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Qualitative Evidence Synthesis to inform health policy

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Editorial

Dear readers,

This year the Health Institute of São Paulo is celebrating its 50th anniversary, and we are very pleased to offer this special issue of the Health Institute Bulletin - BIS, dedicated to discussing the importance of qualitative evidence to shed light on decision-making in health policy.

Across the world there has been a growing interest in qualitative evidence in view of the recognition that decision-making processes should take into account factors related to social, cultural, and axiological context, particularly for the implementation of public policies, in order to improve their effectiveness. In this context, Qualitative Evidence Syntheses (QES) are at the forefront of efforts to transparently systematise and translate scientific knowledge available in primary qualitative studies, so that it can feed into decision-making in an accessible and timely manner.

The first two papers of this issue of BIS were commissioned to present qualitative evidence syntheses and their potential applications in health policy. The other papers were selected through both public call and peer-review, and they provide some examples of how Brazilian researchers are using qualitative evidence to produce knowledge synthesis/translation studies.

In the introductory paper on **qualitative evidence syntheses**, Sousa, Wainwright and Soares present elements of primary qualitative research, provide information on what QES are, and include a short guide on where to find methodological guidance for the conduct of different aspects of the review process, as reviews are increasingly in great demand in health policy making.

Barreto and Lewin address the importance of considering **qualitative evidence** to support actions for policies. The authors present case studies to show how qualitative evidence can inform policy development for key health systems questions. They also discuss what needs to be done in Brazil and the Latin American region to strengthen the use of qualitative evidence in decision making.

Scoping review is a type of evidence synthesis that can map all kind of evidence, and its results are usually expressed in a narrative format, as well as in a visual format such as graphics and figures. With the development of a scoping review of methodological nature, Cordeiro and Soares analyse the use of PRISMA extension for scoping reviews, recommended for the reporting of theses reviews.

Schveitzer and colleagues present the results of a **qualitative systematic review**, which evaluated the application and implementation of clinical management by *kanban* (a card to signal workflow, systems, and processes), in emergency hospital services.

Humanising practices in Primary Health Care is the focus of the **qualitative systematic review** carried out by Pastana and colleagues. Results are provided as summaries, which address perceptions of humanising practices, and barriers and facilitators to their implementation.

An **evidence brief for policy**, carried out by Tesser and colleagues, presents three options to inform health care policies for adolescents with sickle cell disease. The authors also take into consideration barriers to the implementation of these options, concerns about equity when implementing them, and contributions of deliberative dialogue to this synthesis results.

Silva and colleagues address the diabetic foot disease, a multifactorial syndrome that may affect people with diabetes mellitus. In an **evidence brief for policy**, they discuss four options for policies identified in scientific literature, including considerations on their implementation.

Congenital syphilis in Primary Health Care is the focus of an **evidence brief for policy**, carried out by Dias and colleagues as a dissertation in a professional master's degree programme. They discuss four options for policies, focusing on both equity and implementation of these options.

A **rapid qualitative evidence synthesis**, carry out by Tebet and colleagues, approaches men's perceptions of treatment of syphilis. Difficulty in treating sexual partners of women with syphilis are well known. The authors present the results in four categories of analysis that should contribute to a better understanding of the elements of this phenomenon.

Another **rapid qualitative evidence synthesis**, carried out by Bachega and colleagues, addresses the motivations, thoughts, and feelings associated with suicidal ideation among Brazilian children and adolescents. Suicidal ideation as a response to psychological pain was reported in all the studies analysed. However, the authors draw attention to the need for more primary research on this relevant issue of public health.

Farinasso and colleagues report in a **rapid review** the results regarding the main barriers to access or use biologic medicines in the context of users, providers, management and health system. This study informed discussions of the Working Group for the formulation of the National Policy for Biologic Medicines within the Brazilian Unified Health System (SUS), coordinated by the Department of Pharmaceutical Care of the Ministry of Health of Brazil.

Hoarders are known for accumulating excessive amounts of items or domestic animals in a disorderly and disorganized way. To address the potential risk of vector-borne and other zoonotic diseases, Henriques and colleagues conducted an **integrative review**; the findings refer to the health of hoarders themselves, their families and community.

We wish you a good read of this BIS thematic issue; this is the first initiative in Brazil to discuss and present qualitative evidence syntheses as a valuable contribution to inform decision-making on public health issues and problems.

This special edition of BIS was launched during the 'Using qualitative evidence to inform decisions in the Sustainable Development Goals (SDG) era: new frontiers and innovations – QESymposium', in Brasilia from 9-11 October 2019; it is the result of collaboration among different groups and institutions – Health Institute of São Paulo, Oswaldo Cruz Foundation (Fiocruz) - Brasília, Norwegian Institute of Public Health, Durham University, and School of Nursing of the University of São Paulo, among others –, for the promotion of Evidence-Informed Policy-making, and dissemination of knowledge translation products.

Invited Editors

Tereza Setsuko Toma (Health Institute of São Paulo); Jorge Barreto and Sharmila Sousa (Fiocruz Brasília); Simon Lewin and Claire Glenton (Norwegian Institute of Public Health); Megan Wainwright (Durham University); and Cassia Baldini Soares (School of Nursing of the University of São Paulo).

Qualitative Evidence Synthesis: an introductory guide

Maria Sharmila A. de Sousa[I], Megan Wainwright[II], Cassia Baldini Soares[III]

Abstract

Qualitative evidence syntheses (QES) are increasingly used in the health area and new methods are proliferating. The increase in demand for QES is driven in large part by a recognition that evidence on effectiveness is not enough to improve health, we also need evidence on stakeholders' perceptions, acceptability, feasibility, implementation factors, and equity – topics well suited for exploration through qualitative research methods. This introductory paper acts as an overview, serving also as a roadmap to QES methodologies. First, we give an overview of what primary qualitative research is – its paradigm, the kinds of questions it asks, and the kinds of methods and approaches it uses to answer them. Secondly, we provide some background on what a QES is, what kinds of questions it helps answer in health policy and decision-making contexts, and some examples of how it is increasingly used in guideline development and health technology assessment processes. Lastly, we provide readers with a roadmap of where to find methodological guidance on carrying-out different aspects of the review process including formulating a question, searching, choosing a synthesis method, assessing confidence in primary study results and review findings, and reporting a QES.

Keywords: Qualitative Evidence; Qualitative Evidence Synthesis; Systematic Reviews of Qualitative Studies, Qualitative Methodologies.

Apresentação

Health care is undoubtedly complex and historical, that is, dependent on relationships across social contexts. It is therefore essential to consider the evidence about experiences, attitudes, preferences, values and expectations of living and surviving with a health condition from the perspectives of patients, families, carers, legal representatives and health professionals. It is also essential to consider the experiences and/or expectations about the use of a specific health technology, as well as the evidence on the relationship between health needs and broader social dynamics. Such information and analyses can be particularly useful in understanding barriers and facilitators to the provision or implementation of new technologies in health services.

These are the types of evidence that emanate from qualitative research. However, it may not always be possible to conduct (primary) qualitative research due to constraints of time, money or other resources.[1] When this is the case, another option for ensuring that perspectives of key social actors informs practice and policies is to gather existing primary studies on the topic and integrate them into a qualitative evidence synthesis (QES).

The synthesis of findings from primary qualitative studies has become increasingly relevant as a tool to inform health policies and

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decision-making due to its potential to provide in-depth analysis of findings, as well as amplifying the understanding of phenomena of interest. As with other types of evidence, QES depends fundamentally on empirical studies conducted transparently and rigorously, as these will constitute the data to be reviewed and synthesised.

The growing academic recognition of qualitative research in health has culminated in a considerable body of academic health journals specialised in qualitative research. Although the term "qualitative research" was introduced in the Medical Subject Headings (MeSH) only in 2003, a quick search of the term in mid-2019 in the PubMed database identified nearly sixty thousand publications. In Scopus, a multidisciplinary database that comprises human and social sciences, this number exceeds one hundred thousand. It is understandable that, as this evidence database grows, new methods are needed to revise and synthesize such knowledge.

As such, QES (also known as systematic reviews of qualitative research or systematic reviews of qualitative evidence) is increasingly being produced and guidance for conducting it to answer complex questions in the field of health are being published. In this introductory paper, we give an overview of qualitative research and QES and provide a roadmap of specific methods, tools and approaches to guide and support those who wish to conduct a QES.

Qualitative Research Methodologies

Understanding qualitative research methodologies is the first challenge for those who want to produce or synthesize qualitative evidence. Qualitative research aims to analyse individuals' and/or social groups' experiences and interpretations of a phenomenon through systematic collection, organisation and analysis of speech or observational textual data (including textual data from blogs, vlogs, and websites of associations of patients and users of healthcare systems – both public and private – and other types of real-world data that can be analysed).[2] By using data collection methods that involve interaction amongst researchers and participants, qualitative research provides deep understanding of the social world under study.[3]

Qualitative research has an intersubjective nature and aims to produce in-depth understanding of the reality being studied.[4] Its objectives differ from those of quantitative research. As such, evidence from quantitative and qualitative studies informs the field of health in diverse ways, and contradictory findings can surface. These have great potential for responding to health needs. Qualitative research aims to identify, illustrate, describe and explain behaviours, preferences and expectations of patients, their families, caregivers, legal representatives, and health professionals whose responsibility it is to provide healthcare services – key in-depth information that quantitative evidence fails to provide.[5]

Qualitative research makes use of various methodologies, which come from different theoretical-methodological frameworks developed by the human and social sciences. These have been appropriated by the life sciences, notably due to the need to respond to intricate problems, and to understand issues of historical, dynamic, relational or fortuitous interest.[6] Experimental control or fragmentation into measurable variables would obscure or misrepresent the essence of this set of health-related issues. Understanding social and human relations is fundamental for developing and implementing health care whether in the individual or collective dimension, in the form of clinical interventions, in the form of collective programs and policies, or for improving conditions of work and life that determine health and disease gradients.[7]

We can see examples in the literature of qualitative studies grounded in positivist assumptions derived from the natural sciences – an approach that is considered inadequate for understanding processes underlined by micro and macro-social contexts. We also see examples of qualitative studies with no theoretical foundation that present only a simplistic description of empirical data.[8] Therefore, even if only briefly, it is important here to highlight the most commonly used qualitative methodologies in health research and clarify the theoretical-philosophical and theoretical-methodological foundations that form the basis of their respective rigour. Understanding such foundations is a condition for evaluating the methodological quality of a qualitative (empirical) study and, consequently, the degree of confidence one can have in the results of these primary studies, as well as in the findings of QES. We detail the steps for assessing the confidence in review findings and for critically evaluating primary studies in the overview of QES methods discussed later. There, we draw particular attention to the differences between results of primary qualitative studies and findings from qualitative evidence syntheses.

The first foundation of a qualitative study is its theoretically-oriented nature – that is that they are grounded in a theoretical frame appropriate to the analysis of their results. This is why we choose the expression methodology rather than methods, which more adequately expresses the inseparability between the theoretical framework of a research study and the set of methodological procedures coherent with such a framework.[9] These theoretical-methodological frameworks derive from scientific paradigms that, in turn, are shaped by ontological (nature of reality), epistemological (nature of knowledge) and methodological (procedures and methods to understand reality) dimensions. There are numerous publications that shed light on the dimensions of science paradigms. The classic by Guba[10], the book by Minayo[11] and by Denzin and Lincoln[12] widely recognised in health are just a few examples of research methodology texts that can help the reader understand such issues. These paradigms are constantly developing and transforming. The interpretative and critical traditions however are well established, and have had considerable influence on the field of health research. In recent history, the postmodern paradigm[8] for describing empirical observations has also gained recognition.

In the interpretative paradigm, **phenomenology** and its variations represent a significant part of research in health. These studies aim to understand the truth from the meanings that specific individuals give to phenomena.[13] Sodelli[14], for example, applied reflexive collective interviews from the perspective of existential phenomenology to analyse the meaning of drug prevention work for a group of third year pedagogy students.

Ethnographic methodology has been widely used in the health arena, especially in studying “health and disease of the working classes, or specific population groups; popular healing knowledge and practices; knowledge and practices of official medicine; madness and deviation; sexuality; medicalisation; health services; therapeutic choices”.[15] Ethnography originated in anthropology to achieve an in-depth understanding of culture in human societies. While once based on the theoretical assumption of ethnocentrism, this has given way to the concept of cultural relativism. Thus, to understand the culture of a social group, one needs to shed any preconception of the existence of a “standard” society. Theoretical preparation and openness is needed in order to carefully listen to and observe a group and record the information in field notes, diaries and other instruments for detailed and in-depth observation.[15] Originally affiliated with the interpretative paradigm of knowledge production, ethnography currently presents several critical orientations, with anthropologists studying, among other things contemporary urban groups, or the organisation and process of work.[8] Research by Wainwright[16] in Uruguay, for example, has taken an ethnographic approach to understanding the intersections between the sensation of breathlessness for people with chronic obstructive pulmonary disease, the cultural context and the political economy of health, and was recently included in a QES about the experience of chronic breathlessness.[17] Another example is by Souza and Mendes[18] who used **critical ethnography** in a research study based on a Marxist theoretical-philosophical framework. They studied nursing work in hospitals, looking specifically at the organisational structure of hospitals and the flexibilisation of contractual relationships between health professionals within the context of financialised capitalism.

Case studies use one or more ethnographic, phenomenological, or participatory methodologies, among others, to illuminate and illustrate issues specific to certain contexts and under a theoretical-analytical framework that helps us better understand them. For example, case studies about work, needs and social class, based on a Marxist theoretical-philosophical framework and aligned with the critical paradigm, are increasingly being conducted in the health field. Santos, Soares and Campos[19], for example, based on in-depth interviews with nurses about work processes in Primary Care, relate nurses' weary behaviour to the post-Fordism organisation of work. Campos[20], by analysing in-depth interviews with workers of a general practice unit about health needs, showed that work processes had been established prior to recognizing the needs of the social groups served by the unit, thus reversing the rationale that health services should respond to needs. Sousa[21], from the theoretical-philosophical perspective of science and technology studies, conducted a case study about the Brazilian Consortium of Research and Implementation of Multiple Endocrine Neoplasia Type 2 (BrasMEN[26]). Through fieldnotes[12,22] and (self-validated[23]) narratives of oral life history[24,25] she identified the attitudes and perceptions of all stakeholders on the incorporation of familial genetic diagnosis in the Unified Health System (in Portuguese, *Sistema* Único *de Saúde*, SUS) with the National Commission for the Incorporation of Technologies in SUS (CONITEC) – the health technology assessment agency of the Ministry of Health.

Participatory methodologies, and particularly participatory action research[27], are widely used in health research.[28] There is a diverse set of strands within participatory methodologies, but we can say that they all have in common the principles of: joint production of the research process with individuals and social groups whose problems and conditions are the object of study; research questions and objectives developed from the convergence of two perspectives, that of science and practice; co-creation allowing co-researchers to take a step back and question and review established interpretations of a particular problem/condition; convergence of both science and practice perspectives is a very demanding process, which evolves when these two realms of action meet, interact and develop in mutual understanding.[29] **Participatory research** has had a strong presence in Latin America, converging with social movements in the 1960s and 1980s in response to the presence of authoritarian regimes as well as exclusionary and concentrated development models in the region.[30] Freire and Fals Borda's educational strand[31], which emphasises the role of the organic intellectual and the liberating purpose of research, was shaped by humanistic and revolutionary principles. Others have outlined the potential of this kind of research to promote the displacement of universities towards concrete fields of reality.[32] The theoretical-methodological assumptions of bourgeois science were questioned, and new paradigms were sought with the aim to conduct research that would contribute to overcoming serious problems arising from class contradictions and to promote social change.[33] Vecchia and Martins[34], for example, conducted **participatory research** under Vigotsky's historical-cultural strand, which analysed personal and social meanings of mental health care activities developed by professionals who are members of a family health program team.

Qualitative Evidence Synthesis

The practice of conducting in-depth literature reviews – and their systematisation – as a way of taking stock of the state of our knowledge, concepts and theory in certain areas is nothing new. Adopting the "systematic" approach to a literature review of qualitative studies is more so. Systematic reviews differ from literature reviews in their structured methodology, rigorous approach to comprehensively searching the body of literature on a topic, their team approach and an emphasis on transparency in reporting, to name but a few. Systematic reviews, to be labelled as such, follow a specific set of steps (whether of qualitative or quantitative studies), including developing a review question, developing a search strategy, searching databases, screening studies for inclusion and exclusion, extracting data, appraising the quality of primary studies, and analysing and synthesising data. Though the broad steps in QES are similar to those of quantitative reviews of effectiveness, the substance of how these steps are taken differs due to the "dual heritage for QES methodology" – on one side influenced by the methods for systematic reviews of effectiveness and on the other shaped strongly by primary qualitative research methodologies.[35] Due to the number of steps, as well as the time-consuming nature of analysing qualitative data, systematic reviews of qualitative research are no small feat. They require substantial time, energy and commitment. Despite such huge efforts, what has been driving the increased demand for, and investment of resources into, producing qualitative evidence syntheses?

The drive for systematic reviews in the health sector is tied to the movement for evidence-based practice, treatment and policy. To ensure recommendations for health and policy reflect our state of knowledge on the topic, one needs to review what the evidence says. Historically the questions driving the demand to search and synthesize all existing evidence are those around treatment efficacy, safety or cost effectiveness. However as there is increasing recognition that the existence of effective and cost-effective treatments does not automatically translate into better health outcomes for all, the importance of posing other types of questions becomes obvious. Why do men not attend the GP for preventive care? How does past experience shape perceptions of a new program for people with mental illness? What is the most acceptable option, according to stakeholders, for meeting staff shortages? What are the factors influencing the uptake of clinical guidelines in rural settings? What are the obstacles to delivering healthcare to homeless immigrants? Why do certain preventive programs for drug use not work?

Primary qualitative research (as described in the previous section) is well suited to tackle these complex questions. Popay and collaborators[36] affirmed more than twenty years ago that effectiveness evidence will never be enough – evidence is also needed on topics such as appropriateness as well as the factors that shape decision-making. In this case, the "evidence" is not numbers, correlations or statistics, but rather what people think, feel, say, experience or observe in real-world settings. When these types of questions are being posed in a decision-making context (to inform the best choice of policy, investment, or innovation for example), a qualitative evidence synthesis may be commissioned.

Among those who increasingly use QES is the World Health Organization (WHO).[37] The WHO Handbook for Guideline Development includes a chapter on QES as they are increasingly commissioned as part of the process of development of clinical guidelines and those for the implementation of (new) health technologies.[38] The chapter outlines some of the ways qualitative evidence can be used in a guideline. For one it can be used to determine the scope of the guideline itself by considering all topics relevant to stakeholders, and help to better specify the review question itself. Qualitative evidence can shed light on how different stakeholders value different outcomes, or how important an intervention's potential benefits and harms are to people, the acceptability and feasibility of interventions, the factors that shape implementation, and the effect of interventions on health equity. Lewin and Glenton[39] describe their experience of working with the WHO in producing QES to inform guidelines. They give the example of how qualitative evidence was used to inform the WHO recommendations on "antenatal care for positive pregnancy experience".[40] Firstly, a QES was commissioned to ensure women's voices shaped the development of the guidance from the start. This was on the topic of what women need, want and value in pregnancy [41], and was used to determine broad aims and key outcomes to consider in the guideline. A second QES was commissioned to explore factors influencing women's use of antenatal services, which in turn contributed evidence on acceptability and feasibility to the guideline.[39,42]

In this sense, QES enables researchers and policy and decision-makers to "go beyond" the findings of primary qualitative research studies and produce something that is more than their simple summary[5] – they are interpretative syntheses of findings from these studies.[3] As such, the synthesis of several relevant qualitative studies on a particular topic can offer multiple perspectives, as well as provide evidence of contradictory viewpoints that might otherwise be missed when considering a single study alone. It is important to recognise that qualitative synthesis is different from a summary of qualitative studies. Eaton[44] gives a quick overview of the differences between summarising and synthesising information, some of which are presented in Table 1.

