: March 8, 2020 on StatPearls

1. What is/are the key messages of the evidence provided in the text?

The authors highlight that COVID-19 seems very contagious and has quickly spread globally. Additionally, in a meeting on January 30, 2020, per the International Health Regulations (IHR, 2005), the outbreak was declared by the WHO as a Public Health Emergency of International Concern (PHEIC).

The article stated that the first cases of CoVID-19 infection were linked to direct exposure at the Huanan Seafood Wholesale Market of Wuhan, China; the animal-to-human transmission was presumed to be the main mechanism. Nevertheless, subsequent cases were not associated with this exposure mechanism. Therefore, it was concluded that the virus could also be transmitted from humans to humans, and symptomatic people were the most frequent source of COVID-19 spread. The possibility of transmission before symptoms develop seems infrequent, although it cannot be excluded. Moreover, individuals who remain asymptomatic might transmit the virus. These data suggest that isolation is the best way to contain this epidemic.

The transmission is believed to occur through respiratory droplets from coughing and sneezing. Aerosol transmission is also possible in case of protracted exposure to elevated aerosol concentrations in closed spaces. Analysis of data related to the spread of SARS-CoV-2 in China seems to indicate that close contact between individuals is necessary for the spread. In fact, the spread is primarily limited to family members, healthcare professionals, and other close contacts.

The case studies of Li et al. published in the *New England Journal of Medicine* on January 29, 2020, encompass the first 425 cases recorded in Wuhan [154]. Data indicate that the patients' median age was 59 years (range 15 to 89 years). Thus, no clinical cases were reported in children below 15 years of age [153, 154].

The authors of the Chinese CDC report divided the clinical manifestations of the disease by their severity:

- Mild disease: non-pneumonia and mild pneumonia, which occurred in 81% of cases.
- Severe disease: dyspnea, respiratory frequency ≥ 30/min, blood oxygen saturation (SpO2) ≤ 93%, PaO2/FiO2 ratio (ratio between the blood pressure of oxygen [partial pressure of oxygen, PaO2] and the percentage of oxygen supplied [fraction of inspired oxygen, FiO2]) < 300, and/or lung infiltrates > 50% within 24 to 48 hr, which occurred in 14% of cases.
- Critical disease: respiratory failure, septic shock, and/or multiple organ dysfunction or failure, which occurred in 5% of cases[155].

At the time, the therapeutic strategies to deal with the infection are only supportive, and prevention aimed at reducing transmission in the community. Aggressive isolation measures in China led to a progressive reduction in cases.

The authors also provide a summary of what patients and families should receive instruction on based on previous research, some included and some not provided in their article:

- Avoid close contact with subjects with acute respiratory infections.
- Wash hands frequently, especially after contact with sick people or their environment. Avoid unprotected contact with farm or wild animals.
- People with symptoms of acute airway infection should keep their distance, cover coughs or sneezes with disposable tissues or clothes, and wash their hands.
- Immunocompromised individuals should avoid public exposure and public gatherings. If an immunocompromised individual must be in a closed space with multiple individuals present, such as a meeting in a small room, masks, gloves, and personal hygiene with antiseptic soap should be used by those in close contact with the individual. In addition, prior room cleaning with antiseptic agents should be performed before exposure. However, considering the danger to these individuals, exposure should be avoided unless a meeting, group event, etc. is a true emergency.
- Strict personal hygiene measures are necessary for the prevention and control of this infection.

Key messages without supporting evidence

Preliminary data suggest that the reported death rate ranges from 1% to 2% depending on the study and country. Most of the fatalities have occurred in patients over 50 years of age. Young children appear to be mildly infected but may be a vector for additional transmission.

Long-term complications among survivors of SARS-CoV-2 infection with clinically significant COVID-19 infection are not yet available. The global mortality rates for cases remain from 1% to 2%.

2. What is/are the main source(s) of the evidence?

Scientific article published on the "Statpearls" platform. The authors reported the aim of the platform is to collect information and scientific evidence and to provide an overview of the topic that will be continuously updated. Given the urgency of the COVID-19 pandemic, quick reporting is vital for information to be readily and quickly available for necessary actions as needed[153]. The sources of evidence provided by the authors vary from scientific articles to national and international reports.

3. What is/are the reported research gap(s) and research needed as reported in the evidence?

Mode of transmission: further studies are needed to understand the mechanisms of transmission, incubation times and clinical course as well as the duration of infectivity.

Pathophysiology: research is needed to determine the structural characteristics of SARS-COV-2 that underlie the pathogenetic mechanisms. As compared with SARS, for example, initial clinical data show less extra-respiratory involvement, although because of the lack of extensive data, we cannot draw definitive clinical information.

The article also reported that many uncertainties remain with regard to both the virus–host interaction and the evolution of the epidemic, with specific reference to the times when the epidemic will peak.

Research will be needed to determine the structural characteristics of SARS-COV-2 that underlie the pathogenetic mechanisms. As compared with SARS, for example, initial clinical data show less extra-respiratory involvement, although because of the lack of extensive data, we cannot draw definitive clinical information.

4. What is/are illustration(s) used to present the research gaps?

A figure showing a map of the COVID-19 outbreaks as of March 2, 2020 is available. The authors use the map to highlight the rapid evolution of the outbreak and that new cases may not be immediately represented visually on the map.

5.4. Discussion

When identifying and addressing research gaps in a topic area, the most important step is to first and foremost clearly define what is meant by a research gap in the text. I recommend adopting an existing definition that best describes the research gap or describes the nature of the research gaps. Describing the nature of the research gaps can involve different items, as follows:

1) State the scale of the research gap (i.e., specific or broad)
2) Clearly define the research gap
3) Specify the cause of research gap

When summarizing the methodological guidance, methods to identify research gaps were gathered based on the findings of the scoping review and qualitative study findings. I mainly found that methods to identify research gaps can be classified according to secondary, primary and combined secondary and primary methods. Also, methods can be classified according to identifying research gaps in a specific or broad area.

With secondary research, specifically evidence synthesis to identify research gaps in a specific area, mainly systematic reviews were reported, whereas in a broad area (i.e., to map the existing evidence base), scoping reviews were widely used and reported. For primary research, the focus of the methods used was a specific area.

Similar to methods to identify research gaps, displaying gaps can be classified according to secondary, primary and both secondary and primary findings. Additionally, depending on the items to highlight, tables and figures are the most common format to display gaps in health research. They are used to summarize the study results but have been found to also present data on the study characteristics and methodological quality. Other formats used to display data were bubble blots.

The methodological guidance aims to provide a basis of systematically identifying and reporting research gaps, to provide a clearer picture of the current status of the evidence base. The guidance also highlights the importance of clearly reporting research gaps, for subsequence clear identification of research gaps. The methodological guidance needs to be evaluated to determine its applicability and future implementation and adaption in health research and other fields.

Of note, the applied questions are not aimed to critically appraise the scientific article written by Cascella et al. but rather to show the application of key questions related to gaps in health research to consider when reviewing health research. Therefore, a working example of how the key questions can be applied is presented. These key questions aim to present a basis for systematically identifying research gaps and investigating their nature, including the evidence base. To improve their usability, further evaluation is needed to determine the applicability, relevance and feasibility by different audiences.

5.5. Strengths and limitations

The main strength of this methodological guidance is that it provides an overview of key items to consider while identifying and displaying gaps in health research. This guidance is based on

the findings of a comprehensive scoping review and qualitative study undertaken to inform the development of the methodological guidance. Both the scoping review and qualitative study findings demonstrated the difficulty with defining research gaps and characterising methods to identify and display research gaps. However, the main limitation is that the methodological guidance provided in this PhD is not definitive; further development and evaluation is needed, primarily with a wider audience of experts to provide feedback and advice.

Chapter 6: General Discussion

6.1. Summary of findings

In the first study, I found a comprehensive list of scientific articles that reported the definition, identification and/or prioritization, and display of research gaps. These articles related to defining research gaps and methods used to identify, prioritize and display research gaps varied significantly across different topic areas in health research. Within the studies that reported on terms and definitions used to describe research gaps, I identified a total of 12 different definitions based on the title, yet with some similarities on the description provided of each. Three similar cross-cutting themes were identified: definitions related to missing information, inadequate information and insufficient information. This analysis shows despite that the term "research gap" being commonly used in health research, its meaning can differ. Therefore, having a clear term and description supports the clarity in reporting health research and subsequently communication of what research gaps exist.

Additionally, I identified two main frameworks to identify gaps in health research. These frameworks were of key importance in better understanding ongoing efforts to systematically identify gaps in health research. The first is the framework for determining research gaps during systematic reviews[2] (Robinson et al.). It entails identifying and characterizing research gaps from systematic reviews. The framework characterizes gaps by using PICOS elements (also including setting) and identifying the reason(s) for why the gap exists. It also allows investigators to classify reasons for the existence of a research gap as 1) insufficient or imprecise information, 2) biased information, 3) inconsistency or unknown consistency, and 4) not the right information[2]. The other framework is for rigorously identifying research gaps in qualitative literature reviews[9] was designed to identify research gaps when conducting

literature reviews. The framework consists of the localization of research gaps, which is informed by the characterization, verification and presentation of verified research gaps.

The methods to identify research gaps were more clearly presented than the terms related to research gaps and the definitions used to describe the research gaps. The methods identified were grouped in three main categories: primary, secondary and combined primary and secondary research. The most frequently used method was secondary methods, followed by combined primary and secondary and finally primary methods.

Among the secondary methods, knowledge synthesis was commonly used. The definition of knowledge synthesis is an efficient scientific approach to identify and summarize evidence that allows for generalizability and consistency of the research findings to be assessed and data inconsistencies to be explored [49]. The purpose of knowledge synthesis is to summarize all pertinent studies on a specific question, improve the understanding of inconsistencies in diverse evidence and identify gaps in research evidence to define future research agendas [49].Moreover, within knowledge synthesis the scoping review is one of the only methods used to identify research gaps that explicitly included identification of research gaps as part of the purpose of the method.

I then classified the methods used to identify research gaps. The most frequent methods in the review were aimed at gap identification (including both identification and prioritization) and involved secondary research, including knowledge synthesis (80/116 articles, 69%), specifically systematic reviews and scoping reviews (58/80, 73%) (Figure 7). Overall, 24/116 (21%) articles described the use of both primary and secondary research and 12/116 (10%) only

primary research. I found 7 specific methods for identifying research gaps that are described along with the purpose of the method (Table 2).

A mix of primary and secondary research was most frequently used to determine research prioritization, namely, priority setting (e.g., JLA PSP methods). These methods involve the participation of patients, caregivers and healthcare and social-care professionals in identifying research questions, then prioritizing them by using a combination of primary and secondary research [19, 31, 32, 55, 56, 58, 59, 61, 62, 66, 70-79]. The main method for determining research prioritization with primary research was the Delphi survey, which is a practical and productive approach to obtaining opinions from a wide number of relevant experts for identifying potential priority topic areas for research [63, 66-68].

To display research gaps (Figure 7), half of the methods still used traditional ways to present findings (e.g., summary table and bar charts), and the other half used more advanced ways to display information (e.g., tree map charts, radial bar plots and bubble plots). The non-traditional methods used more colors and diagrams to present research gaps or priorities. For example, bubble plots use different shapes, sizes and colors to display information and can be used to present up to three different variables in one diagram. These characteristics could be further explored to determine the appropriate method to be used along with specified methods to identify research gaps and/or determine research priorities.