Finally, we outline that, for the purpose of health technology assessment (HTA) contexts, institutions like the National Institute of Health and Clinical Excellence (NICE) and the Scottish Medical Council (SMC) are also using qualitative evidence in their guidelines. Carroll[5] outlines how the NICE

Table 1. Summary versus Synthesis

Summary	Synthesis
Pulls together information in order to highlight the important points.	Pulls together information not only to highlight the important points, but also to draw your own conclusions.
Addresses one set of information (e.g. article, chapter, document) at a time. Each source remains distinct.	Combines parts and elements from a variety of sources into one unified entity.
Demonstrates an understanding of the overall meaning.	Achieves new insight.

guideline for long-term management of stroke integrated findings from quantitative and qualitative evidence syntheses. In particular, the influence of the qualitative evidence and its synthesis is quite clear: the quantitative evidence only noted that standard procedures were not conducive to shared decision making; the qualitative evidence emphasised the importance of shared decision making and the specifics of how it should be achieved, and these were integrated in detail into the recommendations. Such use of qualitative evidence within HTA contexts in Brazil is also starting to be integrated with the Clinical Protocols and Therapeutic Guidelines (PCDT) formulated by CONITEC.[45]

Overall, there is increasing recognition of the contribution qualitative evidence can make and in turn syntheses of this evidence are being incorporated into decision-making and guideline processes. The paper by Barreto and Lewin[46] in this BIS issue provides additional commentary on using qualitative evidence to inform decisions. Though typically this has been driven within the field of health, interest is increasing in other domains, such as environmental policy and management[47]. Undoubtedly, it is an exciting time for qualitative research. Upcoming publications of interest in this area include using qualitative evidence to determine guideline scope[48], to inform evidence to decision frameworks[49], and to develop implementation considerations.[50] Let us now know look at some of the specific methods used for undertaking a QES.

Qualitative Evidence Synthesis Methods

Just as there are many types of analysis for qualitative data, there are numerous approaches and methods for synthesizing primary qualitative studies. It is a crucial step of the review process to decide what type of synthesis will be conducted and later to report this clearly in the final outputs and publications. There have been a proliferation of names for different synthesis methods making the navigation and selection of the appropriate method that much more complex for reviewers. A starting point for lists with practical and conceptual definitions of different synthesis methods can be found in the critical review of methods by Barnett-Page and Thomas[51], the book by Hannes and Lockwood[52] or the guide by Ring and collaborators[53] for synthesizing qualitative research in health technology assessment. These authors focus on the five to ten most commonly used, or best developed, synthesis methods. Though it is common to hear from experts in the field that there exists more than forty types of synthesis (many only used in one review), we could not find a comprehensive list of all potential options. It seems far easier to find guidance on specific approaches to synthesis (e.g. thematic synthesis[54], meta-ethnography[55], critical interpretive synthesis[56], meta-aggregation[57,58], framework synthesis[59], and best-fit framework synthesis[60]) than to find a starting list of all potential options – something which may pose a challenge for new and experienced reviewers alike.

Currently, there does exist general guidance on how to choose an appropriate synthesis method. Booth and collaborators[61] developed the RETREAT criteria for selecting qualitative evidence synthesis methods. The RETREAT mnemonic makes reference to seven domains on which the choice of method should be made – the Review question (more or less fixed), Epistemology (philosophical underpinning), Time/timescale (how long do you have to finish the review), Resources (financial and human), Expertise (what skills and background reviewers have), Audience and purpose (who and what the findings of the review are for), and Type of data (in the primary qualitative studies). Rather than try to summarize all potential synthesis methods, the authors developed a series of prompts for reviewers to respond to in order to guide them towards a well-justified choice of synthesis method. These domains and prompts can be applied when considering any choice of synthesis method.

Guidance has also been developed for choosing methods in specific review circumstances, for example in the context of a complex intervention and guideline development[62-65]. Flemming and collaborators[62] identify three QES synthesis methods particularly suited to exploring complexity: thematic synthesis, framework synthesis or best-fit framework synthesis, and meta-ethnography. Noyes and collaborators[63], on behalf of the Cochrane Qualitative and Implementation Methods Group, also recommend these same methods when the QES will subsequently be integrated with an intervention effect review for use in decision-making. They provide a useful comparison of the pros and cons of each method when producing a synthesis for a decision-making process. Booth and collaborators[64] also provide a comparison of different synthesis methods (Table 1, p. 16 from this reference) in their guide for selecting a synthesis method for health technology assessment reviews of complex interventions. Kastner and collaborators[65] provide an algorithm for assisting with the selection of a synthesis method for complex evidence (qualitative or quantitative) based on purpose, outputs and applicability and compare a variety of types of synthesis.

Fundamentally, choosing a synthesis method takes careful consideration about the purpose of its production and uses. With the aim of producing health practice statements, meta-aggregation is the QES process advocated by the Joanna Brigs Institute (JBI). Findings from primary studies are accurately extracted and grouped into categories according to similarity of meaning. The gathering of several categories in a line of conduct constitutes the aggregative synthesis.[57,66] Meta-ethnographies in turn are intended to produce theories and, for such purpose, one needs conceptually rich and thickly described findings in the primary studies. For this reason at least, reviewers are recommended to make a final decision on synthesis method only once they know what they will be working with in terms of primary studies, purpose and uses.[63]

Other Tools and Guides for Qualitative Evidence Synthesis

Some organizations that publish "branded" reviews (e.g. Cochrane or a JBI Review) provide their own manuals to guide the whole review process – Cochrane Handbook[67], JBI Reviewer's Manual.[58] However, most reviews will be produced outside of these contexts and the peer-reviewed literature is an ever-expanding source of guides and tools to assist reviewers in different stages of conducting a QES. Here we briefly signpost those for developing a review question and searching, preparing a protocol, assessing confidence in review findings, and writing-up and reporting the review.

Developing a review question and searching the literature

The review question, as emphasised above in the RETREAT framework, is key to choosing a synthesis method. It is also key for developing an effective literature search. A number of frameworks for developing review questions specific to qualitative evidence synthesis have been developed. Here is a small selection (for a more comprehensive list see Booth[1]) of acronyms:

- SPIDER[68] (Sample, Phenomenon of Interest, Design, Evaluation, Research type);
- PICOC[69] (Patient/Population, Intervention, Comparison, Outcomes, Context) – a variation of PICO better suited to qualitative evidence synthesis for its inclusion of "context";
- PerSPEcTiF[35] (Perspective, Setting, Phenomenon of Interest/Problem,

Environment, Comparison (optional), Time/Timing, Findings);

- PICo[70] (Participants, Phenomenon of Interest and Context): mimicking the PICO of efficacy studies, fits into qualitative research, emphasising key elements of this type of research.

The Cochrane Qualitative and Implementation Methods Group consider the SPICE[69] (Setting, Perspective, Intervention/phenomenon of Interest, Comparison, Evaluation) to be the simplest framework. However, although useful for defining concepts that will go into a search strategy it might not be as suited to reviews of complex interventions for which the issue of context is fundamental[69]. Booth and collaborators[35] developed PerSPEcTiF to address perceived weaknesses in other question formulation frameworks for QES. Having a well-defined question is crucial for designing effective databases searches. When it comes to searching, the review by Booth[1] provides some starting principles for searching for qualitative studies including: sampling, sources, structured questions, search procedures, search strategies and filters. These are also summarized in Harris and collaborators.[69]

Preparing a Protocol

QES can be registered in databases such as PROSPERO[IV] to help reduce the likelihood of duplication.[71] Harris and collaborators[69] provide a brief overview of what to include in a protocol, and Butler and collaborators[72] provide an extensive guide to writing QES protocol. In this sense, considering social theories is important from the very start of a review [73], as we have previously discussed in the section on qualitative research methodologies. Specific methods for protocol development are provided by evidence synthesis centres such as Cochrane and JBI, which provides guidance for the preparation of the qualitative evidence synthesis protocol, and publishes protocols accepted after submission and peer review in the Cochrane Library and JBI Database of Systematic Reviews and Implementation Reports[70,74], respectively.

Assessing confidence in review findings

A qualitative evidence synthesis will produce a multitude of review findings and in a decision-making context, it is important for the user of the review to know how much confidence to place in each individual review finding. The GRADE-CERQual (Grading of Recommendations Assessment, Development and Evaluation – Confidence in the Evidence from Reviews of Qualitative research) approach was developed to provide a transparent and systematic way of assessing confidence in individual review findings. [75,76] Detailed guidance exists to assist reviewers in applying the CERQual approach – a process that involves first assessing each individual component (methodological limitations[77], coherence[78], relevance[79], and adequacy[80]) before making an overall assessment of confidence for each individual review finding[81]. Guidance is also included for presenting synthesized findings and GRADE-CERQual assessments of confidence in a Summary of Qualitative Findings Table. [81] GRADE-CERQual approach was designed to be applied to findings of qualitative evidence syntheses using any type of synthesis method. Reviewers using the JBI meta-aggregation method also have the option of using the ConQual tool for assessing confidence in their review findings.[82]

Critical appraisal of primary studies

Critical appraisal (also called quality appraisal) is also a key step in a systematic review. In order to assess the methodological limitations component when making a GRADE-CERQual

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[IV] https://www.crd.york.ac.uk/prospero/

assessment of confidence (section above), reviewers must apply a critical appraisal tool to their primary studies and make a judgement as to whether there are any methodological concerns that might lower our confidence in the review finding. Some types of review method, like the JBI method, may exclude studies from the review based on methodological quality and therefore apply appraisal tools at the start of the review, before synthesis. It is imperative to use an appraisal tool designed to appraise qualitative research. Applying an appraisal tool for quantitative studies to a qualitative study would be fundamentally flawed. A recent compendium and appraisal-tool selection guide can help reviewers choose the best tool for their needs.[83]

It is important not to confuse findings of primary studies and findings of a qualitative evidence synthesis when it comes to appraisal of quality and confidence. For instance, while confidence in a primary study may relate primarily to its methodological quality, confidence in a review finding depends on much more than just the methodological quality of the primary studies underlying it. For example the GRADE-CERQual approach assesses confidence in a review finding based on the coherence, relevance and adequacy of data of review findings, and not exclusively on the methodological limitations of individual primary studies that have contributed to each review finding.

Writing up and reporting the review

When it comes to reporting and publishing one's review, there are an increasing number of reporting guidelines available for different aspects of the review. ENTREQ is an overall reporting guideline for all types of QES[84], while eMERGe is specific to meta-ethnography.[85] PRISMA diagrams help represent the screening and inclusion/exclusion process.[86] Authors can also draw on the STARLITE[87] mnemonic in reporting their search strategies (sampling strategy, type of study, approaches, range of years, limits, inclusion and exclusions, terms used, electronic sources). There are new reporting guidelines developing all the time and reviewers can use the EQUATOR[v] Network database to help find existing reporting guidelines for QES.

Final Considerations

QES is a systematic and structured process that has as a starting point in qualitative categories of analysis – i.e. those related to understanding the reality of human and social relationships and those seeking contextual, historical, and intersubjective explanations for the reality of these relationships. Therefore, QES have the potential to synthesise empirical categories such as perceptions, meanings, experiences, beliefs, and representations, amongst many others.

By synthesizing categories within the realm of human and social relations, we expect to contribute the necessary elements for enhancing the purpose and implementation of technologies, interventions, programs and policies that improve health care, individually and collectively. Ultimately, we expect QES to help us respond to the health needs of different individuals, families, and social groups that make up society.

References

1. Booth A. Searching for qualitative research for inclusion in systematic reviews: a structured methodological review. Syst Rev. 2016;5(1). doi:10.1186/s13643-016-0249-x
2. Malterud K. Qualitative research: standards, challenges, and guidelines. *The Lancet*. 2001;358(9280):483-8.
3. Snape D, Spencer L. The Foundations of Qualitative Research. In: Ritchie J, Lewis J, editors. Qualitative Research Practice A Guide for Social Science Students and Researchers: SAGE; 2009.

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[v] http://www.equator-network.org/

4. Minayo MCdeS, Costa AP. Técnicas que fazem uso da palavra, do olhar e da empatia: Pesquisa qualitativa em ação. Oliveira de Azemeis: Aveiro (PT); 2019.
5. Carroll C. Qualitative evidence synthesis to improve implementation of clinical guideline. BMJ. 2017;356:j80 doi: 10.1136/bmj.j80
6. Soares CB. Revisão qualitativa da literatura com enfoque na revisão sistemática. In: Costa AP. Revisão da literatura com apoio de software: Contribuição da pesquisa qualitativa. Ludomedia: Aveiro (PT); 2019.
7. Soares CB, Yonekura T, Campos CMS. Marxismo como referencial teórico-metodológico em saúde coletiva: implicações para a revisão sistemática e síntese de evidências. Rev Esc Enferm USP. 2013;47(6):1403-1409.
8. Prasad P. Crafting qualitative research: Working in the postpositivist traditions. New York: Routledge; 2015.
9. Cordeiro L, Soares CB, Rittenmeyer L. Unscrambling method and methodology in action research traditions: theoretical conceptualization of praxis and emancipation. Qualitative Research. 2017;17(4):395–407. https://doi.org/10.1177/1468794116674771
10. Guba EC. The alternative paradigm dialog. In: Guba EC (org). The paradigm dialog. Newbury Park, London, New Delhi: Sage Publications. 1990:17-27.
11. Minayo MCS. O desafio do conhecimento: pesquisa qualitativa em saúde. 3ªed. São Paulo: Hucitec-Abrasco; 1992.
12. Denzin NK, Lincoln YS, Editors. The handbook of qualitative research. Thousand Oaks: Sage. 2011.
13. Willis JW. Foundations of qualitative research: interpretive and critical approaches. London: Sage. 2007.
14. Sodelli M. Aproximando sentidos: formação de professores, educação, drogas e ações redutoras de vulnerabilidade. 250 f. Tese (Doutorado em Psicologia) – Pontifícia Universidade Católica de São Paulo, São Paulo, 2006.
15. Nakamura E. The ethnographic method in health researches: an anthropological thinking. Saúde e Sociedade. 2011;20(1):95-103.
16. Wainwright M. Sensing the airs: The cultural-context for breathing and breathlessness in Uruguay. Medical Anthropology. 2017;36(4):332-347. Doi: 10.1080/01459740.2017.1287180
17. Hutchinson A., Barclay-Klingle, N., Galvin, K., Johnson, M.J. (2018). Living with breathlessness: a systematic literature review and qualitative synthesis. Eur Respir J. 2018; 51:1701477 [https://doi.org/10.1183/13993003.01477-2017].
18. Souza HS, Mendes ÁN. A terceirização e o “desmonte” do emprego estável em hospitais. Rev. esc. enferm. USP [Internet]. 2016 Apr; 50(2): 286-294. http://dx.doi.org/10.1590/S0080-623420160000200015.
19. Santos VC, Soares CB, Campos CMS. A relação trabalho-saúde de enfermeiros do PSF no município de São Paulo. Rev Esc Enferm USP. 2007;41(Esp):777–781.
20. Campos CMS. Necessidades de saúde pela voz da sociedade civil (os moradores) e do Estado (os trabalhadores de saúde). [tese]. São Paulo: Escola de Enfermagem, Universidade de São Paulo; 2004.
21. Sousa MSA. Consórcio Brasileiro ‘Neoplasia Endócrina Múltipla Tipo 2’: Medicina Translacional, Bioética e Sociedade. 725 f. Tese (Doutorado) – Escola Paulista de Medicina, Universidade Federal de São Paulo (UNIFESP), São Paulo, 2015. Disponível em: http://repositorio.unifesp.br/handle/11600/48737
22. Geertz C. Works and Lives: The Anthropologist as Author, Stanford University Press. 1988.
23. Mays N, Pope C. Qualitative research in health care. Assessing quality in qualitative research. BMJ. 2000;320:50-2.
24. Meihy JCSB, Holanda F. História Oral: como fazer, como pensar, São Paulo, Contexto. 2007.
25. Meihy JCSB, Ribeiro SLS. Guia prático de história oral: para empresas, universidades, comunidades e famílias, São Paulo, Contexto. 2011.
26. Maciel RMB, Camacho CP, Assumpção LVM, et al. Genotype and phenotype landscape of MEN2 in 554 medullary thyroid cancer patients: the BrasMEN study. Endocrine Connections. 2019;8(3):289-298.
27. Cordeiro L, Soares CB, Rittenmeyer L. Unscrambling method and methodology in action research traditions: theoretical conceptualization of praxis and emancipation. Qualitative Research. 2017;17(4):395–407. https://doi.org/10.1177/1468794116674771
28. Toledo RF, Eosa TEC, Keinert TM, Cortizo CT, organizadores. Pesquisa Participativa em Saúde: vertentes e veredas. São Paulo - SP: Editora do Instituto de Saúde. 2018;24:153-165.
29. Bergold J, Thomas S. Participatory Research Methods: A Methodological Approach in Motion. Forum: Qualitative Social Research. 2012 Jan[S.l.];13(1). ISSN 1438-5627. Available at: <http://www.qualitative-research.net/index.php/fqs/article/view/1801/3334>.
30. Cajardo M. Pesquisa participante: propostas e projetos. In: Brandão CR, organizador. Repensando a pesquisa participante. 3a.ed. São Paulo: Brasiliense. 1987:15-50.

31. Haguete TMF. Metodologias qualitativas na sociologia. 3a.ed. Petrópolis: Vozes. 1992.
32. Borda OF. Aspectos teóricos da pesquisa participante: considerações sobre o significado e o papel da ciência na participação popular. In: Brandão CR. (Org.) Pesquisa participante. São Paulo: Brasiliense. 1981:42-62.
33. Peruzzo CMK. Da observação participante à pesquisa-ação: pressupostos epistemológicos e metodológicos. Apresentado no III Colóquio Brasil-Itália de Ciências da Comunicação, realizado em Belo Horizonte – MG 2 e 3 de setembro de 2003. Promoção: INTERCOM – Sociedade Brasileira de Estudos Interdisciplinares da Comunicação.
34. Vecchia MD, Martins STF. Concepções dos cuidados em saúde mental por uma equipe de saúde da família, em perspectiva histórico-cultural. Ciênc. saúde coletiva. 2009 Feb;14(1):183-193. Available from: http://dx.doi.org/10.1590/S1413-81232009000100024.
35. Booth A. Harnessing Energies, Resolving Tensions: Acknowledging a Dual Heritage for Qualitative Evidence Synthesis. Qualitative Health Research. 2019;29(1):18–31. https://doi.org/10.1177/1049732318808247
36. Popay J, Rogers A, Williams G. Rationale and standards for the systematic review of qualitative literature in health services research. Qualitative Health Research. 1998;8(3):341-351.
37. Langlois EV, Tunçalp Ö, Norris SL, Askew I, Ghaffar A. Qualitative evidence to improve guidelines and health decision-making. Bulletin of the World Health Organization 2018;96:79-79A. doi: http://dx.doi.org/10.2471/BLT.17.206540
38. Glenton C, Lewin S, Norris SL. Usando evidencias de pesquisa qualitativa para desenvolver diretrizes da OMS. (Traduzido para o Português por Maria Sharmila Alina de Sousa e Ananyr Porto Fajardo). WHO handbook for guideline development. 2014. (ISBN 978 92 4 154896 0)
39. Lewin S, Glenton C. Are we entering a new era for qualitative research? Using qualitative evidence to support guidance and guideline development by the World Health Organization. International Journal for Equity in Health. 2018;17:126 https://doi.org/10.1186/s12939-018-0841-x
40. WHO. WHO recommendations on antenatal care for a positive pregnancy experience. Geneva: World Health Organization. 2016.
41. Downe S, Finlayson K, Tuncalp Ö, Metin Gulmezoglu A. What matters to women: a systematic scoping review to identify the processes and outcomes of antenatal care provision that are important to healthy pregnant women. BJOG. 2016;123(4): 529–39.4
42. Downe S, Finlayson K, Tunçalp Ö, Gülmezoglu AM. Factors that influence the uptake of routine antenatal services by pregnant women: a qualitative evidence synthesis (Protocol). Cochrane Database Syst Rev. 2016;10: CD012392.
43. Campbell R, Pound P, Pope C, Britten N, Pill R, Morgan M, et al. Evaluating meta-ethnography: a synthesis of qualitative research on lay experiences of diabetes and diabetes care. Social Science & Medicine. 2003;56:671-84.
44. Eaton SE. Reading Strategy: Differences between summarising and synthesising information. 2010. Available from: https://www.scribd.com/doc/38175256/Differences-Between-Summarizing-and-Synthesizing-Information.
45. Silva AS, Sousa MSA, Silva EV, Galato D. Participação Social no Processo de Incorporação de Tecnologias em Saúde no SUS. Revista de Saúde Pública. 2019 (no prelo).
46. Barreto J, Lewin S. Using qualitative evidence to inform decisions in Brazil and the Latin American region. Boletim de Saúde – BIS. 2019 (no prelo).
47. Macura B, Suškevičs M, Garside R, Hannes K, Rees R, Rodela R. Systematic reviews of qualitative evidence for environmental policy and management: an overview of different methodological options. Environmental Evidence. 2019;8:24.
48. Downe S, Finlayson KW, Lawrie TA, Lewin SA, Glenton C, Rosenbaum S, Barriex M, Tuncalp Ö. Qualitative Evidence Synthesis for Guidelines: Paper 1 - Using Qualitative Evidence Synthesis to Inform Guideline Scope and Develop Qualitative Findings Statements. Health Research Policy and Systems. 2019;17(76).
49. Lewin SA, Glenton C, Lawrie TA, Downe S, Finlayson KW, Rosenbaum S, Barriex M, Tuncalp Ö. Qualitative Evidence Synthesis for Guidelines: Paper 2 - Using Qualitative Evidence Synthesis Findings to Inform Evidence-to-Decision Frameworks and Recommendations. Health Research Policy and Systems. 2019;17(75).
50. Glenton C, Lewin SA, Lawrie TA, Barriex M, Downe S, Finlayson KW, Tamrat T, Rosenbaum S, Tuncalp Ö. Qualitative Evidence Synthesis for Guidelines: Paper 3 - Using Qualitative Evidence Syntheses to Develop Implementation Considerations and Inform Implementation Processes. Health Research Policy and Systems. 2019;17(74).
51. Barnett-Page E, Thomas J. Methods for the synthesis of qualitative research: a critical review. NCRM Working Paper. NCRM. 2009. Available from: http://eprints.ncrm.ac.uk/690/

52. Hannes K, Lockwood C. Synthesizing Qualitative Research: Choosing the Right Approach John Wiley & Sons, Ltd, Chichester, UK. 2012.
53. Ring N, Ritchie K, Mandava L, Jepson R. A guide to synthesising qualitative research for researchers undertaking health technology assessments and systematic reviews. 2010. Available from: http://www.nhshealthquality.org/nhsqis/8837.html
54. Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. BMC Med Res Methodol. 2008 Jul 10;8:45 http://ukpmc.ac.uk/articlerender.cgi?artid=1593387
55. Noblit GW, Hare RD. Meta-ethnography: synthesising qualitative studies Sage: Thousand Oaks, CA. 1988:88.
56. Dixon-Woods M, Cavers D, Agarwal S, et al. Conducting a critical interpretive synthesis of the literature on access to healthcare by vulnerable groups. BMC Medical Research Methodology. 2006;6(35).
57. Lockwood C, Munn Z, Porritt K. Qualitative research synthesis: methodological guidance for systematic reviewers utilizing meta-aggregation. International journal of evidence-based healthcare. 2015;13(3):179-187.
58. Aromataris E, Munn Z (Editors). Joanna Briggs Institute Reviewer's Manual. The Joanna Briggs Institute> 2017. Available from https://reviewersmanual.joannabriggs.org/
59. Dixon-Woods M. Using framework-based synthesis for conducting reviews of qualitative studies. BMC Med. 2011 Apr;14(9):39.
60. Carroll C, Booth A, Leaviss J, Rick J. "Best fit" framework synthesis: refining the method. BMC Medical Research Methodology. 2013;13(1):1-16.
61. Booth A, Noyes J, Flemming K, Gerhardus A, Wahlster P, van der Wilt GJ, et al. Structured methodology review identified seven (RETREAT) criteria for selecting qualitative evidence synthesis approaches. J Clin Epidemiol. 2018;99:41-52.
62. Flemming K, Booth A, Garside R, Tunçalp Ö, Noyes J. Qualitative evidence synthesis for complex interventions and guideline development: clarification of the purpose, design and relevant methods. BMJ Glob Health. 2019;4:e000882. doi:10.1136/ bmjgh-2018-000882
63. Noyes J, Booth A, Flemming K, Gerhardus A, Wahlster P, van der Wilt GJ, et al. Cochrane Qualitative and Implementation Methods Group guidance paper 3: methods for assessing methodological limitations, data extraction and synthesis, and confidence in synthesized qualitative findings J Clin Epidemiol. 2018; 97:49-58
64. Booth A, Noyes J, Flemming K, Gerhardus A, Wahlster P, Van Der Wilt GJ, Mozygemba K, Refolo P, Sacchini D, Tummers M, Rehfuess E. Guidance on choosing qualitative evidence synthesis methods for use in health technology assessments of complex interventions. 2016. Available from: http://www.integrate-hta.eu/downloads/
65. 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. Journal of Clinical Epidemiology. 2016;73:43-49.
66. Hoga LAK, Matheus MCC, Soares CB. Metassíntese: síntese de estudos qualitativos. In Enfermagem baseada em evidências. São Paulo: Barbosa, Dulce; Taminato, Mônica; Fram, Dayana; Belasco, Angélica. 2014:105-116.
67. Noyes J, Hannes K, Booth A, Harris J, Harden A, Popay J, Pearson A, Cargo M, and Pantoja T on behalf of the Cochrane Qualitative and Implementation Methods Group. Chapter 20: Qualitative research and Cochrane reviews. In: Higgins JPT, Green S (editors), Cochrane Handbook for Systematic Reviews of Interventions Version 5.3.0 (updated October 2015). The Cochrane Collaboration. 2015. Available from http://qim.cochrane.org/supplemental-handbook-guidance [Current update process January 2018 onwards]
68. Cooke A, Smith D, Booth A. Beyond PICO: The SPIDER Tool for Qualitative Evidence Synthesis. Qual Health Res. 2012;22(10):1435-1443.
69. Harris JL, Booth A, Cargo M, Hannes K, Harden A, Flemming K, Garside R, Pantoja T, Thomas J, Noyes J. Cochrane Qualitative and Implementation Methods Group guidance series-paper 2: methods for question formulation, searching, and protocol development for qualitative evidence synthesis. J Clin Epidemiol. 2018 May;97:39-48. doi: 10.1016/j.jclinepi.2017.10.023.
70. Lockwood C, Porrit K, Munn Z, Rittenmeyer L, Salmond S, Bjerrum M, … Stannard D. Systematic reviews of qualitative evidence. In E. Aromataris & Z. Munn (Orgs.), Joanna Briggs Institute Reviewer's Manual. The Joanna Briggs Institute. 2017. Recuperado de https://reviewersmanual.joannabriggs.org/
71. Centre for Review and Dissemination. Guidance notes for registering a systematic review protocol with PROSPERO. 2016 Recuperado de https://www.crd.york.ac.uk/PROSPERO/documents/Registering a review on PROSPERO.pdf
72. Butler A, Hall H, Copnell B. A Guide to Writing a Qualitative Systematic Review Protocol to Enhance

Evidence-Based Practice in Nursing and Health Care. Worldviews on Evidence-Based Nursing. 2016;3(3):241–249. doi:10.1111/wvn.12134

73. Noyes J, Hendry M, Booth A, Chandler J, Lewin S, Glenton C, Garside R. Current use was established and Cochrane guidance on selection of social theories for systematic reviews of complex interventions was developed. Journal of Clinical Epidemiology. 2016;75:78-92. doi:10.1016/j.jclinepi.2015.12.009
74. Matheus MCC, Hoga LAK, Soares CB. Projeto de revisão sistemática e estudos qualitativos (RSEQ). In Barbosa D, Taminato M, Fram D, Belasco A (Orgs.), Enfermagem Baseada em Evidências. 1º ed. São Paulo: Atheneu. 2014:93-103.
75. Lewin S, Glenton C, Munthe-Kaas H, Carlsen B, Colvin CJ, Gülmezoglu M, ... Booth A, Rashidian A. Using qualitative evidence in decision making for health and social interventions: an approach to assess confidence in findings from qualitative evidence syntheses (GRADE--CERQual). PLoS Med. 2015;12(10):e1001895.
76. Lewin S, Booth A, Glenton C, et al. Applying GRADE--CERQual to qualitative evidence synthesis findings: introduction to the series. Implement Sci. 2018;13(Suppl 1):2. doi: 10.1186/s13012-017-0688-3.
77. Munthe-Kaas H, Bohren MA, Glenton C, Lewin S, Noyes J, Tunçalp Ö, Booth A, et al. Applying GRADE--CERQual to qualitative evidence synthesis findings--paper 3: how to assess methodological limitations. Implement Sci. 2018;13(Suppl 1):9. doi: 10.1186/s13012-017-0690-9.
78. Colvin CJ, Garside R, Wainwright M, Munthe-Kaas H, Glenton C, Bohren MA, Carlsen B, Tunçalp Ö, Noyes J, Booth A, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 4: how to assess coherence. Implement Sci. 2018;13(Suppl 1):13. doi:10.1186/s13012-017-0691-8
79. Noyes J, Booth A, Lewin S, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 6: how to assess relevance of the data. Implement Sci. 2018;13 (Suppl 1):4. doi: 10.1186/s13012-017-0693- 6
80. Glenton C, Carlsen B, Lewin S, Munthe-Kaas H, Colvin CJ, Tunçalp Ö, Bohren MA, Noyes J, Booth A, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 5: how to assess adequacy of data. Implement Sci. 2018;13(Suppl 1):14. doi: 10.1186/s13012-017-0692-7.
81. Lewin S, Bohren M, Rashidian A, Munthe-Kaas H, Glenton C, Colvin CJ, Garside R, Noyes J, Booth A, et al. Applying GRADE-CERQual to qualitative evidence synthesis findings-paper 2: how to make an overall CERQual assessment of confidence and create a Summary of Qualitative Findings table. Implement Sci. 2018;13(Suppl 1):10. doi: 10.1186/s13012-017-0689-2.
82. Munn Z, Porritt K, Lockwood C, Aromataris E, Pearson A. Establishing confidence in the output of qualitative research synthesis: The ConQual approach. BMCMedical Research Methodology. 2014;14(108):1-7. https://doi.org/10.1186/1471-2288-14-108.
83. Majid U., Vanstone M. Appraising Qualitative Research for Syntheses: A Compedium of Quality Appaisal Tools. Qual Health Res. 2018;28(13):2115-2131. doi: 10.1177/1049732318785358.
84. Tong A, Flemming K, McInnes E, Oliver S, Craig J. Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. BMC Medical Research Methodology. 2012;12(1):181.
85. France EF, Cunningham M, Ring N, Uny I, Duncan EAS, Jepson RG, Maxwell M, Roberts RJ, Turley RL, Booth A, et al. Improving reporting of Metaethnography: the eMERGe reporting guidance. J Adv Nurs. 2019. https://doi.org/10.1111/jan.13809.
86. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med. 2009;6(6):e1000097
87. Booth A. ‘‘Brimful of STARLITE’’: toward standards for reporting literature search. J Med Libr Assoc. 2006;94(4):421-429.

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Using qualitative evidence to inform decisions in Brazil and the Latin American region

Jorge Barreto[I], Simon Lewin[II, III, IV]

Abstract

Interest in how to strengthen the use of research evidence in decision-making processes has grown rapidly in many settings. Evidence informed policy making has now become widely institutionalised within the health sector and is also being implemented within many other areas of social policy. Qualitative evidence – empirical data drawn from research that uses qualitative methods of data collection and analysis – can play a critical role in evidence informed policy making because such evidence can capture the views and experiences of those affected by an intervention or option. Qualitative evidence also has the potential to humanise decision making processes, and can help ensure that the interventions or options selected to address health and social concerns are acceptable to stakeholders, feasible to implement and do not worsen inequities or access to rights. This paper discusses qualitative evidence and how it can be used to inform decisions, especially in the Brazilian context. We present several case studies showing how qualitative evidence can inform policy development for key health systems questions. We then discuss what needs to be done in Brazil and the region to strengthen the use of qualitative evidence in decision making.

Introduction

In many settings, we have seen rapid growth of interest in how to strengthen the use of research evidence in decision-making processes. A wide range of initiatives in this area have now been implemented[1,2], involving policymakers and other stakeholders at international, national and subnational levels. These initiatives have been accompanied by growth in the number of publications on knowledge translation[1,3,4]. This approach to using research evidence – known widely as ‘evidence informed policy making’ – is based on knowledge translation processes and platforms that promote the use of systematic and transparent methods and also adopt effective communication, participatory approaches, and the use of both global and local evidence to inform decisions[5].

Evidence informed policy making has perhaps become mostly widely institutionalised within the health sector. However, the approach has gradually been taken up within other areas of social policy, building on the learning gained over the years within the health sector and helping to expand the vision of the wider use of evidence to inform policy-making processes for social and other systems[6-8].

Implementing evidence informed policy making is a significant challenge because the process of contextualising and adapting evidence on interventions or options needs to take into account the views, experiences and interests of stakeholders. Putting such participatory approaches into practice within health and social care systems is complex because of the range of potential stakeholders, the need to manage many

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different interests, the need for skilled facilitation and the time required. We have suggested elsewhere that qualitative evidence can play a critical role in evidence informed policy making because such evidence can capture the views and experiences of those affected by an intervention or option. Such evidence can complement the views of stakeholders participating directly in policy processes and can help to answer questions – such as the acceptability and feasibility of interventions – that are generally addressed poorly within the quantitative comparative studies used to assess the effects of clinical or policy interventions[9-11].

Qualitative evidence, then, has the potential to humanise decision processes and help ensure that the interventions or options selected to address health and social concerns are acceptable to stakeholders, feasible to implement and do not worsen inequities or access to rights. However, such evidence is not yet systematically used in policy processes in most settings. Awareness of this gap has led a number of research groups to develop methods to support the wider use of qualitative evidence in decision making[12-16]. Many groups are also making efforts to build an environment that is open to the use of this evidence to inform decision for different policy questions.

In this commentary, we introduce the concept of 'qualitative evidence' and discuss how it can be used to inform decisions in Brazil. We do this through presenting several case studies showing how qualitative evidence might be used to inform policy development for key health systems questions. We then discuss what needs to be done by stakeholders in Brazil and the region to strengthen the use of qualitative evidence in decision making.

Evidence informed health policy making in Brazil

In recent years, the institutionalization of systematic and transparent processes for the development of clinical practice guidelines has grown in Brazil, especially in relation to the management of new health technologies by the Ministry of Health[17,18]. Currently, the Ministry of Health uses the GRADE approach to guide the development of new guidelines related to prioritised health technologies and other complex care processes, such as normal childbirth and the management of chronic diseases[18,19].

Alongside this, the issue of how to implement evidence-informed clinical practice guidelines is receiving increasing attention in Brazil. There is particular interest in the barriers and facilitators to implementation, such as the accessibility of guideline recommendations; how easily these can be applied; the extent to which there is ownership of a guideline by health care providers and other stakeholders; and the organizational environment, including the extent of institutional support and human resources.

In Brazil, we and others have been involved in a number of recent initiatives to build capacity for adopting a systematic and transparent approach to including qualitative evidence as inputs to decision-making. These activities include training in qualitative evidence synthesis methods and in applying the GRADE-CERQual approach for assessing how much confidence to place in evidence from reviews of qualitative research[20]. We have also translated and disseminated key materials needed for capacity strengthening (for example,[21,22]. Although stakeholders' views of the value of qualitative evidence for informing decision-making processes is still very varied across the country, there is growing institutional and academic interest in the principles and methods for incorporating this type of evidence. We would argue that this shows a demand for capacity strengthening and sharing of experiences in this area.

What is qualitative evidence?

Qualitative research aims to describe the social world and to explore people’s perceptions and experiences of the social world. This includes people’s perceptions and experiences of health and illness, health and social care services, institutions, the built environment and other aspects of the social world. Qualitative research also aims to explain and interpret the social world by developing hypotheses, theories or models[23]. Primary qualitative research commonly uses data collection methods such as focus group discussions, individual interviews, observations and document analysis. Qualitative studies use a large number of different data analysis approaches, but many involve analyzing text (such as interview transcripts and documents) to identify key themes or categories.

Qualitative evidence is empirical data drawn from research that uses qualitative methods of data collection and analysis. Qualitative evidence includes evidence emerging from individual primary qualitative studies as well as evidence from the findings of qualitative evidence syntheses (sometimes called systematic reviews of primary qualitative studies – a form of secondary research). Individual’s opinions and other similar types of anecdotal information are not, in themselves, considered to be qualitative evidence as they are not based on the systematic data collection and analysis that is an integral part of a research process. Quantitative data that is expressed in words is also not considered to be qualitative evidence. As with other types of evidence, tools are available to assess the methodological limitations and the trustworthiness of qualitative evidence. Such assessments are often part of preparing such evidence for use in decision making – for example, the GRADE-CERQual approach can be used to assess how much confidence to place in findings from qualitative evidence syntheses[14,20].