In the second study, I found a variety of terms and definitions to describe research gaps. The study also showed that the use of both primary and secondary methods (JLA method) to identify gaps was the most robust method for gap identification. The main reported advantage of this method is that it identified gaps (treatment uncertainties); involved different stakeholders

including patients and public involvement, to confirm and prioritize gaps. Whereas, the main disadvantage is that it is labor intensive (requires a team of different specialists) and expensive (administrative support, meeting rooms, catering among others) compared to secondary methods (evidence synthesis) or primary methods (survey). As for experiences with data visualisation, participants mainly expressed the importance of data visualisation in communicating research, no specific methods or formats to present gaps was expressed.

The qualitative study also showed that among methods to identify gaps, various methods can be adopted i.e. primary, secondary and both primary and secondary methods. Of all the methods to identify gaps, secondary methods is the most commonly used, specifically systematic reviews, which are considered the gold standard, they address a highly focused question related to the existing evidence and thus present difficulties for explicitly identifying research gaps in a general area [2, 8, 98]. Other secondary research methods reported were overviews of reviews, also known as umbrella reviews, scoping reviews and evidence mapping. Overviews of reviews focus on a much broader area, compiling evidence from multiple reviews into one accessible and usable document and highlighting other reviews within the specified topic area [100, 156]. Given the resource requirements of formal evidence reviews, topic prioritization is needed to best allocate resources to those areas deemed the most relevant for the health system. Regardless of the topic, the prioritization process is likely to be stakeholder-dependent. Priorities for evidence synthesis will vary depending on the mission of the health care system and the local needs of the health care stakeholders[152].

Hence using both primary and secondary methods is the most robust, since it involves the participation of patients, caregivers and health and social care professionals in identifying

research questions, then prioritizing them by using a combination of primary and secondary research [19, 31, 32, 55, 56, 58, 59, 61, 62, 66, 70-79].

In the last part of my PhD project, I focused on development of methodological guidance, aimed to systematically identify and report research gaps, to provide a clearer picture of the status of the evidence base. The guidance highlights the importance of clearly reporting research gaps, for subsequence clear identification of research gaps. The methodological guidance merged findings from the scoping review and the qualitative. I mainly found, when identifying and addressing research gaps in a topic area, the most important step is to first and foremost clearly define what is meant by a research gap in the text. I recommend adopting an existing definition that best describes the research gap or describes the nature of the research gaps. Describing the nature of the research gaps can involve different items including, state the scale of the research gap (i.e., specific or broad), clearly define the research gap and specify the cause of research gap.

Moreover, I found that methods to identify research gaps can be classified according to secondary, primary and combined secondary and primary methods. Also, methods can be classified according to identifying research gaps in a specific or broad area. The methodological guidance needs to be evaluated to determine its applicability and future implementation and adaption in health research and other fields.

Figure 7. Overview of specific methods to identify research gaps, prioritize research and display research gaps and/or research priorities

Specific methods to identify research gaps

Primary research methods

- ☐ Quantitative and qualitative survey
- ☐ Case report/case series
- ☐ Health impact assessment

Secondary research methods

- ☐ Evidence/knowledge synthesis
- ☐ Bibliometric study
- ☐ Meta research studies

Combined primary and secondary research methods

- ☐ Evidence needs assessment/needs analysis
- ☐ Priority setting
- ☐ Global evidence-mapping methods
- ☐ Comparative effectiveness trials
- ☐ Academic crowd-sourcing

Specific methods to identify research gaps

Primary research methods

- ☐ Delphi survey
- ☐ Quantitative survey

Secondary research methods

- ☐ Evidence/knowledge synthesis
  - Systematic review *
  - Scoping review

Combined primary and secondary research methods

- ☐ Priority setting Example: JLA PSP methods
- ☐ Global evidence-mapping method

Specific methods to identify research gaps

- ☐ Table
- ☐ Box plot
- ☐ Bar graph/horizontal bar graph
- ☐ Scatter plot
- ☐ Funnel plot
- ☐ Pie chart
- ☐ Mind maps
- ☐ Tree map chart
- ☐ Word cloud
- ☐ Geographic map
- ☐ Dot plot
- ☐ Radial bar plot/polar histogram
- ☐ Schematic representation
- ☐ Bubble plot/chart

6.2. Strengths and limitations

This section focuses on the overall strengths and limitations of the work presented in this thesis. The scoping review's overall strength was the comprehensive nature of the study and the overview of ongoing efforts for defining research gaps and methods to identify and display research gaps. It addressed current activities in reducing research waste by identifying research gaps, clarifying the critical information needs for stakeholders on methods to identify and display gaps in clinical care and public health research. The main limitation in relation to the scoping review is the lack of time to conduct an exhaustive study; for example, expanding the search strategy and having more reviewers to conduct full-text screening and data extraction would have improved the rigor of the study, particularly in identifying other studies that would be informative in shaping this topic area.

The qualitative study that followed the scoping review complimented the scoping review findings by exploring the experiences of key stakeholders to provide a clear additional perspective on what methods are available and where they are being used in practice. It was a major strength to have carried out the scoping review because it greatly informed the design and conduct of the qualitative study. The scoping review findings also helped provide a variety of terms related to research gaps in health research that improved understanding of research gaps and there characteristics One of the main limitations of the qualitative study was that it would have benefited from patient and public participation in the design and conduct of the study. This would have greatly improved the relevance and importance of the study to ensure that the in-depth interview guide was tailored to this population, thus improving the use of the research findings among patients and the public.

Both the scoping review and qualitative study provided the basis to develop the methodological guidance to identify research gaps. The main limitation of the methodological guidance is lack of additional time to be able to test the applicability of the methodological guidance developed. Future work to better understand the methodological guidance, including implementation and evaluation, is needed.

6.3. Implications

Recognizing the gaps of the evidence is important if we are to consider the existing body of evidence before undertaking future research and applying evidence to inform policy, health practice, and research and project funding. Researchers should clearly highlight the research gaps in their field and elaborate on the nature of the gaps by considering the methodological guidance and key questions developed from this PhD and described in Chapter 5. Clearly describing research gaps can also help researchers elaborate the justification of their study and other priority areas of the topic area.

This research project also provides a basis for further discussions highlighting what is unknown in the body of evidence. Moreover, systematically exploring the reasons for the unknown and its potential importance and relevance can inform health practice, policy and future research. Despite some research on research gap identification from systematic reviews and research prioritization, little research has been conducted to explore what constitutes a research gap and its impact on the body evidence. Yet, with the growth of technology and information widely available both scientific and non-scientific, identifying ways to sieve through the body of evidence can be of great value. Additionally, basic health interventions on what is known from the body of evidence and also what is unknown gives patients, health professionals and policy-makers the opportunity to make more informed decisions.

6.4. Conclusions

This PhD provides broad considerations for defining, identifying and displaying gaps in health research. It also helped to characterise the methods to identify and display research gaps and operationalized the definitions, based on what contributed to their existence. I studied how to improve the definitions of gaps in health research and develop methodological guidance on approaches to identify and display gaps in health research. Exploring definitions of research gaps, particularly based on the reason for the gap, will inform appropriate methodological approaches to identify the gaps and also better characterize the gaps, which can better inform research, practice and policy decisions guided by more concrete and clear characteristics of the gaps in research. Additionally, existing studies on this topic have been limited in the use of secondary studies to identify gaps in health research. The scoping review explored more broadly the different methods used to identify, prioritize, and display gaps in health research and this study aimed to explore key stakeholder's experiences in practice.

The comprehensive scoping review demonstrated the difficulty with defining research gaps and characterizing methods to identify, prioritize and display gaps in health research. The review also showed the various methods used to identify, prioritize and display gaps in health research and the current uncertainty around which of these is the most suitable method. Nevertheless, classifying methods to identify research gaps for methodological guidance is of key importance to help future researchers mainly think about potentially clearly defining research gaps and the methods that can be used to identify, display and report research gaps. The existing frameworks mainly presented in Chapter 1 are primarily focused on identifying research gaps from reviews mainly literature and systematic reviews, yet both primary and secondary methods can be

applied, depending on the researcher's needs, purpose of the research gaps identification, resources and capacity to invest in the identification of the research gaps.

Then I conducted a qualitative study to explore how research gaps are defined and identified in practice to obtain a more in-depth overview of what is applied in practice in comparison to what is captured in research. The perspectives identified in the qualitative study are not fully representative of experts and people using evidence to make health decisions for future research, health care policy or practice. Perspectives were limited to convenience sampling of participants identified in the scoping review and from evidence based scientific conferences, as well as MiRoR consortium expert contact recommendations. Nevertheless, participants represented a range of perspectives from patients, health care providers, policy makers and researchers, mainly based in Europe. Future work may consider expanding representation from other part of the world.

Lastly, I combined the findings of the scoping review and qualitative study to inform the development of methodological guidance for identifying and displaying research gaps. The methodological guidance provides a foundation for researchers, health care providers, patients, policy makers and funding bodies to consider when thinking about defining, identifying and displaying research gaps. I was not able to test the applicability and usability of the methodological guidance despite combining the items considered important to systematically identify and display research gaps. This project would benefit from future research on the application and evaluation of the methodological guidance. The guidance would be able to guide efforts on making healthcare decisions for individual patients and public health policies, and clinical guidelines should be informed by the best available research evidence while taking

into account missing, inadequate and insufficient evidence. Further work is needed to clarify terminology and methods to identify and display gaps in health research.