In some settings, evidence from qualitative evidence syntheses now feeds into decision-making processes, alongside evidence on the effects of different interventions or options and evidence on resource use or costs[10]. For instance, qualitative evidence is being used to develop clinical guideline and health systems guidance[12,13,15]. Qualitative evidence is also used increasingly in decision support tools such as GRADE evidence-to-decision frameworks[15,24,25], evidence briefs for policy making[26] and health technology assessments[27,28]. As we have noted elsewhere[11,15], for qualitative evidence to inform decision making for health and social care, it needs to move through the elements of the *evidence ecosystem* (Figure 1). Like other types of evidence, qualitative evidence needs to move

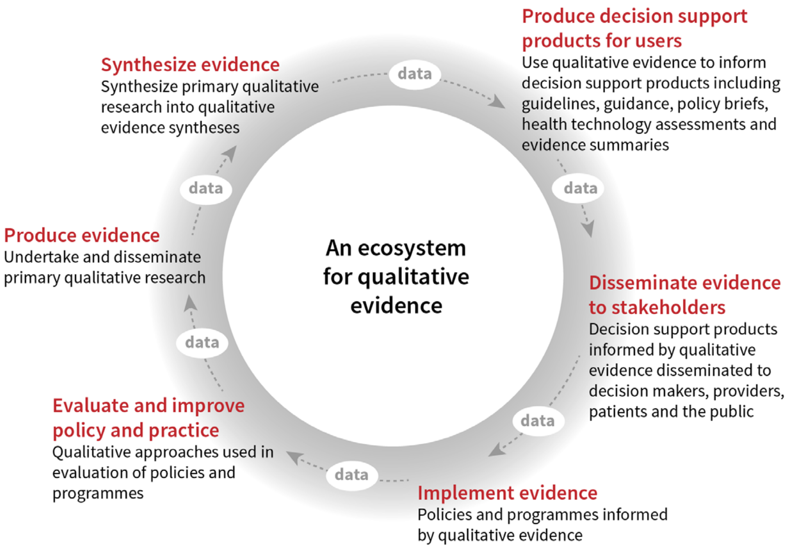


Figure 1: The ecosystem for qualitative evidence (adapted from [15])

seamlessly between *evidence producers* (those undertaking primary qualitative research); *evidence synthesizers* (those conducting qualitative evidence syntheses); *evidence processers and disseminators* (those producing evidence-informed decision products such as guidelines and HTAs); and *evidence implementers* (those responsible for implementing evidence-informed decisions, such as programme managers, and those involved in delivering and using health services such as service providers, and the public)[11,29]. The connections between each stage of the ecosystem are critical to ensuring the flow of evidence and its translation into decisions and actions to improve policy and practice[30]. This conceptual model is helpful in considering where there are bottlenecks in relation to evidence production and demand, and in identifying what might be needed to address these.

Case studies of how qualitative evidence might be used to inform decisions

Chronic diseases guidelines in Brazil – the case of hypertension

In Brazil, clinical practice guidelines are available on pharmacological treatments for the most prevalent non-communicable diseases, including arterial hypertension. According to a recent study, the methodological rigor and transparency of these guidelines are poor. Using the AGREE II tool (Appraisal of Guidelines Research and Evaluation instrument, version II), the evaluation assessed sixteen of the 26 included clinical practice guidelines as low quality, and none were rated as high quality[31]. No clinical practice guideline was recommended for implementation within health services without modification (77% were not recommended at all). The study also showed that guideline development did not consider patients' preferences, and that insufficient information was provided regarding a number of other critical steps in the guideline process. The evaluation concluded that Brazilian healthcare professionals should be concerned about the quality of national clinical practice guidelines for the treatment of selected non-communicable diseases.

These results demonstrate the need to improve the process of developing guidelines in Brazil, including through the use of relevant qualitative evidence. An illustrative example is provided by the 'Cadernos de Atenção Básica' - a collection of primary care guidelines that outlines the strategies recommended by the Brazilian Ministry of Health for care of people with arterial hypertension[32]. A strength of these guidelines is that they used the GRADE approach for the assessing the strength of recommendations[33]. However, the guidelines do not appear to have considered qualitative evidence. Including such evidence could have helped to ensure that the discussions of the guideline group were informed by the experiences of patients and health care providers. Such evidence could also have contributed to prioritizing the questions to be addressed by the guideline, to defining their scope and to assessing the acceptability and feasibility of the clinical interventions considered[10,12,13,15]. Qualitative evidence could also have helped to identify barriers to implementing the guideline recommendations in primary care. For instance, when discussing how care should be implemented, the guideline simply notes that this should be informed by the person's needs and degree of risk, as well as her capacity for adherence and motivation for self-care (p42). However, our quick search of the Epistemonikos database of systematic reviews (www.epistemonikos.org) identified two qualitative evidence syntheses relevant to the care of people with hypertension[34,35]. Guideline developers could have drawn on these, or could have used their findings as the starting point for an updated synthesis to inform the guideline and help close the ‘know-do’ gap.

The growth of digital interventions for health systems strengthening - telemedicine in Brazil

The World Health Organization (WHO) recently released evidence-informed recommendations on how countries can use digital interventions, accessible via cell phones and tablets, to strengthen health systems and improve essential services and people’s health[36]. In this guideline, the WHO recommends the use of client-to-provider telemedicine to complement face-to-face delivery of health services and also recommends the use of provider-to-provider telemedicine. Both of these interventions are recommended in settings where patient safety, privacy, traceability, accountability and security can be monitored.

In Brazil, telemedicine is seen as an important tool for addressing key health system challenges. These include ensuring diagnostic coverage to support primary healthcare in isolated regions and improving the quality and performance of basic health services in cities. A study focusing on scaling up telemedicine in the country highlighted that these technologies constitute a strategic area for innovation and for including technological advances from other areas[37]. The study suggests that digital interventions for health can help democratize access to health services by integrating remote regions with central health services. However, digital strategies in Brazil should not be implemented in ways that ignore the many differences between regions and population groups, particularly since these disparities may impact on people’s access to quality health services.

We would argue that qualitative evidence can play an important role in planning the implementation of digital interventions for health systems strengthening and can help to ensure that these implementation strategies do not widen disparities. A telemedicine programme implemented in a remote area in the northern Brazilian state of Rondônia, and intended to promote the inclusion of hard-to-reach communities, provides an interesting example in this regard[38]. In this programme, telemedicine was used to provide communities with access to information, medical care and lectures on health promotion and prevention. The project showed that telemedicine can improve access to health services, including for more isolated or marginalised populations. Those describing the programme suggest that this, in turn, facilitated the social reintegration of groups who were disadvantaged by geographic isolation and also helped to disseminate key health information[38]. Within such programmes, qualitative evidence could be used to identify factors affecting the implementation of the interventions and to support the development of implementation guides for the project. Relevant evidence is available from a qualitative evidence synthesis conducted for the recent WHO digital interventions guideline[36,39]. This synthesis explored healthcare providers’ perceptions and experiences regarding their use of digital technologies for health, and included findings on disadvantaged or hard-to-reach groups.

HPV vaccination and vaccine hesitancy in Brazil

Brazil’s National Immunization Programme, which includes both children and adolescents, is widely viewed as a success. The HPV (Human papillomavirus) vaccination programme was initiated by the Brazilian Ministry of Health in 2014 for female adolescents aged nine to 14 years, and extended in 2017 to male adolescents aged 11 to 14 years. As with many other HPV vaccination programmes, vaccine hesitancy is an issue for the programme in Brazil. Vaccine hesitancy has been defined as a delay in accepting, or refusing, the recommended vaccines when they are available in health services, a phenomenon that varies over time, the location and types of vaccines[40,41]. While vaccine hesitancy is not a new problem in Brazil or the region, it is an increasingly important concern for Brazilian immunization managers and researchers. Implementation of the HPV vaccine campaign by the Ministry of Health led to a number of controversies in the press and other media. Official government communication on the programme focused on the advantages and risks of the vaccine and on vaccination procedures. However, the media raised issues regarding the social and ethical risks and implications of HPV vaccination, including people's values regarding this intervention. Government communication did not respond actively these public concerns in the first phase of HPV vaccination (which focused on the first vaccine dose). Later, the Ministry of Health attempted to respond to the concerns through a new communication strategy, but without considerable success. Reports of adverse effects emerged most prominently during the second phase, which focused on the second vaccine dose for all those vaccinated in the first phase. These reports contributed to reducing vaccination coverage[42].

How best to communicate with the public about HPV vaccination is therefore a key concern in the Brazilian context. Qualitative evidence has the potential to inform the implementation of communication strategies to address key vaccination barriers that have been identified through local research[43,44]. However, a recently published systematic scoping overview of systematic reviews focusing on communication with adolescents, parents and other stakeholders around HPV vaccination for adolescents highlights the small number of relevant studies from LMICs[45]. In this case, then, primary qualitative research would be needed to support the development of relevant communication strategies for Brazil.

Strengthening the use of qualitative evidence in decision making in Brazil and the region

As part of a broader approach to improving how organisations support the use of research evidence to inform policymaking[46], the following steps might help to strengthen organisational capacity to use of qualitative evidence in particular (Table 1):

1. Assess the organisation's capacity to use qualitative evidence to inform decision making: Capacity to interpret and use qualitative evidence varies widely across organisations, and is likely to be low in many places as the systematic use of qualitative evidence in decision making is relatively new. A key step in strengthening capacity is to assess existing capacities, for example for finding and interpreting qualitative evidence in relation to policy questions.

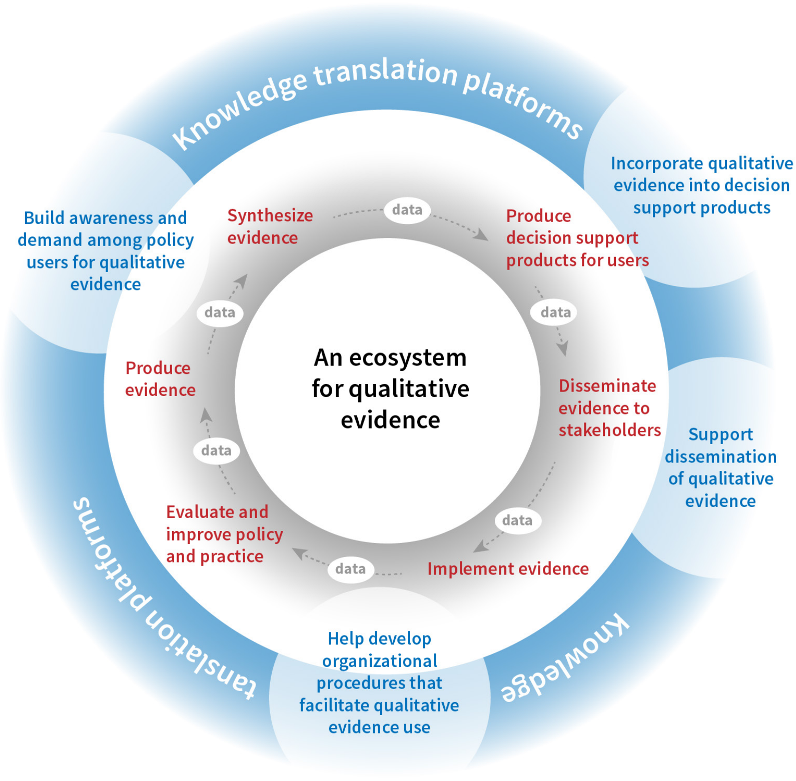
Table 1: Steps to strengthen organisational capacity to use qualitative evidence

1. Assess the organisation's capacity to use qualitative evidence to inform decision making
2. Strengthen capacity in existing knowledge translation platforms to find, appraise and use qualitative evidence
3. Increase awareness of what a qualitative evidence synthesis is, why it is a valid and trustworthy approach and how the findings can be used in decision processes
4. Develop priority setting mechanisms for qualitative research, building on and informed by knowledge gaps identified through qualitative evidence syntheses, public consultations and decision processes

2. Strengthen capacity in existing knowledge translation platforms – collaborations among decision makers, evidence producers and synthesizers, civil society representatives, evidence implementers and other stakeholders to implement evidence informed policy making[47] – to find, appraise and use qualitative evidence:

- Knowledge translation platforms can play a key role in developing and supporting an ecosystem for qualitative evidence in the region. For instance, these platforms can help to build demand among policy users for qualitative evidence; can incorporate qualitative evidence into decision support products such as evidence briefs for policy and guidelines; and can disseminate these products to evidence implementers and other stakeholders (Figure 2). This requires that these platforms build their capacities in at least two areas. Firstly, they need to strengthen their capacity to *find, undertake and use* qualitative evidence syntheses, including undertaking assessments of confidence in the evidence, using approaches such a GRADE-CERQual[14]. Secondly, platforms need to strengthen their capacity to *package* qualitative evidence for decision making, including making this evidence accessible using layered entry formats and plain language[13,15].
- Routinising the use of qualitative evidence may require changes in organisational culture in relation to evidence use. This may include strengthening capacity to interpret this evidence and putting in place organisational procedures to support the use of this evidence in decisions. Where appropriate, knowledge translation platforms could build qualitative evidence into their standard operating procedures for responding to requests for evidence briefs for policy making, guidelines, health systems guidance and other knowledge translation products. This can help to ensure

Figure 2. The ecosystem for qualitative evidence and how knowledge translation platforms can develop and support this (adapted from[15])



that systematic and transparent methods are used, that gaps in capacity are identified and that methods are improved over time.

- Strengthening capacity to interpret and use qualitative evidence needs to extend to the full range of stakeholders, including civil society representatives who may be involved in decision processes. Including qualitative evidence in decision processes may increase upfront costs and the time needed to produce decision support products. However, the use of qualitative evidence has the potential to substantially increase the appropriateness of selected policy options and the likelihood of successful implementation. This is because such evidence can identify contextual and other factors that may affect the acceptability and feasibility of policy options and their implementation. Over time, these benefits are likely to outweigh the upfront costs involved.

3. Increase awareness of what a qualitative evidence synthesis is, why it is a valid and trustworthy approach and how the findings can be used in decision processes: Qualitative evidence synthesis is a key method for bringing together the best available qualitative evidence on a topic, in order to inform decision making. Efforts are needed to raise awareness and build demand among policy users and other stakeholders for this product. Most policy users want information on the acceptability and feasibility of policy options, and on implementation considerations, but may not be aware of the role that findings from qualitative evidence syntheses can play in providing such evidence.
4. Develop priority setting mechanisms for qualitative research, building on and informed by knowledge gaps identified through qualitative evidence syntheses, public consultations and decision processes. This will also help to reduce qualitative research waste[48-50].

Strengthening the use of qualitative evidence for decision making also raises a number of challenges. Firstly, there is limited capacity globally and in the region to undertake qualitative evidence syntheses, to assess confidence in the findings from these syntheses and, more broadly, to support the use of qualitative evidence in decision making. Collaboration across organisations, geographies and sectors may help to address this, and structures such as the Global Evidence Synthesis Initiative (GESI: www.gesiinitiative.com) and EVIPNet (the Evidence Informed Policy Network: www.who.int/evidence/en/) can play an important role in bringing together evidence producers, users and other stakeholders. Secondly, methods for packaging qualitative evidence for use in decision making are still evolving[9-13,15,51,52] and work is needed to understand how best to do this for different types of questions and decision support products. Qualitative evidence needs to be presented in ways that are accessible while not oversimplifying and stripping away key aspects of context and theory that are important to interpreting this evidence[53]. Thirdly, efforts are needed to build the capacity of policy users and other stakeholders to interpret and use qualitative evidence. These efforts could build on existing initiatives in many settings to promote and support evidence-informed policymaking. We believe that local, national and global collaboration are critical to ensuring that qualitative evidence is used more widely to inform decision making in health and other sectors.

Conclusion

The wider use of qualitative evidence has the potential to humanise decision making for health and other sectors. Qualitative evidence may also help us to achieve the health and other Sustainable Development Goals in Brazil and in other countries. We therefore invite decision makers, researchers, knowledge brokers and other stakeholders to actively seek opportunities in their settings to increase the use of qualitative evidence and to build capacity for this.

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References

1. Oliver K, Innvar S, Lorenc T, Woodman J, Thomas J: A systematic review of barriers to and facilitators of the use of evidence by policymakers. BMC Health Serv Res 2914, 14(2).
2. WHO: World report on knowledge for better health. Geneva: World Health Organization; 2004.
3. Lavis JN, Lomas J, Hamid M, Sewankambo NK: Assessing country-level efforts to link research to action. Bull World Health Organ 2006, 84(8):620-628.
4. Pantoja T, Barreto J, Panisset U: Improving public health and health systems through evidence informed policy in the Americas. Bmj 2018, 362(k2469).
5. Oxman AD, Lavis JN, Lewin S, Fretheim A: SUPPORT Tools for evidence-informed health Policymaking (STP) 1: What is evidence-informed policymaking? Health research policy and systems / BioMed Central 2009, 7 Suppl 1:S1.
6. Cook C, Nichols S, Webb J, Fuller R, Richards R: Simplifying the selection of evidence synthesis methods to inform environmental decisions: A guide for decision makers and scientists. Biological Conservation 2017, 213(A):135-145.
7. Godfrey D, Brown C: How effective is the research and development ecosystem for England's schools? London Review of Education 2018, 16(1):136-151.
8. Stewart R: Using evidence in international development. In: What Works Now?: Evidence-Informed Policy and Practice. 1st edn. Edited by Boaz A, Davies H, Fraser A, Nutley S. United Kingdom: Policy Press; 2019.
9. Glenton C, Lewin S, Gulmezoglu AM: Expanding the evidence base for global recommendations on health systems: strengths and challenges of the OptimizeMNH guidance process. Implement Sci 2016, 11:98.
10. Glenton C, Lewin S, Norris SL: Using evidence from qualitative research to develop WHO guidelines (Chapter 15). In: WHO Handbook for Guideline Development. 2nd edn. Edited by World Health Organization. Geneva: WHO; 2016.
11. Lewin S, Glenton C: Are we entering a new era for qualitative research? Using qualitative evidence to support guidance and guideline development by the World Health Organization. Int J Equity Health 2018, 17(1):126.
12. Downe S, Finlayson K, Lawrie TA, Lewin S, Glenton C, Rosenbaum S, Barreix M, Tunçalp Ö: Qualitative Evidence Synthesis (QES) for guidelines: Paper 1 - Using qualitative evidence synthesis to inform guideline scope and develop qualitative findings statements. Health Research Policy and Systems 2019, 17(76).
13. Glenton C, Lewin S, Lawrie TA, Barreix M, Downe S, Finlayson K, Tamrat T, Rosenbaum S, Tunçalp Ö: Qualitative Evidence Synthesis (QES) for guidelines: Paper 3 - Using qualitative evidence syntheses to develop implementation considerations and inform implementation processes. Health Research Policy and Systems 2019, 17(74).
14. Lewin S, Booth A, Glenton C, Munthe-Kaas HM, Rashidian A, Wainwright M, Bohren MA, Tunçalp Ö, Colvin CJ, Garside R et al: Applying GRADE-CERQual to qualitative evidence synthesis findings: introduction to the series. Implementation Science 2018, 13(Suppl 1):2.
15. Lewin S, Glenton C, Lawrie T, Downe S, Finlayson K, Rosenbaum S, Barriex M, Tuncalp Ö: Qualitative Evidence Synthesis (QES) for Guidelines: Paper 2 - Using qualitative evidence synthesis findings to inform

evidence-to-decision frameworks and recommendations. Health Research Policy and Systems 2019, 17(75).

16. Noyes J, Booth A, Cargo M, Flemming K, Garside R, Hannes K, Harden A, Harris J, Lewin S, Pantoja T et al: Cochrane Qualitative and Implementation Methods Group guidance series-paper 1: introduction. Journal of clinical epidemiology 2018, 97:35-38.
17. Comissão Nacional de Incorporação de Tecnologias no Sistema Único de Saúde (CONITEC): Manual de elaboração de Diretrizes Clínicas irá preencher lacunas sobre as melhores práticas na elaboração de diretrizes em saúde CONITEC, Ministério da Saúde; 2016. Available at: http://conitec.gov.br/manual-de-elaboracao-de-diretrizes-clinicas-ira-preencher-lacunas-sobre-as-melhores-praticas-na-elaboracao-de-diretrizes-em-saude.
18. Ministério da Saúde: Diretrizes metodológicas: elaboração de diretrizes clínicas. Brasilia: Ministério da Saúde, Secretaria de Ciência, Tecnologia e Insumos Estratégicos, Gestão e Incorporação de Tecnologias em Saúde; 2016. Available at: http://conitec.gov.br/images/Relatorios/2016/DiretrizMetodologica.pdf.
19. Ministério da Saúde: Diretriz Nacional de Assistência ao Parto Normal. Protocolo. Brasilia: Comissão Nacional de Incorporação de Tecnologias no Sistema Único de Saúde (CONITEC), Ministério da Saúde; 2016. Available at: http://conitec.gov.br/images/Consultas/2016/Relatorio_Diretriz-PartoNormal_CP.pdf.
20. Lewin S, Glenton C, Munthe-Kaas H, Carlsen B, Colvin CJ, Gulmezoglu M, Noyes J, Booth A, Garside R, Rashidian A: Using qualitative evidence in decision making for health and social interventions: an approach to assess confidence in findings from qualitative evidence syntheses (GRADE-CERQual). PLoS Med 2015, 12(10):e1001895.
21. Glenton C, Lewin S, Norris S: 15. Usando evidências de pesquisa qualitativa para desenvolver diretrizes da OMS. In: WHO Handbook for Guideline Development. 2nd edn. Edited by World Health Organization. Geneva: WHO; 2016.
22. Lewin S, Glenton C, Munte-Kaas H, Carlsen B, Colvin C, Gülmezoglu M, Noyes J, Booth A, Garside R, Rashidian A: Uso de Evidência Qualitativa em Decisões para Intervenções Sociais e em Saúde: Uma Abordagem para Avaliar a Confiança em Achados de Sínteses de Evidências Qualitativas (GRADE-CERQual). PLOS Medicine 2015, 12(10):e1001985. Portuguese translation by Maria Sharmila A. Sousa e Ananyr P. Fajardo. Available at: http://bit.do/e1001983SPt
23. Green J, Thorogood N: Qualitative methods for health research, Fourth edn. London: Sage; 2018.
24. Alonso-Coello P, Schünemann H, Moberg J, Brignardello-Petersen R, Akl E, Davoli M, Treweek S: GRADE Evidence to Decision (EtD) frameworks: 1. A systematic and transparent approach to making well-informed healthcare choices: 1. Introduction. Bmj 2015.
25. Moberg J, Oxman AD, Rosenbaum S, Schunemann HJ, Guyatt G, Flottorp S, Glenton C, Lewin S, Morelli A, Rada G et al: The GRADE Evidence to Decision (EtD) framework for health system and public health decisions. Health research policy and systems / BioMed Central 2018, 16(1):45.
26. Lavis JN, Oxman AD, Grimshaw J, Johansen M, Boyko JA, Lewin S, Fretheim A: SUPPORT Tools for evidence-informed health Policymaking (STP) 7: Finding systematic reviews. Health research policy and systems / BioMed Central 2009, 7 Suppl 1:S7.
27. Booth A, Noyes J, Flemming K, Gerhardus A, Wahlster P, Van Der Wilt GJ, Mozygemba K, Refolo P, Sacchini D, Tummers M et al: Guidance on choosing qualitative evidence synthesis methods for use in health technology assessments of complex interventions. In. Available from: http://www.integrate-hta.eu/downloads/ 2016.
28. Ring N, Jepson R, Ritchie K: Methods of synthesizing qualitative research studies for health technology assessment. International journal of technology assessment in health care 2011, 27(4):384-390.
29. MAGIC Project: The Evidence Ecosystem: Personalised eHealth Solutions. In. Available from: http://magicproject.org/research-and-tools/the-evidence-ecosystem/. Accessed 10 July 2019; 2019.
30. Shepherd JP: How to achieve more effective services: the evidence ecosystem. In. Cardiff, UK: What Works Network/Cardiff University; 2014. Available at: http://www.scie-socialcareonline.org.uk/how-to-achieve-more-effective-services-the-evidence-ecosystem/r/a11G0000006z7vXIAQ.
31. Molino CG, Romano-Lieber NS, Ribeiro E, de Melo DO: Non-Communicable Disease Clinical Practice Guidelines in Brazil: A Systematic Assessment of Methodological Quality and Transparency. PLoS One 2016, 11(11):e0166367.
32. Ministério da Saúde: Estratégias para o cuidado da pessoa com doença crônica: hipertensão arterial sistêmica. Brasilia: Ministério da Saúde, Secretaria de Atenção à Saúde, Departamento de Atenção Básica; 2013.

Available at http://bvsms.saude.gov.br/bvs/publicacoes/estrategias_cuidado_pessoa_doenca_cronica.pdf.

33. Guyatt GH, Oxman AD, Vist GE, Kunz R, Falck-Ytter Y, Alonso-Coello P, Schunemann HJ, Group GW: GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. BMJ, 2008, 336(7650):924-926.
34. Khatib R, Schwalm JD, Yusuf S, Haynes RB, McKee M, Khan M, Nieuwlaat R: Patient and healthcare provider barriers to hypertension awareness, treatment and follow up: a systematic review and meta-analysis of qualitative and quantitative studies. PLoS One 2014, 9(1):e84238.
35. Marshall IJ, Wolfe CD, McKevitt C: Lay perspectives on hypertension and drug adherence: systematic review of qualitative research. Bmj 2012, 345:e3953.
36. WHO: WHO guideline: recommendations on digital interventions for health system strengthening. Geneva, Switzerland: World Health Organization; 2019.
37. Maldonado JM, Marques AB, Cruz A: Telemedicine: challenges to dissemination in Brazil. Cad Saude Publica 2016, 32(Suppl 2):e00155615.
38. Machado F, Pinto de Carvalhol M, Mataresi A, Mendonça E, Cardosol L, Yogi M, Rigato H, Salazar M: Utilização da telemedicina como estratégia de promoção de saúde em comunidades ribeirinhas da Amazônia: experiência de trabalho interdisciplinar, integrando as diretrizes do SUS. Ciência & Saúde Coletiva 2010, 15(1):247-254.
39. Odendaal WA, Goudge J, Griffiths F, Tomlinson M, Leon N, Daniels K: Healthcare workers' perceptions and experience on using mHealth technologies to deliver primary healthcare services: qualitative evidence synthesis (Protocol). Cochrane Database of Systematic Reviews 2015(11):Art. No.: CD011942.
40. Larson HJ, Jarrett C, Eckersberger E, Smith DM, Paterson P: Understanding vaccine hesitancy around vaccines and vaccination from a global perspective: a systematic review of published literature, 2007-2012. Vaccine 2014, 32(19):2150-2159.
41. MacDonald NE, Sage Working Group on Vaccine Hesitancy: Vaccine hesitancy: Definition, scope and determinants. Vaccine 2015, 33(34):4161-4164.
42. Poerschke de Quevedo J, Myrrena I, Moro Wieczorkievicz A, Invernizzi N: A política de vacinação contra o HPV no Brasil: a comunicação pública oficial e midiática face à emergência de controvérsias. Revista Tecnologia e Sociedade 2016, 12(24):1-26.
43. Kops N, Hohenberger G, Bessel M, Correia Horvath J, Domingues C, Kalume Maranhão A, Alves de Souza F, Benzaken A, Pereira G, Wendland E: Knowledge about HPV and vaccination among young adult men and women: Results of a national survey. Papillomavirus Res 2019, 7:123-128.
44. Mendes Lobao W, Duarte FG, Burns JD, de Souza Teles Santos CA, Chagas de Almeida MC, Reingold A, Duarte Moreira EJ: Low coverage of HPV vaccination in the national immunization programme in Brazil: Parental vaccine refusal or barriers in health-service based vaccine delivery? PLoS One 2018, 13(11):e0206726.
45. Foss H, Oldervoll A, Fretheim A, Glenton C, Lewin S: Communication around HPV vaccination for adolescents in low- and middle-income countries: a systematic scoping overview of systematic reviews. Systematic Reviews 2019, 8(190).
46. Oxman AD, Vandvik PO, Lavis JN, Fretheim A, Lewin S: SUPPORT Tools for evidence-informed health Policymaking (STP) 2: Improving how your organisation supports the use of research evidence to inform policymaking. Health research policy and systems / BioMed Central 2009, 7 Suppl 1:S2.
47. Ongolo-Zogo P, Lavis JN, Tomson G, Sewankambo NK: Assessing the influence of knowledge translation platforms on health system policy processes to achieve the health millennium development goals in Cameroon and Uganda: a comparative case study. Health Policy Plan 2018, 33(4):539-554.
48. Chalmers I, Glasziou P: Avoidable waste in the production and reporting of research evidence. Lancet 2009, 374(9683):86-89.
49. Chalmers I, Glasziou P: Systematic reviews and research waste. Lancet 2016, 387(10014):122-123.
50. Macleod MR, Michie S, Roberts I, Dirnagl U, Chalmers I, Ioannidis JP, Al-Shahi Salman R, Chan AW, Glasziou P: Biomedical research: increasing value, reducing waste. Lancet 2014, 383(9912):101-104.
51. Lewin S, Glenton C: Using evidence synthesis in the development of health systems guidance. In: Evidence synthesis for health policy and systems: a methods reader. Edited by Langlois EV, Daniels K, Akl E. Geneva: World Health Organization; 2018.
52. Carroll C: Qualitative evidence synthesis to improve implementation of clinical guidelines. Bmj 2017, 356:j80.
53. Britten N, Garside R, Pope C, Frost J, Cooper C: Asking More of Qualitative Synthesis: A Response to Sally Thorne. Qualitative health research 2017, 27(9):1370-1376.

Scoping review: potential for the synthesis of methodologies used in qualitative primary research

Luciana Cordeiro[I], *Cassia Baldini Soares*[II]

Abstract

Introduction: Scoping reviews have global expression in the health area; they are meant to scrutinize types of research, how and by whom they were performed among other unknown categories. **Objectives:** To describe the process of developing a scoping review of a methodological nature that gathered qualitative primary studies; and to analyze particularities of this type of review. **Method:** Experience report, which was referenced in the PRISMA-ScR review report guide, describing the empirical lessons learned in the development of a scoping review which mapped the use of action research in health. **Results:** The PCC mnemonic was adapted, being the concept C decomposed into participation, knowledge production and transformation of practices, the three principles of action research. These principles were included as categories in the instrument of data extraction, whose elaboration is one of the major challenges in this type of review. The review mapped "how" evidence from action research are produced and proved potent to systematically expose the theoretical-methodological frameworks of the primary studies examined, from rigorous application of the steps. **Conclusion:** The PRISMA-ScR extension was detailed and aligned with the guide of the Joanna Briggs Institute, constituting a fundamental instrument for the transparency and reliability of this type of review.

Keywords: Systematic review; Scoping review; Methodological studies

Introduction

Since the 1990s, evidence-based practice has become an important tool for clinical, programmatic, and policy decision-making. With the increase in production of and search for systematic knowledge, there has been an increase in the publication of literature reviews. A broader recognition of different forms of research evidence, aims and questions has favoured the development of various approaches to systematization of literature.[1-4] In public health policy, for instance, it is notorious the move towards the development of comprehensive methodologies such as realist review,[5] and syntheses guided by the EVIPNet (Evidence-Informed Policy Network).[6]

Scoping review[1-4] has gained international prominence in the field of evidence synthesis in health,[7] with a significant growth since 2012.[8] It aims to map literature in a specific field of interest, especially when reviews on a topic have not been published yet. Unlike systematic review, scoping review is appropriate to address broad topics, gather diverse study designs, and identify evidence that has been produced.[1] In other words, it is not intended to search for the best evidence on a health intervention or experience but to gather several types of evidence and show how they have been produced. It is not focused on assessing how robust an evidence is, but on tracing evidence and / or potentialities, in order to support researchers and, to some extent, health workers, managers, and policy makers.

The aims of scoping reviews have been described as: to examine the extent and nature of

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evidence production and / or to clarify conceps that underpin a given domain; to evaluate the relevance and feasibility of carrying out a systematic review, and, by doing so, to improve systematic review questions; to systematise and disseminate findings that may contribute to research, practice and policy; to identify gaps in existing literature; and to understand how a research is conducted in a certain area. In consequence, scoping review can help systematic review authors to examine emerging evidence from recently published or ongoing studies, as well as to examine how research is being conducted in well-established areas.[1,3,4,8,9]

Scoping review is also appropriate to examine studies for decision-making in theoretical-methodological field, by mapping theories and methodologies that may inform researchers. As in primary studies, the question guides the review methodology to be used. Moreover, it is necessary to consider if the question is related to either qualitative or quantitative dimension of healthcare, or if there are questions that can be complementary.

The adoption of a methodology as an object may make it easier to understand its use. It is advocated that the function of methodology is to integrate all the other elements of scientific inquiry, ensuring organicity and internal coherence of the research process. It is seen as the path taken for the development of a research, articulating in a coherent manner its design (aims and methods) to the theoretical and philosophical[III] assumptions.[10,11] Basically, for reality to be understood in order to answer the research question, it is needed an appropriate methodology, aligned with epistemological and theoretical assumptions, which also leads to a proper selection and application of methods of data analysis.[12]

Taking into account the above, Cordeiro and Soares[13] conducted a scoping review in accordance with Joanna Briggs Institute (JBI) methodology[3,4] to understand how action research[IV], which has multiplicity of designs and ways of operationalisation, has been used in the health area across the world. They categorised the studies included in three dimensions – organizational, individual and collective –, and concluded that the principles of action research were used in a variety of ways, according to the theoretical assumptions underlying primary studies.

Although it is expected that scoping reviews should be conducted in a systematic way according to literature and JBI guidelines,[3,4] with transparency and rigour in every step, a scoping review of scoping reviews has shown the need to improve reporting.[8] Therefore, the aim of this paper is to describe the process of conducting a scoping review of methodological nature that gathered qualitative primary studies; and to analyse particularities of this type of review, based on PRISMA-ScR (Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews). It is expected to contribute to the improvement of scoping reviews in general, and those mapping methodologies used in primary qualitative studies in particular.

Method

An experience report was developed to describe and explain the empirical lessons learned from the conduct of a scoping review

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[III] Philosophical foundations have already been extensively explored by a number of authors. See the following reference for further details: Cordeiro L. *Pesquisa-ação na área da saúde: uma proposta marxista a partir de revisão de escopo*. [thesis]. São Paulo: *Escola de Enfermagem da Universidade de São Paulo*, 2016.

[IV] Action research is a methodology of participatory research used in a diversity of knowledge fields, which seeks to transform problems encountered in reality through an educational process. The principles of action research are: effective participation of research participants, knowledge production, and transformation of practice, where the transformation process involves a spiral of steps, with no hierarchy or particular order of the steps.

of methodological nature addressing the use of action research in health.[13]

The report was based on PRISMA-ScR,[7] a checklist recently developed to guide the reporting of scoping reviews. It contains 22 items that are included across the required sections of a review report: Title, Abstract, Introduction, Methods, Results, Discussion, and Funding. Detailed information on the conduct of the scoping review, adaptations made, as well as prior knowledge to the review are provided below.

It is important to note that PRISMA-ScR was published after the scoping review that is addressed in this paper, meaning the scoping review and its experience report followed exclusively the JBI guidelines.[3,4]

Results and discussion

An analysis of the experience report development,[13] in accordance with PRISMA-ScR,[8] is presented below, highlighting the sections that were challenging because of the methodological nature of this scoping review.

For the **title**, PRISMA-ScR recommends to identify the report as a scoping review, and the JBI manual suggests to present the elements of PCC mnemonic (Population, Concept, and Context), which relates to the review question. Accordingly, the original title of the scoping review is: 'Action research in the healthcare field: a scoping review'; that is, P - empirical studies that used action research methodology, C - key categories of action research, and C - healthcare field, which indicate the inclusion criteria for primary studies. The first challenge was thus to adapt the mnemonic to a review of methodological nature.

In the **Introduction** section of a review protocol, as well as in its report, there should be relevant information regarding the topic and usefulness of the review, as a structured goal is necessary to conduct not only a scoping review, but reviews in general.

The analysis of the studies included in the scoping review of action research in healthcare indicated that they had approached practices with a wide range of purposes, and there was an apparent lack of uniformity in the use of the methodology, principles or philosophical foundation. Action research is an iterative, non-linear process[14] within a historical and cultural context, but in order to advance it in the healthcare field, in a coherent and rigorous manner, it is necessary to map and systematise its use.

In view of this, the following points were addressed in the Introduction:

1. The action research methodology backgrounds, i.e. its development and first applications.
2. Adaptations and generalisations made in order to inform on strengths and limitations of this methodology. At this point, possible relationships between researcher and subject should be taken into account, as they are important to decide on the methodology and data collection and analysis methods. Adaptations to and generalisations of a methodology are considered a scientific contribution only when there is compatibility between the philosophical assumptions and the path taken to investigate and understand the reality in question.
3. A clear distinction between the concepts of methodology and method, as the former refers to the theoretical-methodological framework and the path to conduct a research, articulating the research design in a coherent form (aims and methods), while the latter refers to a set of techniques, which is instrumental to the methodology.[15,16]
4. Geosocial contexts (continents, countries, local cultural specificities, geopolitical development and subordination, etc.) where the methodology has been applied. These data provide a preliminary picture of what to expect to find in primary studies,

helping with the development of a tool for data extraction, as well as with data categorisation and analysis.

As all scientific work, the aims of a scoping review should be well-defined and based on the research question. It is noteworthy to mention that, unlike the kind of questions that can be answered by systematic reviews, the questions of scoping reviews are, in general, broader in scope. It does not seek effect or association of risk, nor experience regarding a specific phenomenon, as such categories require closed-ended questions and inclusion of studies that are appropriate to answer them. In fact, they seek to understand what kind of research is being done, how it is being done, by whom and to whom, among other categories that may be of interest.

In scoping review of methodological nature, it may be applicable to adapt the PCC elements as mentioned above. Thus, the following review questions were addressed: How does participatory engagement work in action research in healthcare? Do the results of primary studies demonstrate both knowledge production and transformation of practice during the action research processes? The element ‘participant’ or ‘population’ was replaced by action research methodology; ‘context’ became the healthcare field; and ‘concept’ was divided into three categories: participative engagement or participation, knowledge production, and transformation of practice (i.e. the principles of action research).

The **aim** of the scoping review was to explore international literature related to the application of action research in healthcare.

Regarding the **method**, PRISMA-ScR recommends to inform if there is a review protocol, and whether it can be accessed. Although review protocols in healthcare can be registered in PROSPERO, to date, it is not possible to register scoping review in this database.[17] The JBI Database of Systematic Reviews and Implementation Reports publishes scoping review protocols, as is the case for this scoping review.[18]

With respect to **eligibility criteria, information sources, and search strategies**, the search was restricted to English, Spanish and Portuguese, but no time period restriction was applied. Several databases were searched, not only in the healthcare but also in other fields such as education, social sciences, and psychology. A specific search strategy was developed for each database by testing key words and controlled vocabulary terms. In view of the large number of publications in journals, grey literature was not searched. A record of the search strategies used (dates, number of studies included, etc.), and also the reasons for exclusion of studies during data extraction were provided in the appendix of the review report.

In the development of the scoping review protocol, the design of a data extraction form has been a major challenge. Because of its singular nature, every scoping review seeks to identify novel categories, unlike the traditional categories of systematic reviews (e.g. effect and experiences). It is necessary then, to decide what data to extract from primary studies, in accordance with the review question, besides common data such as author, title, year of publication, as specified by JBI manuals.

Thus, a data extraction form was developed based on the review specific objectives, consisting of several items, which were selected after some preliminary tests. An important part of this process was to understand that extraction of some data would require analytic analysis so they could be categorised. For example, action research steps were not often well described in the method section of primary studies. To capture such information, it was necessary to explore the description of the research process in the results or discussion section. Likewise, the dimensions of the studies – relevant data that led the review discussion – were analysed and the studies were classified according to their epistemological assumptions.