REFERENCES

1. Bennett C, Khangura S, Brehaut JC, Graham ID, Moher D, Potter BK, M. Grimshaw J: **Reporting Guidelines for Survey Research: An Analysis of Published Guidance and Reporting Practices**. *PLoS medicine* 2011, **8**(8):e1001069.
2. Robinson KA, Saldanha IJ, McKoy NA: **Development of a framework to identify research gaps from systematic reviews**. *Journal of clinical epidemiology* 2011, **64**(12):1325-1330.
3. Nasser M: **Setting priorities for conducting and updating systematic reviews**. University of Plymouth; 2018.
4. Rudan I, Campbell H, Marusic A, Sridhar D, Nair H, Adeloye D, Theodoratou E, Chan KY: **Assembling GHERG: Could "academic crowd-sourcing" address gaps in global health estimates?** *Journal of global health* 2015, **5**(1):010101.
5. Viergever RF, Olifson S, Ghaffar A, Terry RF: **A checklist for health research priority setting: nine common themes of good practice**. *Health Research Policy and Systems* 2010, **8**:36-36.
6. Chalmers I, Glasziou P: **Avoidable waste in the production and reporting of research evidence**. *The Lancet* 2009, **374**(9683):86-89.
7. Robinson KA, Saldanha IJ, McKoy NA: **Identification of research gaps from evidence-based guidelines: a pilot study in cystic fibrosis**. *Int J Technol Assess Health Care* 2011, **27**(3):247-252.
8. Carey T, Yon A, Beadles C, Wines R: **Prioritizing future research through examination of research gaps in systematic reviews**. *Prepared for the Patient-Centered Outcomes Research Institute* 2012.
9. Müller-Bloch C, Kranz J: **A Framework for Rigorously Identifying Research Gaps in Qualitative Literature Reviews**. In: *Proceedings/International Conference on Information Systems (icis): 2015*; 2015.
10. Trikalinos T, Dahabreh I, Lee J, Moorthy D: **Defining an optimal format for presenting research needs**. 2011.
11. Ballini L, Minozzi S, Negro A, Pirini G, Grilli R: **A method for addressing research gaps in HTA, developed whilst evaluating robotic-assisted surgery: a proposal**. *Health research policy and systems* 2010, **8**:27-27.
12. Guyatt GH, Oxman AD, Schünemann HJ, Tugwell P, Knottnerus A: **GRADE guidelines: A new series of articles in the Journal of Clinical Epidemiology**. *Journal of clinical epidemiology* 2011, **64**(4):380-382.
13. Li T, Vedula SS, Scherer R, Dickersin K: **What Comparative Effectiveness Research Is Needed? A Framework for Using Guidelines and Systematic Reviews to Identify Evidence Gaps and Research Priorities**. *Annals of Internal Medicine* 2012, **156**(5):367-377.
14. Jacobs RL: **Developing a research problem and purpose statement**. In.: San Francisco: Jossey-Bass; 2011: 125-141.
15. Brocke Jv, Simons A, Niehaves B, Niehaves B, Reimer K, Plattfaut R, Cleven A: **Reconstructing the Giant: On the Importance of Rigour in Documenting the Literature Search Process** 2009.
16. Owens DK, Lohr KN, Atkins D, Treadwell JR, Reston JT, Bass EB, Chang S, Helfand M: **AHRQ series paper 5: grading the strength of a body of evidence when comparing medical interventions—agency for healthcare research and quality and the effective health-care program**. *Journal of clinical epidemiology* 2010, **63**(5):513-523.
17. Ghersi D, Pang T: **En route to international clinical trial transparency**. *Lancet* 2008, **372**(9649):1531-1532.

18. Atal I, Zeitoun J-D, Névéol A, Ravaud P, Porcher R, Trinquart L: **Automatic classification of registered clinical trials towards the Global Burden of Diseases taxonomy of diseases and injuries**. *BMC Bioinformatics* 2016, **17**(1):392.
19. Al-Khatib SM, Gierisch JM, Crowley MJ, Coeytaux RR, Myers ER, Kendrick A, Sanders GD: **Future Research Prioritization: Implantable Cardioverter-Defibrillator Therapy in Older Patients**. *Journal of general internal medicine* 2015, **30**(12):1812-1820.
20. Young C, Horton R: **Putting clinical trials into context**. *The Lancet* 2005, **366**(9480):107-108.
21. Arksey H, O'Malley L: **Scoping studies: towards a methodological framework**. *International Journal of Social Research Methodology* 2005, **8**(1):19-32.
22. Peters MD, Godfrey CM, Khalil H, McInerney P, Parker D, Soares CB: **Guidance for conducting systematic scoping reviews**. *International journal of evidence-based healthcare* 2015, **13**(3):141-146.
23. McGowan J, Sampson M, Salzwedel DM, Cogo E, Foerster V, Lefebvre C: **PRESS Peer Review of Electronic Search Strategies: 2015 Guideline Statement**. *Journal of clinical epidemiology*, **75**:40-46.
24. Tricco AC, Lillie E, Zarin W, O'Brien K, Colquhoun H, Kastner M, Levac D, Ng C, Sharpe JP, Wilson K *et al*: **A scoping review on the conduct and reporting of scoping reviews**. *BMC Medical Research Methodology* 2016, **16**:15-15.
25. Tricco AC, Lillie E, Zarin W, et al.: **Prisma extension for scoping reviews (prisma-scr): Checklist and explanation**. *Annals of Internal Medicine* 2018, **169**(7):467-473.
26. Scott NA, Carmen M, Christa H, Jacques M: **Using Health Technology Assessment to Identify Research Gaps: An Unexploited Resource for Increasing the Value of Clinical Research**. *Healthcare Policy* 2008, **3**(3):109.
27. Hiten DP, Emmanuel I, Phillip MP, Stephen MS, Michael HJ, Ritu S, Eric BB, Mohamad EA: **A Systematic Review of Research Gaps in the Evaluation and Management of Localized Renal Masses**. *Urology* 2016, **98**:14.
28. Fulgham SM, Shaughnessy MF: **O & A with Ed Tech Leaders: Interview with Roger Kaufman**. *Educational Technology* 2008, **48**(5):49.
29. Birte S, Martina V, Ami B, Jennifer S, Marie G: **Evidence & Gap Maps: A tool for promoting evidence informed policy and strategic research agendas**. *Journal of clinical epidemiology* 2016, **79**:120.
30. Brearley SG, Stamataki Z, Addington-Hall J, Foster C, Hodges L, Jarrett N, Richardson A, Scott I, Sharpe M, Stark D *et al*: **The physical and practical problems experienced by cancer survivors: a rapid review and synthesis of the literature**. *Eur J Oncol Nurs* 2011, **15**(3):204-212.
31. Yu T, Li T, Lee KJ, Friedman DS, Dickersin K, Puhan MA: **Setting priorities for comparative effectiveness research on management of primary angle closure: a survey of Asia-Pacific clinicians**. *J Glaucoma* 2015, **24**(5):348-355.
32. van Middendorp JJ, Allison HC, Ahuja S, Bracher D, Dyson C, Fairbank J, Gall A, Glover A, Gray L, Masri WE *et al*: **Top ten research priorities for spinal cord injury: the methodology and results of a British priority setting partnership**. *Spinal Cord* 2016, **54**(5):341-346.
33. Kitson A, Straus SE: **The knowledge-to-action cycle: identifying the gaps**. *Canadian Medical Association Journal* 2010, **182**(2):E73-E77.
34. Thompson A, Brennan K, Cox A, Gee J, Harcourt D, Harris A, Harvie M, Holen I, Howell A, Nicholson R *et al*: **Evaluation of the current knowledge limitations in breast cancer research: a gap analysis**. *Breast Cancer Research* 2008, **10**(2):R26.

35. Wahl DA, Cooper C, Ebeling PR, Eggersdorfer M, Hilger J, Hoffmann K, Josse R, Kanis JA, Mithal A, Pierroz DD *et al*: **A global representation of vitamin D status in healthy populations**. *Arch Osteoporos* 2012, **7**:155-172.
36. Chang SM, Carey TS, Kato EU, Guise JM, Sanders GD: **Identifying research needs for improving health care**. *Ann Intern Med* 2012, **157**(6):439-445.
37. Mickenautsch S: **Research gaps identified during systematic reviews of clinical trials: glass-ionomer cements**. *BMC Oral Health* 2012, **12**(1):18.
38. Tunis SR, Turkelson C: **Using Health Technology Assessment to Identify Gaps in Evidence and Inform Study Design for Comparative Effectiveness Research**. *Journal of Clinical Oncology* 2012, **30**(34):4256-4261.
39. Levac D, Colquhoun H, O'Brien KK: **Scoping studies: advancing the methodology**. *Implementation Science* 2010, **5**(1):69.
40. Schwartz SR, Baral S: **Fertility-related research needs among women at the margins**. *Reprod Health Matters* 2015, **23**(45):30-46.
41. Tricco AC, Lillie E, Zarin W, O'Brien K, Colquhoun H, Kastner M, Levac D, Ng C, Sharpe JP, Wilson K *et al*: **A scoping review on the conduct and reporting of scoping reviews**. *BMC Med Res Methodol* 2016, **16**:15.
42. Clavisi O, Bragge P, Tavender E, Turner T, Gruen RL: **Effective stakeholder participation in setting research priorities using a Global Evidence Mapping approach**. *Journal of clinical epidemiology* 2013, **66**(5):496-502 e492.
43. Schmucker C, Motschall E, Antes G, Meerpohl JJ: **[Methods of evidence mapping. A systematic review]**. *Bundesgesundheitsblatt, Gesundheitsforschung, Gesundheitsschutz* 2013, **56**(10):1390.
44. Atal I, Trinquart L, Porcher R, Ravaud P: **Differential Globalization of Industry- and Non-Industry-Sponsored Clinical Trials**. *PLoS One* 2015, **10**(12):e0145122.
45. Ahmad N, Boutron I, Dechartres A, Durieux P, Ravaud P: **Geographical representativeness of published and ongoing randomized controlled trials. The example of: Tobacco consumption and HIV infection**. *PLoS One* 2011, **6**(2):e16878.
46. Ndounga Diakou LA, Ntoumi F, Ravaud P, Boutron I: **Published randomized trials performed in Sub-Saharan Africa focus on high-burden diseases but are frequently funded and led by high-income countries**. *Journal of clinical epidemiology* 2017, **82**(Supplement C):29-36.e26.
47. Marion LM, Fiona C, Susanne K, Renee S, Jenny M, Toni D: **Patient, family-centred care interventions within the adult ICU setting: An integrative review**. *Australian Critical Care* 2016, **29**(4):179.
48. Johnson M, Tod AM, Brummell S, Collins K: **Prognostic communication in cancer: A critical interpretive synthesis of the literature**. *Eur J Oncol Nurs* 2015, **19**(5):554-567.
49. Mulrow CD: **Systematic reviews: rationale for systematic reviews**. *BMJ* 1994, **309**(6954):597-599.
50. Kho ME, Brouwers MC: **The systematic review and bibliometric network analysis (SeBriNA) is a new method to contextualize evidence. Part 1: description**. *Journal of clinical epidemiology* 2012, **65**(9):1010-1015.
51. Mbuagbaw L, Kredo T, Welch V, Mursleen S, Ross S, Zani B, Motaze NV, Quinlan L: **Critical EPICOT items were absent in Cochrane human immunodeficiency virus systematic reviews: a bibliometric analysis**. *Journal of clinical epidemiology* 2016, **74**:66-72.
52. Merigó JM, Yang J-B: **A bibliometric analysis of operations research and management science**. *Omega* 2017, **73**(Supplement C):37-48.