This data extraction process required knowledge of both epistemological domains and the methodology, including possible adaptations to it, in order to be able to develop the data extraction form, as well as to conduct the selection of studies for inclusion. For example, there were studies that reported the use of action research, but then described clinical follow-up. Such studies were excluded after being examined by two reviewers. While one of the key steps in a systematic review is the assessment of methodological quality for eligible studies, based on a standard tool, the critical appraisal of this scoping review focused on trustworthiness, accuracy and rigour in the methodology use in all primary studies included. According to PRISMA-ScR[8], the assessment of methodological quality of primary studies included based on a standard tool is not a requirement, but on occasion this may be the case.

According to JBI[3,4] and PRISMA-ScR[8] guidelines, the **results** of the searches and the number of studies included were presented as a flow diagram; the reasons for exclusion of studies after full text analysis were reported; and the results were expressed in a narrative format and presented in tables. In view of its importance for the discussion, the participation category and its classification were presented in two different tables, so trends in types of participation across the continents could be examined. In addition, the diverse categories were cross-linked and presented in a table. These tables were of great value in presenting the findings of more than a hundred studies included.

In the **discussion**, the results were summarised based on the research questions and objectives. It was taken into consideration some propositions not confirmed in the scoping review, as well as limitations of this type of review process, as recommended by both JBI[3,4] and PRISMA-ScR.[8]

Conclusion

The scoping review of methodological nature mapped ‘how’ evidence from action research are produced, and was effective in addressing the theoretical-methodological frameworks of the primary studies included in a systematic, transparent and rigorous way.

Its conduct was an exhaustive process of analysis, beginning with a well-formulated question, paying special attention to the development of the data extraction form as well to the summarization of the results, different from other types of systematic review that use standard tools. An improper use of action research methodology in primary studies was a major challenge in the development of this scoping review and its report.

The experience report included the first issues to consider when planning a scoping review of methodological nature. Focusing on the methodology as a path for connecting epistemological and theoretical assumptions, and data collection and analysis methods is crutial. Also, it is important to be well acquainted with the methodology’s approaches and backgrounds in order to be able to understand their implications in primary studies. Finally, it recommended a detailed analysis of a sample of studies included in a review, as this may help in the development of the data collection tool, which should be consistent with the review question.

Systematic reviews in healthcare aim to find the best available evidence on an intervention, programme or policy, and, therefore, their results should be graded for confidence. Scoping review is intended to map evidence, in such a way that confidence is basically related to the process and indicative of the improvement in the area.

The application of PRISMA extension for scoping reviews (PRISMA-ScR) was positive, aligned with the manual of the Joanna Briggs Institute, providing detailed information, and constituting a fundamental tool for the transparency and reliability of scoping reviews.

References

1. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. Int J Soc Res Methodol. 2005; 8(1):19-32.
2. Grant MJ, Booth A. A typology of reviews: an analysis of 14 types and associate methodologies. Health Information and Library Journal. 2009; 16: 91-108.
3. Peters MD, Godfrey CM, Khalil H, McInerney P, Parker D, Soares CB. Guidance for conducting systematic scoping reviews. Int J Evid Based Healthc. 2015; 13:141-6.
4. Peters MDJ, Godfrey C, McInerney P, Baldini Soares C, Khalil H, Parker D. Scoping reviews. In: Aromataris E, Munn Z, editors. Joanna Briggs Institute Reviewer's Manual. Adelaide, Australia: Joanna Briggs Inst; 2017.
5. Langlois EV, Daniels K, Akl EA, editors. Evidence synthesis for health policy and systems: a methods guide. Geneva: World Health Organization; 2018.
6. Toma TS, Barreto JOM. *Métodos na EVIPNet Brasil: ferramentas SUPPORT para políticas informadas por evidências*. Bol Inst Saúde (Print). 2016; 17:43-49.
7. Pham H, Rajic A, Greig JD, Sargeant JM, Papadoopulos A, McEwen SA. A scoping review of scoping reviews: advancing the approach and enhancing the consistency. Res Synthesis Methods. 2014; 5:371-385.
8. Tricco A, 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(7):467-473.
9. Munn Z, Peters M, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. BMC Med Res Methodol [Internet]. 2018 [accessed 8 August 2019]. Available at: https://doi.org/10.1186/s12874-018-0611-x.
10. Guba EC. The alternative paradigm dialog. In: Guba EC, editor. The paradigm dialog. Newbury Park, London, New Delhi: Sage Publications; 1990:17-27.
11. Minayo MCS *O desafio do conhecimento: pesquisa qualitativa em saúde*. 3.ed. São Paulo: Hucitec-Abrasco; 1992.
12. Soares CB, Hoga LAK, Matheus MCC. *Revisão sistemática de estudos qualitativos e síntese de evidências*. In: Barbosa D, Taminato M, Fram D, Belasco A, organizadoras. *Enfermagem baseada em evidências*. São Paulo: Atheneu. 2014:79–92.
13. Cordeiro L, Soares CB. Action research in the healthcare field: a scoping review. JBI Database Syst Rev Implement Rep. 2018; 16(4):1003-1047.
14. Reason P, Bradburry H. Introduction. In: Reason P, Bradburry H, editors. The SAGE Handbook of action research: Participative Inquiry and Practice. 3a.ed. London: Sage; 2008.
15. Cordeiro L, Soares CB, Rittenmeyer L. Unscrambling method and methodology in Action Research traditions: theoretical conceptualization of praxis and emancipation. Qualit Res. 2017; 7(4):395-407.
16. Alves-Mazzotti AJ, Gewandsznajder F. *O método nas ciências naturais e sociais: pesquisa quantitativa e qualitativa*. São Paulo: Pioneira Thomson Learning; 1999.
17. Centre for Review and Dissemination. Guidance notes for registering a systematic review protocol with PROSPERO [Internet]. 2016 [accessed 10 Abril 2019]. Available at: https://www.crd.york.ac.uk/PROSPERO/documents/Registering a review on PROSPERO.pdf.
18. Cordeiro L, Soares CB, Rittenmeyer L. Action research methodology in the health care field: a scoping review protocol. JBI Database System Rev Implement Rep. 2015; 13(8):70-78.

Implementation of clinical management using kanban in emergency services: a qualitative systematic review

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Abstract

The objective was to evaluate the application and implementation of clinical management by kanban in the emergency hospital services (EHS). Method: Qualitative systematic review with metassynthesis. Data collection was carried out in the following information sources: Web of Science, Scopus, CINAHL, PsycoInfo, Embase, Eric, Pubmed (included Medline), Lilacs, ScienceDirect, Google Scholar. Following descriptors were used: health personnel; comprehensive health care; patient care team; continuity of patient care; quality improvement; patient centered care; healthcare; guide to clinical practice; lean thinking; quality of health care; lean management; hospital emergency service; hospital/hospital logistics; emergency medical services. Articles in English, Portuguese and Spanish published between 2008 and 2016 were selected. It was used methodological quality analysis tools of qualitative research and data extraction from JBI-SUMARI. The categorization of the findings was performed on the basis of meaning similarity and the categories were aggregated into synthesis. Results: There were included 22 articles, aggregated in eight categories and three synthesis. Kanban appears as a lean-related tool in EHS. The implementation of the lean should be further explored in the emergency services, focusing on sustainability, time of care and user satisfaction.

Keywords: Hospital Administration, Emergency Service, Hospital; Qualitative Review.

Introduction

Technological arrangements for care coordination are a set of technologies, best practices and management levels. Preferably implemented in an articulated manner, they constitute models of multi-professional intervention aimed at the application of scientific knowledge for practical purposes in management and provision of health care.[1,2] Examples of such technological arrangements are systems of risk classification, such as the Manchester Triage System (MTS), and clinical management, such as kanban.

A systematic review[3] on the use of triage scales in emergency departments (ED) found these scales (including MTS risk classification) to be supported by limited or insufficient evidence. However, another systematic review[4] that specifically analysed the efficacy of MTS for risk classification of patients supported its applicability and validity for use in children and adults in ED.

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In regard to clinical management, a systematic review[5] addressed the practice of lean thinking[I] (which includes kanban)[II] in health services. This review identified five articles on ED and six on the use of kanban, published from 1998 to 2008. All articles reported positive results related to lean thinking.

For the purpose of contributing to the implementation of lean thinking in hospitals, Andersen and colleagues[6] conducted a systematic review of review articles published from 2000 to 2012, and developed a list of 23 facilitators to this implementation.

Therefore, considering different technological arrangements in emergency hospital services (EHS) and the existence of recent systematic reviews on the Manchester Triage System and lean thinking in general, this systematic review focused on evaluating the application and implementation of clinical management by kanban in EHS.

Method

Reviews in health care are fundamental to summarize knowledge and inform health workers, managers and researchers. Its purpose is to provide evidence base for the best clinical decision, planning and administration of health services, definition of policies and programmes to be implemented, as well as definition of new research strategies.[7]

On the basis of the Evidence-Based Research Movement, systematic reviews of qualitative data allow to integrate primary studies using the interpretivist and critical paradigm in a summarized but not statistically combined manner.[8]

Moreover, a meta-synthesis is a synthesis of qualitative research on a given topic to understand a phenomenon and expand knowledge, thus improving the use of knowledge in health policy, practice and decision making.[8]

In view of the need to develop the best approaches for the review of qualitative research, Cochrane Collaboration has established the Qualitative Research Methods Group.[8]

Another important international organization in the conduct of systematic reviews of qualitative research is the Joanna Briggs Institute (JBI), based within the University of Adelaide, Australia. JBI collaborates internationally with over 60 countries by promoting and supporting the conduct of meta-aggregation (categorization of findings) and also the transfer and utilization of review results for health decision making.[9]

Review question

The PICo[9] strategy was used to elaborate the review question: ‘How is the application and implementation of clinical management by kanban in EHS?’, that is, P – users, health workers, managers; I – application and implementation of clinical management by kanban; Co - emergency hospital services (EHS).

Controlled vocabulary terms and databases

Based on the review question, some searches were made using the Health Sciences Descriptors (DeCS) through the Virtual Health Library (VHL), and the Medical Subject Headings (MeSH) through the National Center for Biotechnology Information (NCBI). Then, the following descriptors were used: health personnel; comprehensive health care; patient care team; continuity of patient care; quality improvement; patient centered care; healthcare; guide to clinical practice; lean thinking; quality of

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[I] The lean system is a management philosophy that proposes practices to detect and identify problems in real time, to establish the basis for learning and continuous improvement, to reduce waste, and to institutionalise new operating procedures[5].

[II] Kanban is a board to signal workflow, systems and processes. It is used in healthcare, for example, to ensure replenishment of supplies (on time and quantity required) and to reduce stock[5].

health care; lean management; emergency hospital service; hospital/hospital logistics; emergency medical services.

The following databases were searched: Web of Science, Scopus, CINAHL, PsycoInfo, Embase, Eric, Pubmed (included Medline), Lilacs, ScienceDirect, Google Scholar.

Data collection

An initial limited search of MEDLINE and CINAHL was undertaken followed by analysis of the text words contained in the title and abstract, and of the index terms used to describe the article. A second search using all key words and descriptors identified was undertaken for each database defined. Then, the reference list of all articles identified were screened to search for additional studies. The inclusion criteria were articles published in English, Portuguese and Spanish, from 2008 to 2016, which could answer the review question. The exclusion criteria were articles on the use of kanban for inventory management.

After data collection, duplicate articles were excluded, as well as those that did not meet the aims of this review by screening of titles and abstracts. The articles selected for retrieval were assessed by two independent reviewers for methodological validity prior to inclusion in the review using a standardized tool from JBI-SUMARI (System for the Unified Management, Assessment and Review of Information) for critical appraisal of qualitative research. This tool consists of ten questions and is available online[III]. Any disagreements between the reviewers were resolved by consensus or by a third reviewer. In order to establish the level of confidence in evidence produced, the confidence in qualitative (ConQual) approach[10] was used to assess the syntheses found in this review.

Data analysis

Data were extracted from articles included in the review using the standardized data extraction tool from JBI-SUMARI.[9] The data extracted included specific details about the phenomenon of interest, populations, study methods, and results of relevance to the review question and specific objectives.

Categorization of findings was carried out on the basis of similarity in meaning. The categories were then subjected to a meta-synthesis in order to produce a comprehensive set of synthesized findings (syntheses) that could be used as a basis for evidence-based practice.

Ethical Aspects

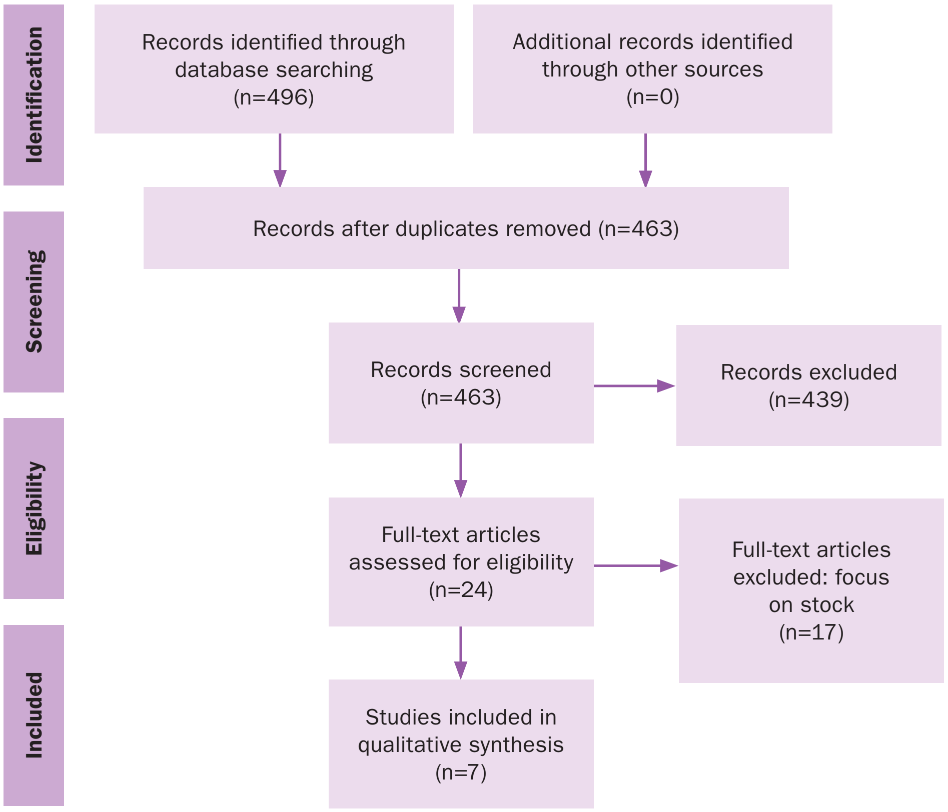
Ethical approval from a Human Research Ethics Committee was not sought as this was a systematic review of the published literature. It is noteworthy to mention that the ethical precepts which deals with the ethics of scientific research, both for data analysis and dissemination of the results, were respected in accordance with the Resolution No. 466/12 of the Brazilian National Council of Health. The review protocol was presented and discussed with the research team of the project of PPSUS (Research programme for the Brazilian Unified Health System - SUS), to which this article is related.

Results

In total, 496 titles were identified through database searches. Then, 33 duplicates were removed and 463 titles and abstracts were screened, of which 24 were selected for full-text review, and 17 were excluded because they focused on stock organization. Finally, 7 articles were assessed for quality. The selection process is presented as a PRISMA flow diagram in Figure 1 (PRISMA stands for Preferred Reporting Items for Systematic Reviews and Meta-Analyses).

[III] Accessed 7/20/2019 at https://joannabriggs.org/sites/default/files/2019-05/JBI_Critical_Appraisal-Checklist_for_Qualitative_Research2017_0.pdf

Figure 1. Flow diagram of the article selection process



Description of included articles

The seven articles included in this review were published from 2008 to 2016.[12-18] Four studies were from the United States,[13,16-18] one from Australia,[12] one from Sweden,[14] and one from United Kingdom.[15]

The methods used in the articles were the following: retrospective case study,[12] multiple case study,[14] semi-structured interviews,[15] participant observation,[16,18] pre-post retrospective analysis,[13] and lean six sigma methodology.[17]

Different urban contexts were presented: public,[14,15] private,[13,16-18] university hospital,[12,14] children's hospital.[13] Participants of the studies were managers, physicians, nurses and nursing assistants, midwives, residents, and patients.[12-18]

Methodological quality of included articles and level of confidence in the syntheses

Regarding the quality assessment of included articles, none scored below six out of ten questions, four scored six[12,13,16,17], and three scored seven (Box 1).[14,15,18] The questions with 'no' response were related to congruity between the philosophical perspective and the research methodology, and no statement locating the researcher and no acknowledgement of their influence on the research. The level of confidence in the syntheses was graded as low, according to the ConQual approach (Box 2).

Box 1. Quality assessment of included articles

ID	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Nota
11	N	S	S	S	S	N	N	U	S	S	6
12	N	S	S	S	S	N	N	U	S	S	6
13	N	S	S	S	S	N	N	S	S	S	7
14	N	S	S	S	S	N	N	S	S	S	7
15	N	S	S	S	S	N	N	U	S	S	6
16	N	S	S	S	S	N	N	U	S	S	6
17	S	S	S	S	S	N	N	U	S	S	7

Box 2. Assessment of the syntheses by the ConQual approach.[18]

Syntheses	Type of research	Dependability	Credibility	ConQual score
1	Qualitative	Downgrade 1 level*	Downgrade 1 level**	Low
2	Qualitative	Downgrade 1 level*	Downgrade 1 level**	Low
3	Qualitative	Downgrade 1 level*	Downgrade 1 level**	Low

* Downgraded one level due to common dependability issues across the included primary studies (the majority of studies had no statement locating the researcher and no acknowledgement of their influence on the research).

** Downgraded one level due to a mix of unequivocal and credible findings.

Findings, categories and syntheses

In the analysis process, 93 findings were identified: 32 were rated as unequivocal and 71 credible. These findings were grouped into eight categories, which were then meta-aggregated into three syntheses described below.

Synthesis 1: The implementation of clinical management in EHS has shown positive effects of Lean on length of stay, flow and admission of patients, and negative effects on direct patient care time. This synthesis included two categories.

Positive effects of Lean

Impact on quality, reduced length of stay, optimised patient flow, strengthened horizontal relationships, improved patient satisfaction, increased admissions, resulting in financial effect. This is illustrated by the following quotes:

> *'All the performance indicators have improved during the first post-Lean year.'*[16, p.506]
>
> *'Patient satisfaction began to improve from post-Lean year 1 and continues to improve during the months after the reported 2- year post-Lean period.'*[16, p.508]

Negative effects on Lean

Less time spent in direct patient care. This is illustrated by the following quotes:

> *'[They] have not been successful in several specific clinical settings.'*[17 p.1103]
>
> *'I would imagine most of it is down to the four hour wait because we are under such*

> *pressure to try and push people through quicker' (nurse practitioner).*[15, p.220]

Synthesis 2: The implementation of clinical management in EHS has required different involvement and support from staff and managers, as not all professionals have had the same understanding of Lean. It has developed healthcare management and critical thinking, empowered staff, changed professional roles, eliminated unnecessary processes, identified problems, and used tools such as kanban. This synthesis included four categories.

Staff involvement

The involvement/support of staff and managers with lean has varied, as not all professionals have had the same understanding of Lean, it has not been a priority for residents, it has overwhelmed nursing staff and generated conflicts across different levels of management. This is illustrated by the following quotes:

> *'At the moment we've seen all the trials and some of them seem to have gone really well, but today I don't think it is going that well to be honest, obviously because we're sitting and we're watching, because we're not busy doing everything like they are, it just seems very chaotic' (Nurse).*[15, p.218]

> *'The junior doctors are only here for three months and they rotate different jobs the whole time they're here. They and the residents, you know, have a lot of work to do. And it's [the eight hour project] probably just not a priority' (Nurse 031).*[12, p.944]

Organizational change

Lean has supported problem solving in healthcare management, team working in the same room, collaboration across hospital's units, and staff's critical thinking, aiming at a cost-effective model of emergency services. This is illustrated by the following quotes:

> *'Oh yes it will be cost effective of course [...] but it's to make it more efficient and [a] more efficient department is effective, [...] towards the staff, for the finances, most of all the patients, for the treatments' (Junior sister).*[15, p.223]

> *'It's about making the department more efficient with the resources that we have currently' (Consultant EM Physician).*[15, p.223]

Professional roles

Lean has contributed to staff empowerment, and it has led to changes such as: increase in the number of professionals (e.g. more daily routine physicians and radiologists), replacement of nursing technicians with nurses, and broadening of the scope of physicians' responsibility for patient care. This is illustrated by the following quotes:

> *'Management took a subordinate role when it came to solving flow issues and let the frontline staff identify problems and come up with their own solutions'.*[18, p.181]

> *'More empowered staff eager to institute their ideas as opposed to a reluctant staff feeling forced to institute top-down process improvements'.*[18, p.181]

Work processes and tools

Lean has contributed to process improvements by eliminating unnecessary processes, identifying problems using tools such as kanban, and focusing on staff and their skills to solve problems, as well as on patient flow and discharge. Only two findings mentioned kanban. The following quotes illustrate these findings.

> *'Tools such as the takt board and the whiteboard helped visualize and identify problems (e.g. deviations from the prototype) in everyday work'.*[14, p.280]

> *'All services introduced a takt board, a computer-based tool to monitor the number of patients assessed by physicians per hour related to the anticipated pace of work, and a whiteboard to capture and manage improvement ideas at the ED.'*[14, p.274]

Synthesis 3: The implementation of clinical management in EHS should observe the principles of Lean, and involve managers and staff. It has been difficult to measure, especially by users, and has raised doubts about its sustainability. This synthesis included two categories.

Barriers to lean implementation

Operational efficiency through Lean has been difficult to measure, raised doubts about its sustainability, and it has taken time for users to realize changes. This is illustrated by the following quotes:

> *'The patient cannot fully quantify the quality of the service.'* [18, p.181]

> *'[They] Have been sustainable without increasing expense per patient or the number of ED treatment areas.'* [18, p.181]

Appropriate lean implementation

The implementation process should observe the principles of Lean, such as adaptation to reality at the request of management or staff. This is illustrated by the following quotes:

> *'[It] Has been associated with both short-term and longer-term positive effects on department operations.'*[18, p.181]

> *'Applied this methodology in our clinical setting to identify four opportunities for improving the operational efficiency in our TRU.'*[17, p.1103]

Discussion

All included studies reported the use of kanban as a tool, that is, as a card, a visual signalling system that indicates new items, supplies or services, on time and quantity required.[19]

However, it is also a method, but the use of kanban as a method were not reported by the included studies. This method has been incorporated by David J. Anderson for software development since 2010.[20]

In Brazil, since the Ordinance No. 1,660/2011,[21] which reformulated the National Emergency Care Policy and established the Emergency Care Network in the Unified Health System (SUS), and Ordinance No. 1,663/2012,[22] which addressed the SOS Emergency Program, the Ministry of Health has been encouraging the adoption of technological arrangements for healthcare management in emergency hospital services (EHS), including the kanban method, in order to optimise bed management and foster continuous improvement.[23] EHS that implemented the kanban method have shown improvement, compared to hospitals that either faced barriers to its implementation or failed to implement it.[23]

On the other hand, unlike in most Brazilian EHS, in those analysed in this review kanban has been used only as a tool of Lean strategy, intended to improve processes and eliminate unnecessary steps, maximize flow and minimize waste. Lean has been implemented in health services since 2002.[24]

The syntheses of this review corroborate the evidence that Lean has the potential to improve services when it is properly applied, including support from staff and especially from managers, otherwise, the changes may be superficial and unsustainable.[24-25]

Recommendations for practice

- Positive effects of Lean in EHS are related to length of stay, flow and admission of patients.
- Negative effects of Lean in EHS are related to direct patient care time.
- Lean in EHS develops healthcare management and critical thinking, empowers staff, changes professional roles, eliminates unnecessary processes, identifies problems, and uses tools such as kanban.
- Implementation in EHS should observe the principles of Lean, and involve managers and staff.
- It is necessary to measure both user satisfaction and sustainability of Lean changes in EHS.

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References

1.Fleury ACC. *Organização do trabalho industrial: um confronto entre teoria e realidade*. São Paulo [tese]. São Paulo: Escola Politécnica, Universidade de São Paulo; 1978.

2. Ramanantso AB. Strategic Technology Management. Chichester, UK: John Wiley & Sons; 1992.

3. Farrohknia N, Castrén M, Ehrenberg A, Lind L, Oredsson S, Johsson H, Asplund K, Goransson K. Emergency Department Triage Scales and Their Components: A Systematic Review of their Scientific Evidence. Scand J Trauma Resusc Emerg Med. 2011; 19:42.

4. Azeredo TR, Guedes HM, Rebelo de Almeida RA, Chianca TC, Martins. Efficacy of the Manchester Triage System: a systematic review. Int Emerg Nurs. 2015; 23(2):47- 52.

5. Mazzocato P, Savage C, Brommels M, Aronsson H, Thor J. Lean thinking in healthcare: a realist review of the literature. Qual Saf Health Care. 2010; 19:376-382.

6. Andersen A, Rovik KA, Ingebrigtsen T. Lean thinking in hospitals: is there a cure for the absence of evidence? A systematic review of reviews. BMJ Open. 2014; 4.

7. Egger M, Davey Smith G, Altman DG (Org). Systematic Reviews in Health Care: Meta- analysis in context. 2001.

8. Lopes ALM, Fracolli LA. *Revisão sistemática de literatura e metassíntese qualitativa: considerações sobre sua aplicação na pesquisa em enfermagem*. Texto Contexto Enferm, Florianópolis. 2008; 17(4): 771-8.

9. JBI. The Joanna Briggs Institute. Joanna Briggs Institute Reviewer's Manual: 2011 edition. Adelaide: The Institute Joanna Briggs; 2011.

10. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med. 2009; 6(6).

11. Munn Z, Porrit K, Lockwood C, Aromataris E, Pearson A. Establishing confidence in the output of qualitative research synthesis: the ConQual approach BMC Medical Research Methodology. 2014; 14:108.

12. G Leggat S, Gough R, Bartram T, Stanton P, Bamber GJ, Ballardie R, Sohal A. Process redesign for time-based emergency admission targets. J Health Organ Manag. 2016; 19;30(6):939-49.

13. Beck MJ, Okerblom D, Kumar A, Bandyopadhyay S, Scalzi LV. Lean intervention improves patient discharge times, improves emergency department throughput and reduces congestion. Hosp Pract (1995). 2016; 44(5):252-259.

14. Mazzocato P, Thor J, Bäckman U, Brommels M, Carlsson J, Jonsson F, Hagmar M, Savage C. Complexity complicates lean: lessons from seven emergency services. J Health Organ Manag. 2014; 28(2):266-88.

15. Timmons S, Coffey F, Vezyridis P. Implementing lean methods in the emergency department: the role of professions and professional status. J Health Organ Manag. 2014; 28(2):214-28.

16. Dickson EW, Anguelov Z, Vetterick D, Eller A, Singh S. Use of lean in the emergency department: a case series of 4 hospitals. Ann Emerg Med. 2009; 54(4):504-10.

17. Parks JK, Klein J, Frankel HL, Friese RS, Shafi S. Dissecting delays in trauma care using corporate lean six sigma methodology. J Trauma. 2008; 65(5):1098-104.
18. Dickson EW, Singh S, Cheung DS, Wyatt CC, Nugent AS. Application of lean manufacturing techniques in the Emergency Department. JEmergMed. 2009; 37(2):111-82.
19. Santos, VM. *O que é Lean Healthcare e como isso está mudando a saúde?* 2017 [accessed 5 July 2017]. Available at: https://www.fm2s.com.br/o-que-e-lean-healthcare/.
20. InfoQ. *Kanban com o pioneiro: Entrevista com David J. Anderson*. 2012 [accessed 10 July 2017]. Available at: https://www.infoq.com/br/articles/kanban-david-anderson-conceitos-e-mitos.
21. *Ministério da Saúde (BR). Portaria 1.660/2011. Reformula a Política Nacional de Atenção às Urgências e institui a Rede de Atenção às Urgências no Sistema Único de Saúde (SUS),* 2012.
22. *Ministério da Saúde (BR). Portaria n1663 de 6 de agosto de 2012. Dispõe sobre o Programa SOS Emergências no âmbito da Rede de Atenção às Urgências e Emergências (RUE)*, 2012.
23. Petry D. *Análise de implantação do Kanban em Hospitais do Programa SOS Emergências. [dissertação] - Curso de Gestão de Serviços de Saúde*, UFBA, Palmas. 2016:151.
24. Womack JP, Byrne AP, Fiume OJ, Kaplan GS, Toussaint J, Miller D. Going Lean in Health Care. Int Health Care Improv. Cambridge. 2005.
25. Joosten T, Bongers I, Janssen R. Application of lean thinking to health care: issues and observations. Int J Qual Health Care. 2009; 21:341-347.

Humanising practices in Primary Health Care: a qualitative systematic review

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Abstract

Objective: To identify humanising practices in Primary Health Care. Method: Qualitative systematic review. Studies were searched in the following information sources: PubMed, CINAHL, Scielo, using the key word primary health care, together with the following key words: humanisation of assistance, holistic care/health, patient centred care, user embracement, personal autonomy, holism, attitude of health personnel. Results: We included 17 studies published between 2012 and 2018. Data were systematized into seven categories: (a) Offer of humanising practices, (b) Difficulties of humanising practices, (c) Facilitators of humanising practices, (d) Need for training on humanising practices, (e) Professional-patient bond, (f) Positive effect of humanising practices in teamwork, and (g) Attitudes and beliefs that reflect humanising practices. Conclusions: Humanising practices continue to be developed in Primary Health Care and their positive effects are recognized by health professionals. Therefore, investments in the development of these practices and in research should continue, also including the perceptions of patients and management bodies.

Keywords: Primary Health Care, Primary Health Care, Humanisation of Assistance; Health personnel; Qualitative Systematic Review.

Introduction

In 2006, the Brazilian National Policy for Humanisation of Health Care and Management was published,[1] also known as the National Humanisation Policy (PNH, in Portuguese), or HumanizaSUS. The PNH promotes the use of soft technologies, such as autonomy and empowerment of healthcare users, bond and qualified listening between patients and health professionals, extended clinical care and singular therapeutic project for the management of complex cases, among others.

Primary Health Care (PHC) represents the entrance door to the health system and organizes the service network. In 2013, a systematic review on the PNH identified that humanisation permeates the work processes and those involved in primary care.[2]

In 2016, another systematic review addressed the health workforce's understanding of the role of humanisation practices in PHC, and reported on the following difficulties to make these practices effective: fragmented concepts of health and care, and lack of investment in multidisciplinary teamwork, community empowerment, professional-patient bond, and user embracement.[3]

In view of these difficulties and the importance and need of further research on this topic, the objective of this review was to identify humanisation practices in Primary Health Care.

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Method

The purpose of a systematic review is to enable the translation of the best scientific evidence into policies, practices and decisions in the healthcare context.[4] This design allows to group findings of primary studies into categories, which are then subjected to a meta-synthesis.[5] This is a process consisting of six steps: (a) identifying the theme and formulating the research question; (b) defining data sources for search; (c) establishing study inclusion and exclusion criteria; (d) selecting and processing data, and excluding duplicates; (e) detailing the studies selected, and analysing data - this analysis consists of (e_1) assessing the quality of included studies, and (e_2) interpreting the findings.[5]

The PICo strategy was used for the research question construction, identifying its key-aspects: Population (P), Phenomenon of Interest (I), and Context (Co).[6] In this review, the Population was PHC professionals, the Phenomenon of Interest was the humanisation practices, and the Context was PHC services. After adjusting the research objectives to PICo, the review question was defined: ‘How do health professionals offer humanisation practices in PHC services?’

The concepts of humanisation of the Brazilian National Humanisation Policy (PNH) focus on key principles such as access, empathy, humanisation of assistance, personal autonomy, comprehensive health care, and user embracement. In data collection, the controlled descriptors used were based on these concepts. The following key words were used: primary health care with other descriptors, separately: humanisation of assistance, holistic care/health, patient centred care, user embracement, personal autonomy, holism, and health personnel attitude.

The following databases were searched in May 2018: PubMed, CINAHL and Scielo, and the search was limited to the period from 2012 to 2018, in order to locate articles published after the data collection of the above mentioned systematic review.[3] The inclusion criteria were articles on how PHC professionals offer humanisation practices, available in Portuguese, English and Spanish languages. Articles on patients and students, and/or studies conducted in hospital settings were excluded. The articles were organized using Excel.

Two independent reviewers assessed the articles and the final selection was made by consensus, based on a comparison of the evaluation of both reviewers. The quality of the articles was assessed using a tool from JBI-SUMARI (System for the Unified Management, Assessment and Review of Information) for qualitative research, which consists of ten questions and is available online.[V] The following information was extracted from each article: country of origin, number of professionals, objectives, data collection and analysis methods, protection of human subjects, findings.

Data were organized into empirical categories that emerged from the analysis and review by Della Nora and Junges.[2] Four independent reviewers assessed the categories in terms of their respective scope and definition. Any disagreements were resolved by consensus. The aim was to provide a meta-synthesis by integrating the categories.

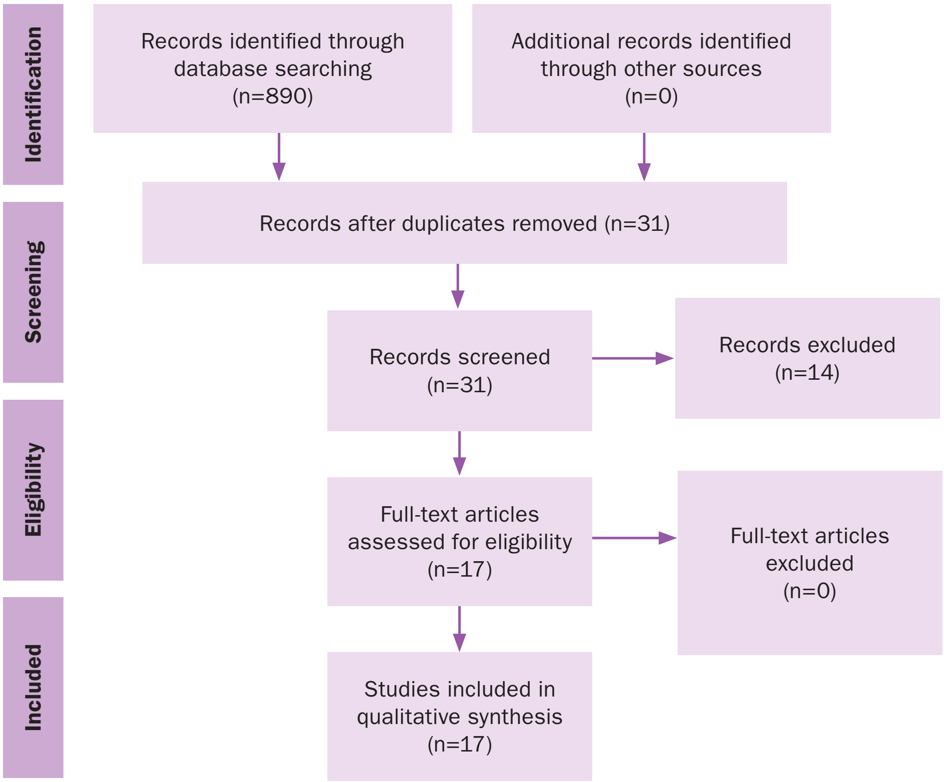
In regard to the ethical aspects, this study was developed in accordance with the guidelines and standards of the Resolution No. 466/12 of the Brazilian National Council of Health, however, ethical approval from a Human Research Ethics Committee was not sought as this was a systematic review of the published literature.

Results

From potential 890 references, reduced to 31 non duplicated studies, 17 qualitative studies were included and systematically reviewed,[7-23] as Figure 1 shows. Studies retrieved were conducted from 2012 to 2018. Three studies were published in English and 14 in Portuguese.

[V] Accessed 7/20/2019 at https://joannabriggs.org/sites/default/files/2019-05/JBI_Critical_Appraisal-Checklist_for_Qualitative_Research2017_0.pdf

Figure 1. Flow diagram for the inclusion and exclusion process of the studies[24]



In total, the studies included 248 PHC professionals from United States[11-13,23] and Brazil.[7-10,14-22] Seven studies[12,14,15,17,20-22] did not mentioned the number of professionals by category, and among those that mentioned it,[7-11,13,16,18,19,21,23] 61% of these professionals were nurses, and the other 39% mostly were nursing technicians, community health workers, physicians, pharmacy technicians, and administrative personnel.

In regard to the quality assessment of included studies, four articles scored seven,[8,16,17,21] eleven scored eight[7,9-15,19,20,23] and two scored nine.[18,22]

The findings were aggregated in seven empirical categories: (1) offer of humanising practices; (2) difficulties of humanising practices; (3) facilitators of humanising practices; (4) need for training on humanising practices; (5) professional-patient bond; (6) positive effect of humanising practices in teamwork; (7) attitudes and beliefs that reflect humanising practices.

Category 1: Offer of humanising practices

This category included perceptions of health care professionals about the offer of humanising practices, their organization, conduct and consequences. This category was present in 12/17 articles.

Health care professionals identified as offer of humanising practices: educational groups, home visits, health assistance by free demand (access and use of health services whenever necessary), patient centred care, and user embracement. They reported that these practices improve the access of the community to health services, and promote longitudinality and shared care between professional-user.[7,8,14,16,23]

> *The doctors (More Doctors Program) always see everybody, sometimes more 5 or 6 cases than they should, because they also take into consideration the riparians and community people who come with the expectation of*

> *being seen by a doctor'. Sometimes the boat is delayed, or there is bad weather, and when they arrive there are no appointments available. [...] The users make all this sacrifice, but the doctors see everybody very well.* [7]

For health care professionals, embracement is qualified listening, it is the moment when the user, even without appointment, is heard by the health care professional (usually a nurse or physician), who identifies the user's needs to decide for the best treatment. Embracement was the humanising practice more mentioned by professionals.[8,10,11,16,17,21,22]

> *[...] most of them (users), just to talk to him, by talking you already solve the problem. He needs someone to talk to, someone who listens, [...] right here in the service, the Family Health Strategy worker does this kind of thing, which is the embracement, talking to the patient, helping him.*[10]

In two articles the offer of humanising practices was taken into consideration not only in the PHC context, but also in the health care network and intersectoral articulation. Based on the analysis of these articles, it was possible to notice that the users' needs go beyond the health care sector, and referral to other sectors and/or services to meet their needs depends on the sensitivity and character of health care professionals. [9.20]

> *A woman was frequently spanked by her son, a drug addict. One day I was at her house, attending to her bedridden husband, when her son threatened her with a knife... I had to go up to him and hit him with my clipboard, which was the only thing I had at hand... She stayed behind me. So, I stood between the aggressor and the victim. If I hadn't done this, I think he would have killed her...[...] I took this case to the mayor. The son was arrested [...] I referred her [not officially, but by friendship] to the social worker, nurse and psychologist [...]*[20]

Category 2: Difficulties of humanising practices

This category included perceptions of health care professionals about difficulties, barriers and limitations related to humanising practices. This category was present in 8/17 articles.