53. Boney O, Bell M, Bell N, Conquest A, Cumbers M, Drake S, Galsworthy M, Gath J, Grocott MP, Harris E *et al*: **Identifying research priorities in anaesthesia and perioperative care: final report of the joint National Institute of Academic Anaesthesia/James Lind Alliance Research Priority Setting Partnership**. *BMJ Open* 2015, **5**(12):e010006.
54. Buckley BS, Grant AM, Glazener CM: **Case study: a patient-clinician collaboration that identified and prioritized evidence gaps and stimulated research development**. *Journal of clinical epidemiology* 2013, **66**(5):483-489.
55. Buckley BS, Grant AM, Tincello DG, Wagg A, Firkins L: **Reaching a consensus and ranking research priorities in urinary incontinence**. *Nurs Times* 2010, **106**(24):36-37.
56. Heazell AE, Whitworth MK, Whitcombe J, Glover SW, Bevan C, Brewin J, Calderwood C, Canter A, Jessop F, Johnson G *et al*: **Research priorities for stillbirth: process overview and results from UK Stillbirth Priority Setting Partnership**. *Ultrasound Obstet Gynecol* 2015, **46**(6):641-647.
57. Knight SR, Metcalfe L, O'Donoghue K, Ball ST, Beale A, Beale W, Hilton R, Hodkinson K, Lipkin GW, Loud F *et al*: **Defining Priorities for Future Research: Results of the UK Kidney Transplant Priority Setting Partnership**. *PLoS One* 2016, **11**(10):e0162136.
58. Welsh E, Stovold E, Karner C, Cates C: **Cochrane Airways Group reviews were prioritized for updating using a pragmatic approach**. *Journal of clinical epidemiology* 2015, **68**(3):341-346.
59. Rees SE, Chadha R, Donovan LE, Guitard AL, Koppula S, Laupacis A, Simpson S, Johnson JA: **Engaging Patients and Clinicians in Establishing Research Priorities for Gestational Diabetes Mellitus**. *Can J Diabetes* 2016.
60. Jennifer MG, Evan RM, Kristine MS, Douglas CM, Remy RC, Matthew JC, Ranee C, Amy SK, Gillian DS: **Prioritization of Patient-Centered Comparative Effectiveness Research for Osteoarthritis**. *Annals of Internal Medicine* 2014, **160**(12):836.
61. Ingram JR, Abbott R, Ghazavi M, Alexandroff AB, McPhee M, Burton T, Clarke T: **The Hidradenitis Suppurativa Priority Setting Partnership**. *Br J Dermatol* 2014, **171**(6):1422-1427.
62. Gadsby R, Snow R, Daly AC, Crowe S, Matyka K, Hall B, Petrie J: **Setting research priorities for Type 1 diabetes**. *Diabet Med* 2012, **29**(10):1321-1326.
63. Li T, Vedula SS, Scherer R, Dickersin K: **What comparative effectiveness research is needed? A framework for using guidelines and systematic reviews to identify evidence gaps and research priorities**. *Ann Intern Med* 2012, **156**(5):367-377.
64. Jaramillo A, Welch VA, Ueffing E, Gruen RL, Bragge P, Lyddiatt A, Tugwell P: **Prevention and self-management interventions are top priorities for osteoarthritis systematic reviews**. *Journal of clinical epidemiology* 2013, **66**(5):503-510 e504.
65. Katz DL, Williams AL, Girard C, Goodman J, Comerford B, Behrman A, Bracken MB: **The evidence base for complementary and alternative medicine: methods of Evidence Mapping with application to CAM**. *Alternative therapies in health and medicine* 2003, **9**(4):22-30.
66. Yoshida S: **Approaches, tools and methods used for setting priorities in health research in the 21(st) century**. *Journal of global health* 2016, **6**(1):010507.
67. Evelina C, Ludovic R, Amy C, Stephanie S, Xavier B: **Cochrane systematic reviews are useful to map research gaps for decreasing maternal mortality**. *Journal of clinical epidemiology* 2013, **66**(1):105.

68. Mitchell RB, Hussey HM, Setzen G, Jacobs IN, Nussenbaum B, Dawson C, Brown CA, 3rd, Brandt C, Deakins K, Hartnick C *et al*: **Clinical consensus statement: tracheostomy care**. *Otolaryngol Head Neck Surg* 2013, **148**(1):6-20.
69. Boney O, Bell M, Bell N, Conquest A, Cumbers M, Drake S, Galsworthy M, Gath J, Grocott MPW, Harris E *et al*: **Identifying research priorities in anaesthesia and perioperative care: final report of the joint National Institute of Academic Anaesthesia/James Lind Alliance Research Priority Setting Partnership**. *BMJ Open* 2015, **5**(12).
70. Wald HL, Leykum LK, Mattison ML, Vasilevskis EE, Meltzer DO: **A patient-centered research agenda for the care of the acutely ill older patient**. *J Hosp Med* 2015, **10**(5):318-327.
71. Lophatananon A, Tyndale-Biscoe S, Malcolm E, Rippon HJ, Holmes K, Firkins LA, Fenton M, Crowe S, Stewart-Brown S, Gnanapragasam VJ *et al*: **The James Lind Alliance approach to priority setting for prostate cancer research: an integrative methodology based on patient and clinician participation**. *BJU Int* 2011, **108**(7):1040-1043.
72. Pollock A, St George B, Fenton M, Firkins L: **Top 10 research priorities relating to life after stroke--consensus from stroke survivors, caregivers, and health professionals**. *Int J Stroke* 2014, **9**(3):313-320.
73. Meremikwu M, Udoh E, Nwagbara B, Effa E, Oringanje C, Edet B, Nwagbara E, Bello S, Eke F: **Priority setting for systematic review of health care interventions in Nigeria**. *Health Policy* 2011, **99**(3):244-249.
74. Brian SB, Adrian MG, Cathryn MAG: **Case study: A patient-clinician collaboration that identified and prioritized evidence gaps and stimulated research development**. *Journal of clinical epidemiology* 2011, **66**(5):483.
75. Mitnick CD, Rodriguez CA, Hatton ML, Brigden G, Cobelens F, Grobusch MP, Horsburgh R, Lange C, Lienhardt C, Oren E *et al*: **Programmatic Management of Drug-Resistant Tuberculosis: An Updated Research Agenda**. *PLoS One* 2016, **11**(5):e0155968.
76. Pollock A, St George B, Fenton M, Crowe S, Firkins L: **Development of a new model to engage patients and clinicians in setting research priorities**. *J Health Serv Res Policy* 2014, **19**(1):12-18.
77. van Furth EF, van der Meer A, Cowan K: **Top 10 research priorities for eating disorders**. *Lancet Psychiatry* 2016, **3**(8):706-707.
78. Chapman E, Reveiz L, Sangalang S, Manu C, Bonfill X, Munoz S, Abalos E: **A survey study identified global research priorities for decreasing maternal mortality**. *Journal of clinical epidemiology* 2014, **67**(3):314-324.
79. Gierisch JM, Myers ER, Schmit KM, Crowley MJ, McCrory DC, Chatterjee R, Coeytaux RR, Kendrick A, Sanders GD: **Prioritization of research addressing management strategies for ductal carcinoma in situ**. *Ann Intern Med* 2014, **160**(7):484-491.
80. Thompson A, Brennan K, Cox A, Gee J, Harcourt D, Harris A, Harvie M, Holen I, Howell A, Nicholson R *et al*: **Evaluation of the current knowledge limitations in breast cancer research: a gap analysis**. *Breast Cancer Res* 2008, **10**(2):R26.
81. Knight R, Small W, Pakula B, Thomson K, Shoveller J: **A scoping study to identify opportunities to advance the ethical implementation and scale-up of HIV treatment as prevention: priorities for empirical research**. *BMC Med Ethics* 2014, **15**:54.

82. Sun C, Dohrn J, Omoni G, Malata A, Klopper H, Larson E: **Clinical nursing and midwifery research: grey literature in African countries**. *Int Nurs Rev* 2016, **63**(1):104-110.
83. Wan YL, Beverley-Stevenson R, Carlisle D, Clarke S, Edmondson RJ, Glover S, Holland J, Hughes C, Kitchener HC, Kitson S *et al*: **Working together to shape the endometrial cancer research agenda: The top ten unanswered research questions**. *Gynecol Oncol* 2016, **143**(2):287-293.
84. DeFrank JT, Barclay C, Sheridan S, Brewer NT, Gilliam M, Moon AM, Rearick W, Ziemer C, Harris R: **The psychological harms of screening: the evidence we have versus the evidence we need**. *Journal of general internal medicine* 2015, **30**(2):242-248.
85. Viergever RF, Terry RF, Karam G: **Use of data from registered clinical trials to identify gaps in health research and development**. *Bull World Health Organ* 2013, **91**(6):416-425C.
86. Kumar MB, Wesche S, McGuire C: **Trends in Metis-related health research (1980-2009): identification of research gaps**. *Can J Public Health* 2012, **103**(1):23-28.
87. De Luca Canto G, Pacheco-Pereira C, Aydinoz S, Major PW, Flores-Mir C, Gozal D: **Biomarkers associated with obstructive sleep apnea and morbidities: a scoping review**. *Sleep Med* 2015, **16**(3):347-357.
88. Tavender EJ, Bosch M, Fiander M, Knott JC, Gruen RL, O'Connor D: **Implementation research in emergency medicine: a systematic scoping review**. *Emerg Med J* 2016, **33**(9):652-659.
89. Singh Ospina N, Rodriguez-Gutierrez R, Brito JP, Young WF, Jr., Montori VM: **Is the endocrine research pipeline broken? A systematic evaluation of the Endocrine Society clinical practice guidelines and trial registration**. *BMC Med* 2015, **13**:187.
90. Azeredo TB, Luiza VL, Oliveira MA, Emmerick IC, Bigdeli M: **Stakeholders' perspectives on access-to-medicines policy and research priorities in Latin America and the Caribbean: face-to-face and web-based interviews**. *Health Res Policy Syst* 2014, **12**:31.
91. Umscheid CA: **A Primer on Performing Systematic Reviews and Meta-analyses**. *Clin Infect Dis* 2013, **57**(5):725-734.
92. van den Eertwegh V, van Dulmen S, van Dalen J, Scherpbier AJ, van der Vleuten CP: **Learning in context: identifying gaps in research on the transfer of medical communication skills to the clinical workplace**. *Patient Educ Couns* 2013, **90**(2):184-192.
93. Andrea CT, Erin L, Wasifa Z, Kelly OB, Heather C, Monika K, Danielle L, Carmen N, Jane Pearson S, Katherine W *et al*: **A scoping review on the conduct and reporting of scoping reviews**. *BMC Medical Research Methodology* 2016, **16**:15.
94. Bhavisha V, Emma J, Iris G, Cova B, Elena S: **Availability of evidence on cataract in low/middle-income settings: a review of reviews using evidence gap maps approach**. *British Journal of Ophthalmology* 2016.
95. Miake-Lye IM, Hempel S, Shanman R, Shekelle PG: **What is an evidence map? A systematic review of published evidence maps and their definitions, methods, and products**. *Syst Rev* 2016, **5**:28.
96. Wang DD, Shams-White M, Bright OJ, Parrott JS, Chung M: **Creating a literature database of low-calorie sweeteners and health studies: evidence mapping**. *BMC Med Res Methodol* 2016, **16**:1.
97. Impellizzeri FM, Bizzini M: **SYSTEMATIC REVIEW AND META-ANALYSIS: A PRIMER**. *International Journal of Sports Physical Therapy* 2012, **7**(5):493-503.

98. Tricco AC, Zarin W, Ghassemi M, Nincic V, Lillie E, Page MJ, Shamseer L, Antony J, Rios P, Hwee J *et al*: **Same family, different species: methodological conduct and quality varies according to purpose for five types of knowledge synthesis**. *Journal of clinical epidemiology* 2018, **96**:133-142.
99. Nikolakopoulou A, Mavridis D, Furukawa TA, Cipriani A, Tricco AC, Straus SE, Siontis GCM, Egger M, Salanti G: **Living network meta-analysis compared with pairwise meta-analysis in comparative effectiveness research: empirical study**. *BMJ* 2018, **360**:k585.
100. Grant MJ, Booth A: **A typology of reviews: an analysis of 14 review types and associated methodologies**. *Health Info Libr J* 2009, **26**(2):91-108.
101. Althuis MD, Weed DL: **Evidence mapping: methodologic foundations and application to intervention and observational research on sugar-sweetened beverages and health outcomes**. *Am J Clin Nutr* 2013, **98**(3):755-768.
102. Hetrick SE, Parker AG, Callahan P, Purcell R: **Evidence mapping: illustrating an emerging methodology to improve evidence-based practice in youth mental health**. *J Eval Clin Pract* 2010, **16**(6):1025-1030.
103. Snilstveit B, Vojtkova M, Bhavsar A, Stevenson J, Gaarder M: **Evidence & Gap Maps: A tool for promoting evidence informed policy and strategic research agendas**. *Journal of clinical epidemiology* 2016.
104. Nyanchoka L, Tudur-Smith C, Thu VN, Iversen V, Tricco AC, Porcher R: **A scoping review describes methods used to identify, prioritize and display gaps in health research**. *Journal of clinical epidemiology* 2019, **109**:99-110.
105. Pham MT, Rajić A, Greig JD, Sargeant JM, Papadopoulos A, McEwen SA: **A scoping review of scoping reviews: advancing the approach and enhancing the consistency**. *Research Synthesis Methods* 2014, **5**(4):371-385.
106. Whittemore R, Chao A, Jang M, Minges KE, Park C: **Methods for knowledge synthesis: An overview**. *Heart & Lung: The Journal of Acute and Critical Care* 2014, **43**(5):453-461.
107. Kastner M, Antony J, Soobiah C, Straus SE, Tricco AC: **Conceptual recommendations for selecting the most appropriate knowledge synthesis method to answer research questions related to complex evidence**. *Journal of clinical epidemiology* 2016, **73**:43-49.
108. Manson H: **Systematic reviews are not enough: policymakers need a greater variety of synthesized evidence**. *Journal of clinical epidemiology* 2016, **73**:11-14.
109. Perrier L, Lightfoot D, Kealey MR, Straus SE, Tricco AC: **Knowledge synthesis research: a bibliometric analysis**. *Journal of clinical epidemiology* 2016, **73**:50-57.
110. Pluye P, Hong QN, Bush PL, Vedel I: **Opening-up the definition of systematic literature review: the plurality of worldviews, methodologies and methods for reviews and syntheses**. *Journal of clinical epidemiology* 2016, **73**:2-5.
111. Sales A: **Generating and using evidence: reflections from the perspective of implementation**. *Journal of clinical epidemiology* 2016, **73**:6-7.
112. Straus SE, Kastner M, Soobiah C, Antony J, Tricco AC: **Introduction: Engaging researchers on developing, using, and improving knowledge synthesis methods: a series of articles describing the results of a scoping review on emerging knowledge synthesis methods**. *Journal of clinical epidemiology* 2016, **73**:15-18.
113. Tricco AC, Antony J, Soobiah C, Kastner M, Cogo E, MacDonald H, D'Souza J, Hui W, Straus SE: **Knowledge synthesis methods for generating or refining theory: a scoping review reveals that little guidance is available**. *Journal of clinical epidemiology* 2016, **73**:36-42.

114. Wong G: **Knowledge synthesis approaches–spoilt for choice?** *Journal of clinical epidemiology* 2016, **73**:8-10.
115. Bouma GD, Ling R: **The research process**: Oxford University Press, USA; 2004.
116. Dowling M: **Approaches to reflexivity in qualitative research**. *Nurse researcher* 2006, **13**(3).
117. Palaganas EC, Sanchez MC, Molintas VP, Caricativo RD: **Reflexivity in qualitative research: A journey of learning**. *Qualitative Report* 2017, **22**(2).
118. Yin RK: **Case study research: Design and methods (applied social research methods)**. *London and Singapore: Sage* 2009.
119. Saunders B, Sim J, Kingstone T, Baker S, Waterfield J, Bartlam B, Burroughs H, Jinks C: **Saturation in qualitative research: exploring its conceptualization and operationalization**. *Quality & Quantity* 2017.
120. Nyanchoka L, Tudur-Smith C, Porcher R, Hren D: **Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study protocol**. *BMJ Open* 2019, **9**(8):e027926.
121. Shenton A: **Strategies for Ensuring Trustworthiness in Qualitative Research Projects**, vol. 22; 2004.
122. Hennink MM, Kaiser BN, Marconi VC: **Code Saturation Versus Meaning Saturation: How Many Interviews Are Enough?** *Qualitative Health Research* 2016, **27**(4):591-608.
123. Britten N: **Qualitative interviews in medical research**. *BMJ* 1995, **311**(6999):251-253.
124. Kvale S, Brinkmann S: **Interviews: Learning the craft of qualitative research interviewing**: Sage; 2009.
125. Gill P, Stewart K, Treasure E, Chadwick B: **Methods of data collection in qualitative research: interviews and focus groups**. *Br Dent J* 2008, **204**(6):291-295.
126. Braun V, Clarke V: **Using thematic analysis in psychology**. *Qualitative research in psychology* 2006, **3**(2):77-101.
127. Guba EG, and Yvonna S. Lincoln: **Naturalistic inquiry**; 1985.
128. Glonti K, Hren D: **Editors' perspectives on the peer-review process in biomedical journals: protocol for a qualitative study**. *BMJ Open* 2018, **8**(10):e020568.
129. Macnee CL, McCabe S: **Understanding nursing research: Using research in evidence-based practice**: Lippincott Williams & Wilkins; 2008.
130. Holloway I, Wheeler S: **Ensuring trustworthiness and quality**. *Holloway I, Wheeler S Research in nursing 2nd Ed Blackwell Publishing, India1996* 2002:250-263.
131. Anney VN: **Ensuring the quality of the findings of qualitative research: Looking at trustworthiness criteria**. *Journal of Emerging Trends in Educational Research and Policy Studies (JETERAPS)*, **5**(2):272-281.
132. Tobin GA, Begley CM: **Methodological rigour within a qualitative framework**. *J Adv Nurs* 2004, **48**(4):388-396.
133. Connelly LM: **Trustworthiness in qualitative research**. *Medsurg Nursing* 2016, **25**(6):435-437.
134. Safdar N, Abbo LM, Knobloch MJ, Seo SK: **Research Methods in Healthcare Epidemiology: Survey and Qualitative Research**. *Infect Control Hosp Epidemiol* 2016, **37**(11):1272-1277.
135. Boney O, Bell M, Bell N, Conquest A, Cumbers M, Drake S, Galsworthy M, Gath J, Grocott MPW, Harris E *et al*: **Identifying research priorities in anaesthesia and perioperative care: final report of the joint National Institute of Academic Anaesthesia/James Lind Alliance Research Priority Setting Partnership**. *BMJ Open* 2015, **5**(12):e010006.

136. Wright J, Williams R, Wilkinson JR: **Development and importance of health needs assessment**. *BMJ* 1998, **316**(7140):1310-1313.
137. Scott-Samuel A: **Assessing how public policy impacts on health**. *Healthlines* 1997, **47**:15-17.
138. Lock K: **Health impact assessment**. *BMJ (Clinical research ed)* 2000, **320**(7246):1395-1398.
139. Sox HC: **Defining comparative effectiveness research: the importance of getting it right**. *Med Care* 2010, **48**(6 Suppl):S7-8.
140. Sayre JW, Toklu HZ, Ye F, Mazza J, Yale S: **Case Reports, Case Series - From Clinical Practice to Evidence-Based Medicine in Graduate Medical Education**. *Cureus* 2017, **9**(8):e1546-e1546.
141. Timulak L: **Meta-analysis of qualitative studies: A tool for reviewing qualitative research findings in psychotherapy**. *Psychotherapy Research* 2009, **19**(4-5):591-600.
142. Haidich AB: **Meta-analysis in medical research**. *Hippokratia* 2010, **14**(Suppl 1):29-37.
143. Medicine Io, Sciencespolicy IoMDoH, Use CfEMTiC, Policy DoHS, Promotion DoH, Prevention D: **Assessing medical technologies**: Nomad Press; 1985.
144. Hunt H, Pollock A, Campbell P, Estcourt L, Brunton G: **An introduction to overviews of reviews: planning a relevant research question and objective for an overview**. *Systematic Reviews* 2018, **7**(1):39.
145. Aromataris E, Fernandez R, Godfrey CM, Holly C, Khalil H, Tungpunkom P: **Summarizing systematic reviews: methodological development, conduct and reporting of an umbrella review approach**. *International journal of evidence-based healthcare* 2015, **13**(3):132-140.
146. Khangura S, Konnyu K, Cushman R, Grimshaw J, Moher D: **Evidence summaries: the evolution of a rapid review approach**. *Systematic Reviews* 2012, **1**(1):10.
147. Tricco AC, Antony J, Zarin W, Strifler L, Ghassemi M, Ivory J, Perrier L, Hutton B, Moher D, Straus SE: **A scoping review of rapid review methods**. *BMC Medicine* 2015, **13**(1):224.
148. Ioannidis JPA: **Meta-research: Why research on research matters**. *PLOS Biology* 2018, **16**(3):e2005468.
149. Sandelowski M, Voils CI, Barroso J: **Defining and designing mixed research synthesis studies**. *Research in the schools: a nationally refereed journal sponsored by the Mid-South Educational Research Association and the University of Alabama* 2006, **13**(1):29.
150. Rycroft-Malone J, McCormack B, Hutchinson AM, DeCorby K, Bucknall TK, Kent B, Schultz A, Snelgrove-Clarke E, Stetler CB, Titler M *et al*: **Realist synthesis: illustrating the method for implementation research**. *Implementation Science* 2012, **7**(1):33.
151. Rudan I, Campbell H, Marušić A, Sridhar D, Nair H, Adeloye D, Theodoratou E, Chan KY: **Assembling GHERG: Could "academic crowd–sourcing" address gaps in global health estimates?** *Journal of global health* 2015, **5**(1).
152. Hempel S, Gore K, Belsher B: **Identifying Research Gaps and Prioritizing Psychological Health Evidence Synthesis Needs**. *Medical Care* 2019, **57**:S259-S264.
153. Cascella M, Rajnik M, Cuomo A, Dulebohn SC, Di Napoli R: **Features, Evaluation and Treatment Coronavirus (COVID-19)**. In: *StatPearls [Internet]*. edn.: StatPearls Publishing; 2020.