Health care professionals reported factors related to: authoritarian approach of the health professionals, focusing on their needs instead of being patient centred;[7,13] excess of demand;[8,17,22] barriers to communication between health professionals and users;[15,22] users' lack of knowledge about embracement and familiarity with the 'traditional' model of health care;[15-17] turnover of professionals who perform user embracement;[11] inadequate implementation of user embracement[16,22] and lack of user embracement in the service.[16]

> *'Of course, when you are too stressed or not patient-centred enough it happens, and I, so to speak, push things in my direction. I have my own agenda and I do not invite the patient into a dialogue; that has happened many times.'*[13]

Category 3: Facilitators of humanising practices

This category included perceptions of health care professionals about facilitators and factors that contribute to humanising practices. This category was present in 5/17 articles.

Health care professionals mentioned factors related to: knowledge of the needs of the community served and individualized care;[16,18] embracement;[17] appreciation and recognition of the work of health care professionals;[21] sufficient number of professionals for user embracement;[21] adequate physical space;[21] holding of technical meetings;[21] a well-coordinated team and a system to support the work of health care professionals.[23]

> *According to managers and professionals, low turnover of professionals, and their willingness to do the embracement and to schedule appointments promote the strengthening of bonding and expanding of access. In the words*

> *of the professionals, embracement is seen as a reference for people of the coverage area.*[21]

Category 4: Need for training on humanising practices

This category included perceptions of health care professionals about the need for training and raising awareness on humanising practices, for both health care professionals and users, in different contexts. This category was present in 3/17 articles.

> *'I think we usually give recommendations that are adapted to middle-aged, well-educated Swedish men. It is not that you have always thought about it ahead, I mean, how it is to be a woman from a Muslim country, having a lot of children at home, so it is not for sure that your style will work.'*[13]

Category 5: Professional-patient bond

This category included perceptions of health care professionals about the development of professional-patient bond for the offer of humanising care. This category was present in 9/17 articles.

Health care professionals identified as factors that both contribute to and result from professional-patient bonding: qualified listening;[12,20,21] dialogue;[9,13,17,21] understanding of embracement;[17] professionals' feeling of belonging to the community served;[17] empathy;[18] confidence of users;[19,21] work recognition;[19] the coverage area of the health center;[16,18] humanisation;[18] engagement with the patient's family;[16] holding users accountable for their self-care;[16] treatment adherence;[19,21] the best clinical outcome.[21]

> *[...] I think that when we see suffering in front of us, we try to be more human. There is also empathy, you should put yourself in someone else's shoes. So, I believe that is it. (...) looking at problems more closely, getting to know reality, then you try to provide a better care [...]*[18]

Category 6: Positive effect of humanising practices in teamwork

Humanising practices contribute to teamwork in PHC. This category was present in 9/17 articles. Health care professionals mentioned effects of humanising practices in teamwork related to: interpersonal relationship, unity and communication,[7,8,10,13] work process of the team organized from the perspective of coordination of care,[9,16,18] technical division of work and recognition of the different professional roles.[17,18,20]

> *Everyday, each of us has to do some thinking, to see what is our real role and see if community is also understanding our role. It has to be a share of information [...] more communication, more interaction among workers, between users and workers, it has to be like a team. Everybody trying to speak the same language [...]*[20]

Category 7: Attitudes and beliefs that reflect humanising practices

This category was present in 12/17 articles and is related to the attitudes and beliefs of health care professionals that reflect humanising practices, such as respect, time of care, and recognition of both user knowledge and individuality.[7-13,15,17,19,21,23]

> *[...] time for physical examination, time to look at the patient's face, follow-up, good care, and time for a better patient embracement; the physical examination, for example, sometimes some of our physicians who worked with us here in the municipality didn't have this time of examining every patient; before, people would complain, saying "the doctor didn't even look at me, at my face", but these doctors (More Doctors Program) give full attention.*[7]

Based on these categories, two syntheses were developed: (1) The offer of humanising practices depends on the attitudes and beliefs of health care professionals, improves the bond with the user, and has positive effect on teamwork; (2) The health care professionals in Primary Health Care settings reported more difficulties than facilitators, and focused on the need for training on humanising practices.

The syntheses have shown that health care professionals relate humanising practices to the care that should be provided at PHC according to the Brazilian National Primary Health Care Policy, that is, comprehensive, people-centred care, based on teamwork and extended clinical care.

Discussion

Since the publication of the Brazilian National Humanisation Policy (PNH), there have been increasing reports on the barriers to implement the humanising practices in PHC, as well as the lack of training of health personnel.[2] Among the barriers, it is noteworthy to mention that changes in the work process have been prioritizing the achievement of the contract goals rather than focusing on the health needs of the population, thus hindering the development of the humanising practices and their positive effects in the services.[25]

The process of care in PHC based on the use of soft technologies has been continuously facing increase in demand, limitation of resources, and changes in rules and regulations that leave room for multiple interpretations of the composition of a team and health care model[27]. Therefore, it has been moving further away from the international recommendations to improve PHC as the Health Care Network coordination.[28]

The following recommendations are based on this study's results and intended to contribute to the development of humanising practices: creation of spaces for discussion on the attitudes and beliefs of health care professionals regarding humanising practices; training on humanising practices; appreciation of professional-user bonding; and recognition of the time required to develop teamwork, for instance, meetings and case discussions. These recommendations should be taken into consideration especially in the development of management contracts with social organizations to provide PHC.

In order to meet the need for training on humanising practices, the purpose should be to go beyond one-time training and the banking model of education, and to support innovation, such as training on active reading and active methodologies, which contribute to professional development in ethical and humanistic dimensions, strengthening critical thinking skills and the focus on the needs of the population,[26,29] so as to develop empathy, self-reflection, and closeness to others (both providing and receiving care).

Humanising practices have been developed in Primary Health Care and its positive effects are recognized by health professionals. Therefore, investments in the development of these practices and in research should continue, also including the perceptions of patients and management bodies.

However, there were some difficulties to conduct this research on humanising practices and soft technologies, as proposed in the National Humanisation Policy (PNH), such as the translation of terms into descriptors, which may be a limitation of this study and its results' internationalisation, although efforts to find related terms in other languages contributed to search a wide range of databases.

Regarding the quality assessment of included articles, using the 10 question tool from JBI-SUMARI, questions with 'no' response were related to congruity between the philosophical perspective and the research methodology, no statement locating the researchers and no acknowledgement of their influence on the research. Considering that these questions relate to information that is not necessarily explicit in scientific publications, there were no impact on the quality assessment of the articles. However, researchers should include this information in future publications.

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References

1. *Ministério da Saúde (BR). HumanizaSUS: documento base para gestores e trabalhadores do SUS. Brasília: Ministério da Saúde; 2006*.
2. Della Nora C, Junges JR. Humanization policy in primary health care: a systematic review. Rev Saúde Pública. 2013; 47(6):1-15.
3. Schveitzer MC, Zoboli ELCP, Vieira MMS. Nursing challenges for universal health coverage: a systematic review. Rev Lat Am Enfermagem [Internet]. 2016; 24:2676.
4. Lopes ALM, Fracolli LA. Systematic Review of Literature and Qualitative metasynthesis: considerations about their application in nursing research. *Texto Contexto Enferm*. 2008; 17(4):771-8.
5. Sandelowski M, Barroso J. Handbook for synthesizing qualitative research. New York: Springer; 2007.
6. Lockwood C, Porrit K, Munn Z, Rittenmeyer L, Salmond S, Bjerrum M, Loveday H, Carrier J, Stannard D. Chapter 2: Systematic reviews of qualitative evidence. In: Aromataris E, Munn Z (Editors). Joanna Briggs Institute Reviewer's Manual. The Joanna Briggs Institute [Internet]. 2017 [accessed 8 August 2019]. Available at: https://reviewersmanual.joannabriggs.org/.
7. Comes Y et al. *A implementação do Programa Mais Médicos e a integralidade nas práticas da Estratégia Saúde da Família. Ciência & Saúde Coletiva.* 2016; 21(9):2729-2738.
8. Camelo MS, Lima LR, Volpe CR, Santos WS, Rehem TC. *Acolhimento na atenção primária à saúde na ótica de enfermeiros. Acta Paul Enferm.* 2016; 29(4):463-8.
9. Silva MAC, Aguiar MGG, Moreira TDS. *Entre os nós da rede de saúde mental: as práticas de agentes comunitários de saúde. Rev. Baiana de Saúde Pública.* 2016; 40(3):713-728.
10. Machado LM, Colomé JS, Silva RM et al. *Significados do fazer profissional na estratégia de saúde da família: atenção básica enquanto cenário de atuação*. Rev Fun Care Online. 2016; 8(1):4026-4035.
11. Brasil EGM; Queiroz MVO; Cunha JMH. Receptiveness to the teenager in nursing consultation – a qualitative study. Online braz j nurs [Online]. 2012; 11(2):346-358.
12. Nunstedt H et al. Strategies for healthcare professionals to facilitate patient illness understanding. J Clin Nurs. 2017; 26:4696–4706.
13. Hornsten A et al. Strategies in health-promoting dialogues – primary healthcare nurses' perspectives – a qualitative study. Scand J Caring Sci. 2014; 28:235–244.
14. Silva G, Iglesias A, Dalbello-Araujo M, & Badaró-Moreira MI. *Práticas de Cuidado Integral às Pessoas em Sofrimento Mental na Atenção Básica. Psicologia: Ciência e Profissão.* 2017; 37(2):404-417.
15. Hentges IC, Cogo ALP. *Competências desenvolvidas por equipe de saúde da família sobre o acolhimento após atividade educativa.* J Nurs Health. 2017; 7(1):4-15.
16. Costa RF, Queiroz MVO, Zeitoune RCG. *Cuidado aos adolescentes na Atenção Primária: Perspectivas de integralidade. Esc Anna Nery (impr.).* 2012; 16(3):466-472.
17. Lopes AS, Vilar RLA, Melo RHV, França RCS. *O acolhimento na Atenção Básica em saúde: relações de reciprocidade entre trabalhadores e usuários. Saúde Debate*. 2015; 39(104):114-123.
18. Oliveira AG, Morais IF, Azevêdo LMN, Valença CN, Sales LKO, Germano RM. *O que mudou nos serviços de saúde com a Estratégia Saúde da Família*. Rev Rene. 2012; 13(2):291-9.
19. Bortoli CFC, Bisognin P, Wilhelm LA, Prates LA, Sehnem GD, Ressel LB. *Fatores que possibilitam a atuação do enfermeiro na atenção pré-natal.* Rev Fun Care Online. 2017; 9(4):978-983.
20. Signorelli MC et al. *Violência doméstica contra mulheres e a atuação profissional na atenção primária à saúde: um estudo etnográfico em Matinhos, Paraná, Brasil. Cad. Saúde Pública.* 2013; 29(6):1230-1240.
21. Guerrero P, Mello ALSF, Andrade SR, Erdmann A. *O acolhimento como boa prática na atenção básica à saúde. Texto Contexto Enferm.* L. 2013; 22(1):132-40.
22. Silva MRF, Pontes RJS, Silveira LC. *Acolhimento na estratégia saúde da família: as vozes dos sujeitos do cotidiano. Rev. enferm.* UERJ. 2012; 20(2):784-82.

23. Bikker AP et al. Measuring empathic, person-centred communication in primary care nurses: validity and reliability of the Consultation and Relational Empathy (CARE) Measure. BMC Family Practice. 2015; 16:149.
24. Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med. 2009; 6(6).
25. Silva Vanice Maria da, Lima Sheyla Maria Lemos, Teixeira Marcia. *Organizações Sociais e Fundações Estatais de Direito Privado no Sistema Único de Saúde: relação entre o público e o privado e mecanismos de controle social. Saúde debate.* 2015; 39:145-159.
26. Gallian DMC, *Literatura como remédio: os clássicos e a saúde da alma. 1 edição,* São Paulo: Martin Claret, 2017; 1:214.
27. Melo EA, Mendonça MHM de, Oliveira JR de, Andrade GCL de. *Mudanças na Política Nacional de Atenção Básica: entre retrocessos e desafios. Saúde em Debate*. 2018; 42(1):38–51.
28. Cecilio LC de O, Reis AAC dos. *Apontamentos sobre os desafios (ainda) atuais da atenção básica à saúde. Cad Saúde Pública*. 2018; 34(8):1–14.
29. Mesquita SK da C, et al. *Metodologias ativas de ensino/aprendizagem: dificuldades de docentes de um curso de enfermagem. Trab. educ. saúde.* 2016; 14(2):473-486.

The care of adolescents with sickle cell disease: possibilities and challenges

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Abstract

Sickle cell disease is one of the most common hereditary diseases in the world. People living with this disease may present important symptomatology and serious complications since childhood. Therefore, providing comprehensive care for these individuals is a major challenge for managers and health professionals. Although some measures have improved life expectancy for these people, adolescence period is of great vulnerability, as the young face issues of physical and psychological development, and the changes in their follow-up from child health care services to the adults' ones. This study, based on the SUPPORT methodology, identified three options to inform policies to deal with this transition phase: 1) Use of psychological interventions to improve adherence to treatment; 2) Implementation of a transition programme from paediatric to adult care; 3) Promotion of self-care with adolescents. Barriers and facilitators are discussed, as well as equity considerations for the implementation of these options.

Keywords: Adolescent; Anemia, Sickle Cell; Evidence-Informed Policy

Introduction

Sickle cell disease is one of the most common hereditary diseases in Brazil, therefore, it is important to know its consequences and the scope of care provided by the Unified Health System (SUS), taking into consideration that it primarily affects people of African descent. In this disease, a gene mutation results in the production of haemoglobin S (HbS). Lack or decrease of oxygen tension in the blood causes blood cells to take on a sickle or crescent shape. This leads to occlusion of blood vessels, causing ischemia, pain, necrosis, and permanent tissue and organ dysfunction or damage. Some of these disorders occur in the first year of life, and early diagnosis through neonatal screening is of paramount importance.[1] Given the complexity of the disease, ignorance and institutional racism, comprehensive care for patients is a major challenge for managers and health professionals.

Among the measures that have effectively contributed to improve the quality of life of people with sickle cell disease are infection control through immunization, identification of children at higher risk of cerebrovascular disease by transcranial doppler, and early transfusion of red blood cells. Moreover, educational guidance for families and patients, as well as training of health care professionals are of great relevance. [2,3] All these measures are provided for in the Clinical Protocol and Therapeutic Guidelines for Sickle Cell Disease of the Ministry of Health of Brazil.[4] Recommended medicines and vaccines, as well

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as procedures, including haematopoietic stem cell transplantation, are available at SUS.

Adolescence period is of great vulnerability for those with sickle cell disease, as these patients face the challenges of physical and psychological development, and the changes in their follow-up from paediatric to adult health care services.[5] The transition from paediatric to adult care, in general, happens abruptly, without time for the necessary preparation of the adolescents and their families, hindering the continuity of care.[6,7] In this transfer to adult health care services, adolescents are expected to be autonomous regarding self-care, therefore, there is an understanding that the time of transfer should be decided by considering not only age, but most important the individual readiness. On the other hand, it is also important to consider the feelings of family members, who tend to be overprotective and may feel insecure about these changes.[7]

The objective of this study was to identify health policy option aimed at improving health care of adolescents with sickle cell disease.

Method

We used the knowledge translation approach, based on the SUPPORT tools (SUPporting POlicy relevant Reviews and Trials),[8] including steps for prioritising the research problem, developing an evidence brief for policy, and conducting a deliberative dialogue.

The definition of the priority problem resulted from meetings with an expert in sickle cell disease and with the coordinator of the Technical Area on the Health of the Black Population of the Municipal Health Secretariat of São Paulo.

The development of the evidence brief for policy consisted of the following steps: comprehensive search for evidence in scientific literature databases; selection of relevant systematic reviews; quality assessment of and data extraction from the selected systematic reviews; organization of data in policy options; considerations on equity and implementation of options.

Initial searches were conducted using the terms ‘sickle cell anaemia’ and ‘sickle cell disease’, combined or not with ‘treatment adherence’, in the following databases: Virtual Health Library, PubMed, Health Systems Evidence, Health Evidence, The Cochrane Library, Rx for Change, and PDQ-Evidence. Due to the difficulty in identifying relevant studies, a new search was conducted in PubMed, using the terms ‘treatment adherence’, ‘chronic disease’, and ‘transition health care’, as well as in the Google Scholar. The searches were conducted from October 2015 to March 2016. An additional search was carried out in July 2016, as suggested by deliberative dialogue participants, in order to include an option about self-care, in the following databases: The Cochrane Library, Health Evidence, Health Systems Evidence, PDQ-Evidence, and PubMed, using the terms ‘self-care’ and ‘self-management, combined or not with ‘sickle cell disease’.

In total, 460 articles were identified through database searches, of which 442 were excluded either because they were duplicates or irrelevant, after screening of titles and abstracts. Among 18 eligible articles, 8 were excluded either because they did not meet the objective or presented inconclusive results. One article of a reference list of a systematic review screened was included.

For considerations on implementation, additional searches were carried out in the following databases: SciElo, NICE evidence search and Google Scholar, with a focus on Brazilian studies, as implementation of actions depends on the context.

Searches were carried out by one researcher, while study identification by two researchers independently. All researchers took part in the phase of full-text reading and data extraction, and then a discussion within the research team followed.

Two independent researchers assessed the methodological quality of the systematic reviews using the AMSTAR (A MeaSurement Tool to Assess the methodological quality of systematic Reviews).[9] Any disagreements were resolved by consensus.

The evidence brief was externally peer reviewed. Then, a deliberative dialogue was carried out to discuss about the evidence brief with stakeholders – researchers, health care providers, managers, users, and representatives of user associations.

Results

In the meeting held with the experts, it was informed that diagnosis of sickle cell disease is established early, and paediatric follow-up is relatively well developed in the city of São Paulo. However, the transition to adulthood is a great challenge, when many adolescents may not receive regular health care, and have to go to emergency services in an acute crisis state. For this reason, this evidence brief focused on the care of adolescents.

Options for health policy

We observed that research on interventions that may benefit young people with chronic disease, and particularly those with sickle cell disease, have not been conducted with the necessary methodological rigour. Consequently, systematic reviews are scarce or have inconclusive results. Only ten systematic reviews were identified, and some of them addressed interventions focusing on adolescents with chronic disease, but not specifically with sickle cell disease.

These systematic reviews informed three policy options: 1) Use of psychological interventions to improve adherence to treatment; 2) Implementation of a transition programme from paediatric to adult care; 3) Promotion of self-care with adolescents. Elements of each policy option are presented below, and Table 1 shows the relevant findings regarding benefits, potential harm, cost-effectiveness, knowledge uncertainties / gaps, and applicability of interventions. Some characteristics of the systematic reviews analysed can be found at the end of this article (Appendix).

Option 1. Use of psychological interventions to improve adherence to treatment

Interventions described as psychological include a variety of educational and behavioural strategies. Educational interventions provide information or training for patients on the disease and its treatment. Such verbal or written guidance may be offered by means of single or multiple sessions, telephone calls, consultations in clinics or home visits. Behavioural interventions require a certain behaviour or action on the part of the individual. They may involve techniques, such as establishing targets and monitoring them, reinforcing adherence to medical treatment through rewards, counselling to improve self-esteem. Other strategies are programmes for skill development, emotional peer support, and a combination