154. Li Q, Guan X, Wu P, Wang X, Zhou L, Tong Y, Ren R, Leung KSM, Lau EHY, Wong JY *et al*: **Early Transmission Dynamics in Wuhan, China, of Novel Coronavirus-Infected Pneumonia**. 2020.
155. Wu Z, McGoogan JM: **Characteristics of and Important Lessons From the Coronavirus Disease 2019 (COVID-19) Outbreak in China: Summary of a Report of 72314 Cases From the Chinese Center for Disease Control and Prevention**. *Jama* 2020.
156. Pollock M, Fernandes RM, Becker LA, Featherstone R, Hartling L: **What guidance is available for researchers conducting overviews of reviews of healthcare interventions? A scoping review and qualitative metasummary**. *Syst Rev* 2016, **5**(1):190.
157. **Translational Science and Evidence-Based Healthcare: A Clarification and Reconceptualization of How Knowledge Is Generated and Used in Healthcare**. *Nursing Research and Practice* 2012, **2012**.
158. Snilstveit BV, Martina; Bhavsar, Ami; Gaarder, Marie: **Evidence gap maps - a tool for promoting evidence-informed policy and prioritizing future research**. 2013.
159. Callahan P, Liu P, Purcell R, Parker AG, Hetrick SE: **Evidence map of prevention and treatment interventions for depression in young people**. *Depress Res Treat* 2012, **2012**:820735.
160. Gough D, Thomas J, Oliver S: **Clarifying differences between review designs and methods**. *Syst Rev* 2012, **1**:28.
161. Bragge P, Clavisi O, Turner T, Tavender E, Collie A, Gruen RL: **The Global Evidence Mapping Initiative: scoping research in broad topic areas**. *BMC Med Res Methodol* 2011, **11**:92.
162. Colquhoun HL, Levac D, O'Brien KK, Straus S, Tricco AC, Perrier L, Kastner M, Moher D: **Scoping reviews: time for clarity in definition, methods, and reporting**. *Journal of clinical epidemiology* 2014, **67**(12):1291-1294.

SUPPLEMENTARY INFORMATION

Methods for Identifying, Prioritizing and Displaying Gaps in Health Research: A Scoping Review

Appendix A: Definitions

Evidence Based Healthcare, a process that identifies policy or clinical questions and addresses these questions by generating knowledge and evidence to effectively and appropriately deliver health care in ways that are effective, feasible, and meaningful to specific populations, cultures, and settings[157].

Evidence Gap Map/Evidence Map, a visual tool for presenting the state of evidence in particular thematic areas, with the aim to provide easy access to the best available evidence and highlight knowledge gaps [158].

Evidence Mapping, systematic search of a broad field to identify gaps in knowledge and/or future research needs that presents results in a user-friendly format often a visual figure or graph, or a searchable database [95], for research planning, strategic research prioritization and evidence-informed policies [65, 101, 102, 159-161].

Health Research Gaps, a research question for which missing or insufficient information limits the ability to reach a conclusion [1].

Knowledge Synthesis, efficient scientific approach to identifying and summarizing evidence that allows generalizability and consistency of research findings to be assessed and data inconsistencies to be explored [49].

Research Need, defined as a health gap that limits the ability of decision-makers (policy-makers, patients, practitioners) from making decisions and therefore more research in this area is needed[8].

Research Priority Setting, research priority setting is not commonly defined in a consistent way, although it has been described as any interpersonal activity that leads to the selection of topics or key questions to further investigate [3].

Scoping Reviews, an exploratory study that systematically maps the literature on a topic, identifying key concepts, theories and sources of evidence. They examine the extent range and nature of research activity and also aim at identifying research gaps[162].

Appendix B: Search Strategy

Database: MEDLINE

Date: 12/02/2017

Total hits: 450

1	Biomedical Research/ or Evidence-Based Medicine/	122593
2	(medical research or health research or experimental medicine or investigational medicine or health-care* or healthcare* or public health or clinical health).tw.	192294
3	1 or 2	309869
4	Knowledge/ or Research	203787
5	(research gap* or evidence gap* or knowledge gap* or prioritization gap* research uncertainties) and (identifying or displaying or determine or recognize or show or demonstrate or translate). tw.	1503
6	4 or 5	20527
7	Humans/	16365952
8	(wom#n or men or male or female or child* or adult or infant*).mp.	11114912
9	7or 8	18655386
10	3 and 6 and 9	7332
11	limit 8 to (english language and humans and "reviews (best balance of sensitivity and specificity)")	450

Database: PUBMED

Date: 12/02/2017

Total hits: 291

#Search ((((research OR evidence OR knowledge OR prioritization) gap*)) AND (identifying OR displaying)) AND (health research OR clinical health OR public health)

Sort by: Relevance Filters: published in the last 10 years; Humans; English

Database: EMBASE

Date: 12/02/2017

Total hits: 28

1	medical research/	204099
2	public health/	202515

3	(medical research or health research or experimental medicine or investigational medicine or health-care* or healthcare* or public health or clinical health).tw.	20726
4	1 or 2 or 3	407627
5	Knowledge/ or Research	28322
6	(research gap* or evidence gap* or knowledge gap* or prioritization gap* research uncertainties) and (identifying or displaying or determine or recognize or show or demonstrate or translate). tw.	1961
7	5 or 6	30209
8	Humans/	11463056
9	(wom*n or men or male or female or child* or adult or infant*).tw.	4809642
10	6 or 7	14242346
11	4 and 7 and 10	378
12	limit 9 to (human and embase and "reviews (best balance of sensitivity and specificity)" and yr="2006 -Current")	28
13	limit 10 to english	28

Database: Cochrane Library

Date: 12/02/2017

Total hits: 33

D	Search	Hits
#1	MeSH descriptor: [Biomedical Research] explode all trees	9394
#2	(medical research or health research or experimental medicine or investigational medicine or health-care* or healthcare* or public health or clinical health):ti,ab,kw	97848
#3	MeSH descriptor: [Evidence-Based Medicine] explode all trees	2114
#4	#1 or #2 or#3	100143
#5	MeSH descriptor: [Knowledge] explode all trees	146
#6	MeSH descriptor: [Research] explode all trees	17475
#7	(research gap* or evidence gap* or knowledge gap* or prioritization gap* research uncertainties) and (identifying or displaying or determine or recognize or show or demonstrate or translate): ti,ab,kw	507
#8	#5 or #6 or #7	19788
#9	MeSH descriptor: [Humans] explode all trees	1443
#10	(wom*n or men or male or female or child* or adult or infant*): ti,ab,kw	6938

#11	#9 or #10	8380
#12	#3 and #8 and #11 Publication Year from 2007 to 2017	33

Database: Scopus

Date: 12/02/2017

Total hits: 327

(TITLE-ABS-KEY (research AND gap*) OR TITLE-ABS-KEY (evidence AND gap*) OR TITLE-ABS-KEY (knowledge AND gap*) AND TITLE-ABS-KEY (identifying) OR TITLE-ABS-KEY (displaying) AND TITLE-ABS-KEY (health AND research) AND (clinical AND health) AND (public AND health) AND (humans)) AND (LIMIT-TO (PUBYEAR, 2017) OR LIMIT-TO (PUBYEAR, 2016) OR LIMIT-TO (PUBYEAR, 2015) OR LIMIT-TO (PUBYEAR, 2014) OR LIMIT-TO (PUBYEAR, 2013) OR LIMIT-TO (PUBYEAR, 2012) OR LIMIT-TO (PUBYEAR, 2011) OR LIMIT-TO (PUBYEAR, 2010) OR LIMIT-TO (PUBYEAR, 2008) OR LIMIT-TO (PUBYEAR, 2007)) AND (LIMIT-TO (SUBJAREA, "MEDI ") OR LIMIT-TO (SUBJAREA, "NURS") OR LIMIT-TO (SUBJAREA, "HEAL ")) AND (LIMIT-TO (DOCTYPE, "ar") OR LIMIT-TO (DOCTYPE, "re")) AND (LIMIT-TO (LANGUAGE, "English"))

Database: Web of Science

Date: 12/02/2017

Total hits: 392

TOPIC: (research gap*) *OR* **TOPIC:** (evidence gap*) *OR* **TOPIC:** (knowledge gap*) *OR* **TOPIC:** (prioritization gap*) *AND* **TOPIC:** (identifying) *OR***TOPIC:** (displaying) *AND* **TOPIC:** (health research) OR TOPIC: (clinical health) OR TOPIC: (public health) *AND* **TOPIC:** (human*)

Refined by: WEB OF SCIENCE CATEGORIES: (PUBLIC ENVIRONMENTAL OCCUPATIONAL HEALTH OR HEALTH POLICY SERVICES) AND **RESEARCH AREAS:** (PUBLIC ENVIRONMENTAL OCCUPATIONAL HEALTH OR HEALTH CARE SCIENCES SERVICES) AND **LANGUAGES:** (ENGLISH) AND**PUBLICATION YEARS:** (2016 OR 2012 OR 2010 OR 2007 OR 2015 OR 2013 OR 2009 OR 2014 OR 2011 OR 2008) AND **WEB OF SCIENCE CATEGORIES:** (HEALTH POLICY SERVICES OR HEALTH CARE SCIENCES SERVICES) AND **WEB OF SCIENCE CATEGORIES:** (PUBLIC ENVIRONMENTAL OCCUPATIONAL HEALTH OR PRIMARY HEALTH CARE OR PEDIATRICS OR NURSING)

Indexes=SCI-EXPANDED, SSCI, A&HCI, ESCI Timespan=2007-2017

Database: PROSPERO register

Date: 12/02/2017

Total hits: 27

TITLE-ABSTR-KEY ((research gap* or evidence gap* or knowledge gap* or prioritization gap*) and (identifying or displaying or determine or recognize or show or demonstrate or translate or research uncertainties)) and (health research or clinical health or public health) and (human).

In total, after removing overlapping references, 1222 articles remained After Total N= 1222

Additional searches included - TRIP (735); Google Scholar (23); Google, Hand Searched and Expert consultation (157)

Total N =**1938**

Appendix C: Inclusion criteria for scoping review

	Inclusion
Study methods	All study designs that describe or apply methodology including quantitative and qualitative methods for identifying research gaps
Language	English only
Species	Human only
Time Frame	Published within the last 10 years
Main content	Describing in objective/methods sections the identification of research gaps or determining research prioritization

Appendix D: Data extraction form

This data extraction form was used to gather data from relevant full-text articles on methods of identifying and displaying gaps in health research.

DATA EXTRACTION

1. What type of article is this? (**drop-down list, select one**)
    - Peer reviewed article
    - Opinion/editorial
    - Protocol
    - Other
2. What is the main objective of the article related to health research gap identification and health research prioritization? (**drop-down list, select one that best describes the article**)
    - Identification of research gaps
    - Prioritization of research
    - Both
3. What is the main study methodology used in the article (**drop-down list, select one that best corresponds with the article**)
    - Primary research: Quantitative Research
    - Primary research: Qualitative Research
    - Primary research: Mixed Method (Qualitative and Quantitative)

- Secondary research: Literature Review
- Secondary research: Systematic Review
- Secondary research: Narrative Review
- Secondary research: Scoping Review
- Secondary research: Evidence Mapping
- Secondary and primary research: Review and Qualitative Research
- Secondary and primary research: Review and Quantitative Research
- Secondary and primary research: Review and Mixed Methods
- Other

4. Does the article have a clear definition of health research gap? (**No, Yes**)
    - No

      If no skip to question 6
    - Yes

      If yes, complete question 5
5. What is the gap definition as described in the article (**Free Text**)
6. Does the article describe a specific method to identify health research gaps? (**No, Yes different to study methodology, yes same to study methodology**)
    - No

      If no skip to question 8
    - Yes different to study methodology

      If yes and different to study methodology proceed to question 7 and provide a description of the method as provided in this article
    - Yes same to study methodology

      If yes and same to study methodology skip to question 8, unless gap identification method has additional description information beyond study methodology information on question 3, in this case proceed to question 7 and document additional information.
7. What is the specific method used to identify health research gaps in the article (**Free text**)
8. Does that article describe method to determine research (RP) prioritization in the article? (**No, Yes different to study methodology, Yes same to study methodology**)
    - No

      If no skip to question 10
    - Yes different to study methodology

      If yes and different to study methodology proceed to question 10 and provide a description of the method as provided in this article
    - Yes same to study methodology

If yes and same to study methodology skip to question 10, unless gap identification method has additional description information beyond study methodology information on question 3, in this case proceed to question 10 and provide additional information.

9. What is the specific method used to determine research prioritization (RP) (**Free text**)
10. Did the article provide any display of health research gaps? (**No, Yes**)
11. What was the method used to display health research gaps in the article? **(Free text)**
12. Was there any mention of patient involvement in the study? **(No, Yes)**
13. Was a specific framework used in the article on health research gap identification? **(No, Yes)**
14. Did the article provide information regarding the research gap based on any of the following? **(List below)**
    - Insufficient or imprecise information
    - Biased information
    - Inconsistency or unknown consistency
    - Incorrect information (wrong population or wrong outcome)

Appendix E : Specific methods to display gaps

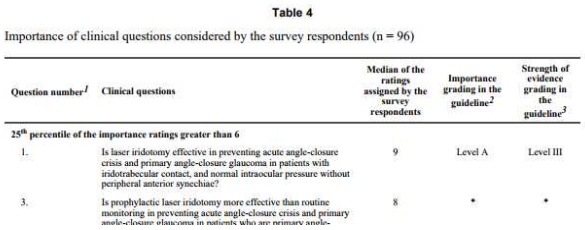
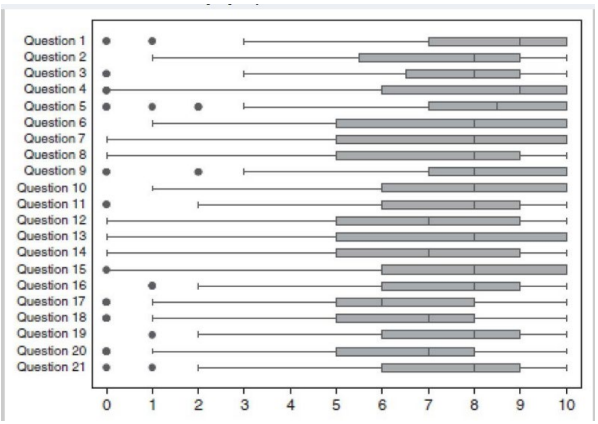
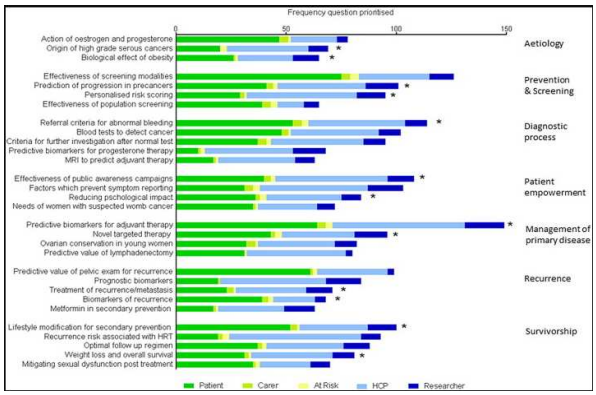
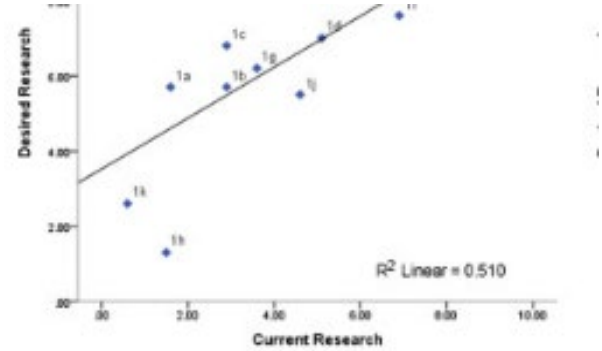
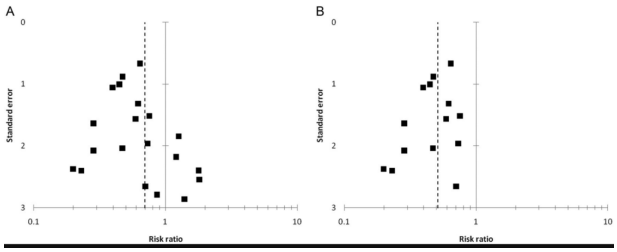
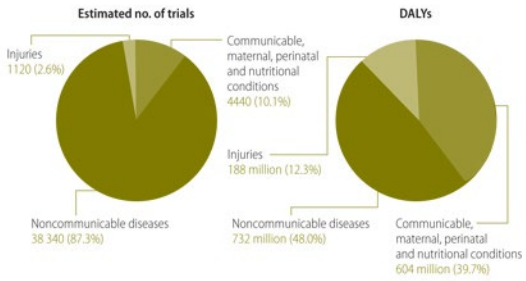
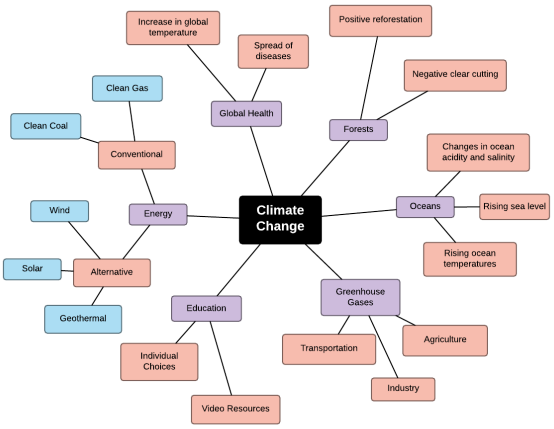
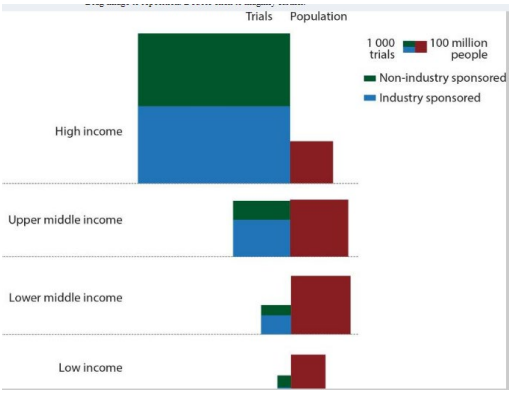
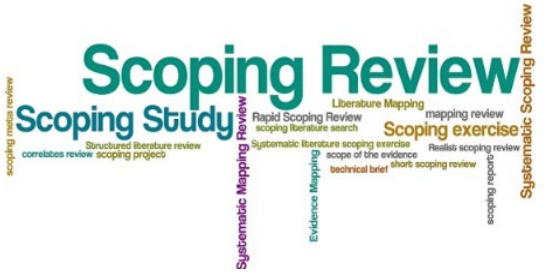
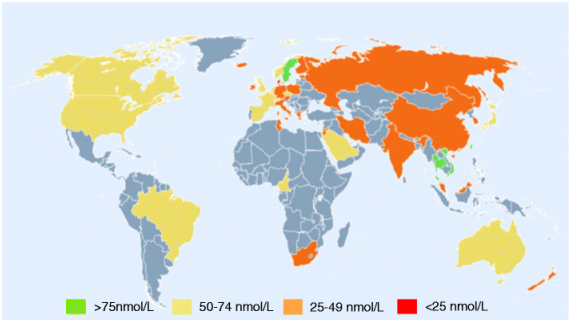
Format	Specific study method involved	Example
Table	Secondary research - Literature review - Health Technology Assessments - Systematic review* - Review of systematic reviews - Scoping Review Primary research - Quantitative survey - Delphi survey - Focus groups - Consensus meeting - Mixed methods	
Box plot	Primary research - Survey	

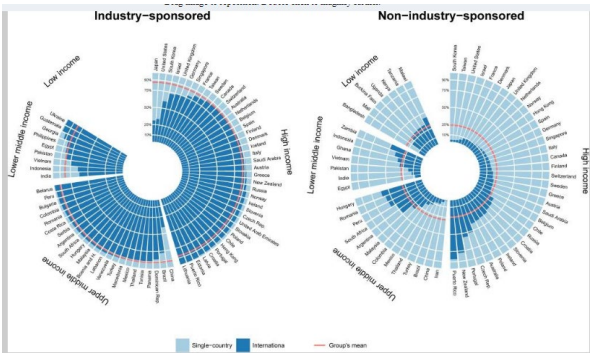
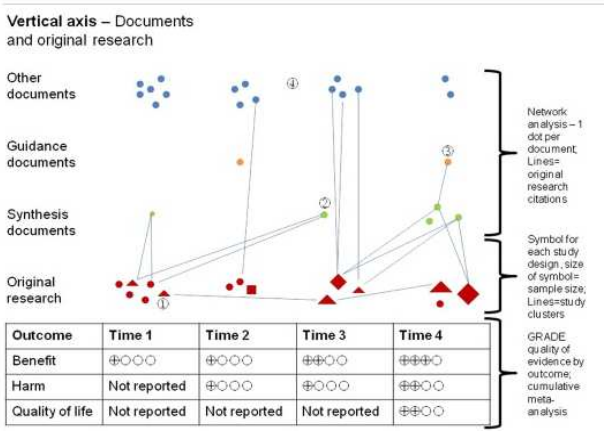
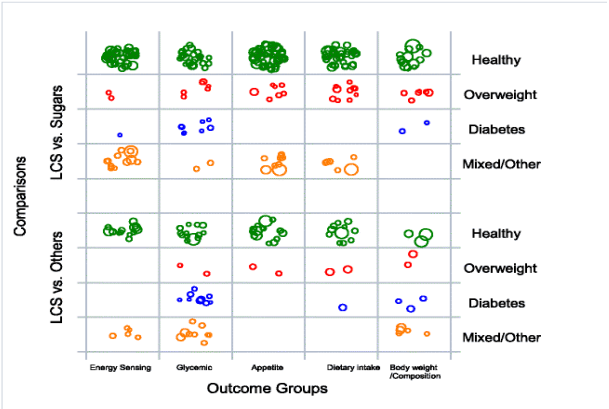
Table 4

Importance of clinical questions considered by the survey respondents (n = 96)

Question number[1]	Clinical questions	Median of the ratings assigned by the survey respondents	Importance grading in the guideline[2]	Strength of evidence grading in the guideline[3]
25th percentile of the importance ratings greater than 6				
1.	Is laser iridotomy effective in preventing acute angle-closure crisis and primary angle-closure glaucoma in patients with iridotrabecular contact, and normal intraocular pressure without peripheral anterior synechiae?	9	Level A	Level III
3.	Is prophylactic laser iridotomy more effective than routine monitoring in preventing acute angle-closure crisis and primary angle-closure glaucoma in patients who are primary angle-	8	*	*

Bar graph/horizontal bar graph	Secondary research - Systematic review - Mapping systematic reviews Primary research - Survey - Consensus meeting	
Scatter plot	Primary research - Qualitative interviews and quantitative survey	
Funnel plot	Secondary research - Methodological review	
Pie chart	Secondary research - Mapping study	Fig. 2. Health problems being investigated by trials registered in the International Clinical Trials Registry Platform (ICTRP), 2012 

Mind maps	Secondary research - Review of evidence	
Tree map chart	Secondary research - Mapping study	
Word cloud	Secondary research - Scoping review	
Geographic map	Secondary research - Systematic review	

Dot plot	Secondary research - Review of evidence	
Radial barplot	Secondary research - Mapping study	
Schematic representation	Secondary research - Systematic review - Bibliometric network analysis	
Bubble plot/chart	Secondary research - Scoping review - Evidence mapping	

*Including Health Technology Assessments

Key stakeholders' perspectives and experiences with defining, identifying and displaying gaps in health research: a qualitative study protocol

Appendix 1: Semi-structured interview guide

Date: Interviewer: Archival #:

In person: Teleconference: Start Time: End Time:

Background?

1. Tell me a little about your work, and what you do?
   What does it involve?
2. Experience with using evidence for decision-making in health choices, policymaking, prioritizing research or funding projects?
3. How did you go about making the decision when the evidence was missing, insufficient or inadequate?

Defining research gaps

4. How would you describe the term "research gaps" in your own words?
   Probe based on participant (Researcher, Policy maker, Funder, Health Professional or Public/Patient)
   - *(Research) Can you walk me through how you use evidence to inform future research/research topics?*
   - *(Policy Makers) Can you walk me through how you use research to influence policies?*
   - *(Funders) Can you walk me through how you use research to determine which project to fund?*
   - *(Health Professionals) Can you walk me through how you use research to inform your practice as a health provider?*
   - *(Public/Patients) Can you walk me through how you use research to inform your health decisions?*

     ✓ What are your thoughts on the importance of identifying research gaps?

     ✓ What are your thoughts on the causes of research gaps?

Experiences, knowledge and perceived needs with methods used to identify research gaps

5. Could you talk about your views/any experience you have in identifying research gaps?
   - *(Research) For example, if you need to apply for funding, how would you select the study?*
   - *(Policy Makers) For example, if you work in developing policies?*
   - *(Funders) or example, if you need to fund projects, how do you determine which ones to fund?*
   - *(Health professionals) For example, in making decisions between treatment choices in your practice where there is uncertainty?*

- *(Public/Patients) For example, when making health decisions where there is uncertainty?*

6. Could you tell me more specifically about the methods you used to identify research gaps?
   ✓ What are some of the strengths of the method(s)/practices you used?
   ✓ What are some of the challenges you experienced using the method(s) /practices?
7. Looking back on your experience using methods to identify research gaps, what is needed to improve the methods you used to identify research gaps?

Experiences, knowledge and perceived needs with methods used to display/present research gaps

8. Could you describe any experience you have in displaying/presenting research gaps?
9. Could you tell me more about the method(s) you used to display/present research gaps?
   ✓ What are some of the strengths of the method(s) you used for displaying research gaps?
   ✓ What are some of the challenges you experienced?
10. Please share any reflection on what you feel is needed to improve the methods you used to display/present research gaps?

General follow-up questions

11. Any additional thoughts you would like to share?

Appendix 2: Participant Information Sheet

Experiences with Methods for Identifying and Displaying Research Gaps

We invite you to take part in our research study. Before you decide whether to participate, you should understand why the research is being done and what it will involve. Please take your time to read the following information carefully and feel free to ask if you need more information or if there is anything that you do not understand. Please also feel free to discuss this with your friends, relatives and anyone else you wish.

What is the purpose of the study?

This study aims to explore the experiences of key stakeholders, including **the public, patients, researchers, clinicians, clinical guideline developers, public health professionals, policymakers and funders**, with methods for identifying and displaying research gaps, to inform **health choices, health practice, future research, policy or funding**. This study aims to help in better understanding the methods used to identify and display research gaps. The overall topic area on methods to identify and display gaps is still not well established, particularly because of no standard definition for the term "research gaps"; therefore a study to better understand the context, as well as the interactions of the factors such as alternative definitions, different audiences and methods used to identify gaps is important to improve our understanding of the audience's needs and the strengths and limitations of methods.

Why have I been chosen to take part?

You have been asked to take part because you are or have been involved in using research, producing research and/or communicating research. Your insight and experience with any methods you have used to identify and display research gaps will be highly appreciated to further guide this topic area.

Do I have to take part?

It is completely up to you whether or not you agree to take part in the study. If you do decide to take part, you will be asked to sign a consent form. If you decide to take part but then change your mind, you are free to do so at any time without giving a reason.

What will happen if I take part?

You will be asked to take part in an interview with a researcher, Linda Nyanchoka, about your experience with and your views of methods for identifying and display research gaps. The interviews will last approximately 20 to 40 minutes or as long as you would like to talk about your experience. With your permission, the interview will be audio-recorded. You can stop the interview at any time, and you do not have to answer a particular question if you don't want to.

Where will the interview take place?

The interview will take place in person at a specific location or over the phone. Participants in the UK have the option of an in-person or teleconference interview, and all other participants will have teleconference interviews at a date and time that is convenient for them.

Are there any risks in taking part?

We do not expect any risks or discomfort associated with this research study. However, if you feel uncomfortable, you can stop the interview at any time, without giving a reason.

Are there any benefits in taking part?

You will be helping develop our understanding of research gaps and methods for identifying and displaying research gaps.

Will my participation be kept confidential?

All the information you give us will be kept strictly confidential. The procedures for handling, processing, storing and destroying the data will comply with the Data Protection Act of 1998.

This means that only the researchers will see what you have said. The audio-recording of your interview will be identified by a code number only. These audio-recordings will be transcribed, and identifying details such as place names and people's names will be removed from the transcripts. We will use quotes from the interviews in the write-up of the study but will ensure that no one can be identified from these quotes.

At the end of the study, the research data, including consent forms, anonymised interview transcripts, field notes and your contact details, will be kept in locked filling cabinets and/or password-protected university computers for up to 10 years.

What will happen to the results of the study?

After the study has finished, the results will be written up as part of the PhD research thesis of Linda Nyanchoka and submitted for examination. The results will also be submitted for publication in an academic journal and presented at conferences.

If you would like to receive a copy of the findings, please let us know by using the contact information provided and we will happily provide you with one.

What will happen if I want to stop taking part?

If you decide at any point that you no longer wish to be part of the study, then you can withdraw without giving a reason. You can also ask for your data to be removed from the study and destroyed.

What if I am unhappy or if there is a problem?

If you are unhappy or if there is a problem, please feel free to let us know by contacting the lead researcher, Linda Nyanchoka, at the University of Liverpool (+33 75 34 29 417; L.Nyanchoka@liverpool.ac.uk). Linda will try to help or put you in touch with someone who can.

If you remain unhappy or have a complaint that you feel you cannot communicate to us, you should contact the Research Governance Officer at the University of Liverpool (0151 794 8290; ethics@liv.ac.uk). When contacting the Research Governance Officer, please provide the name or a description of the study (so that it can be identified), the researcher(s) involved, and the details of the complaint you wish to make.

Who is funding the research?

This research is funded by the European Union's Horizon 2020 research and innovation programme under the Marie Sklodowska-Curie grant (agreement no. 676207). If you want to find out more about the funding body, please contact https://ec.europa.eu/programmes/horizon2020/.

Who is doing this research?

The research and interviews will be conducted by Linda Nyanchoka, a Marie Curie Research Fellow at the University of Liverpool, UK.

How can I find out more?

You can get in touch with Linda Nyanchoka, who will be happy to answer any questions you might have:

Department of Biostatistics,Institute of Translational Medicine Block
F/WaterhouseBuidling,
University of Liverpool,
Liverpool
L69 3BX
Teleconference no.: +33 75 34 29 417
Email address: lnyanchoka@gmail.com

Thank you for taking the time to read this document.
This information sheet is for you to keep

Appendix 3: Participant consent form

Researcher: Linda Nyanchoka

Please initial box

1. I confirm that I have read and have understood the information sheet dated **[ ]** for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my rights being affected. ☐

3. I understand that, under the Data Protection Act 1998, I can at any time ask for access to the information I provide and I can also request the destruction of that information if I wish. ☐

4. I agree for the data I provide to be archived at The University of Liverpool. I understand that other authorised researchers will have access to this data only if they agree to preserve the confidentiality of the information as requested in this form. ☐

5. I agree to take part in the above study. ☐

_____	_____	
Participant name	Date	Signature
_____	_____	
Name of person taking consent	Date	Signature
_____	_____	
Researcher	Date	Signature

Principal Investigator
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Student Investigator
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Biostatistics Department
Block F Waterhouse Building
1-5 Brownlow Street
Liverpool L69 3GL

Tel: +33 75 34 29 417
Email: L.Nyanchoka@liverpool.ac.uk

The information you have submitted will be published as a report; please indicate whether you would like to receive a copy. ☐

I understand that confidentiality and anonymity will be maintained and it will not be possible to identify me in any publications ☐

I agree for the data collected from me to be used in future research and understand that any such use of identifiable data would be reviewed and approved by a research ethics committee. ☐

I understand and agree that my participation will be audio recorded and I am aware of and consent to your use of these recordings for the following purposes: meeting research aims and goals in exploring methods used to identify and display research gaps. ☐

I understand that the information collected about me will be used to support other research in the future, and may be shared anonymously with other researchers. ☐

I would like my name used and I understand and agree that what I have said or written as part of this study will be used in reports, publications and other research outputs so that anything I have contributed to this project can be recognised. ☐

I understand that my responses will be kept strictly confidential. I give permission for members of the research team to have access to my anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in the report or reports that result from the research. ☐

I understand and agree that once I submit my data it will become anonymised and I will therefore no longer be able to withdraw my data. ☐

I understand that the fully anonymised data will be held securely at the University of Liverpool and I can request access to the data collected, and/or request that the data is destroyed at any time until the data is submitted for publication. ☐

I understand that other authorised researchers may use my words in publications, reports, webpages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form. ☐

Appendix 4: Participant Teleconference Consent Form

Teleconference: Oral Consent Example Script:

Hello, I am Linda Nyanchoka, a PhD student from the University of Liverpool. I will be talking to you about my research project on defining research gaps and on methods to identify and display research gaps in health. Additional information is on the information sheet you have received.

Are you still interested in taking part in the project? *[Await confirmation]*. Now I'd like to confirm some of the details of the project to make sure you are clear about what's involved for you:

- We do not expect any risks or discomfort associated in this research study. However, if you feel uncomfortable, you can stop the interview at any time, without giving a reason.
- You do not have to say yes to take part; you can ask me any questions you want before or during the interview; you can also withdraw at any stage without giving a reason and without any negative consequences.
- You do not have to answer any questions that you do not wish to.
- You are aware that a University of Liverpool Research Ethics committee has approved this research project; for further information email me at L.Nyanchoka@liverpool.ac.uk
- I may use brief quotes of what you say during the interview in the write-up of this study, but they will remain anonymous.
- I will safely store your data electronically in encrypted, secure files. All identifiable data will be destroyed at the end of the study.
- I will audio-record you unless you say that I can't.
- Are you still willing to take part?

  Do you give your permission for me to re-contact you to clarify information?

[*Await confirmation]* So if you're happy with all of that, and have no more questions, let's start.

Researcher: Linda Nyanchoka

Participant:

Date:

Time: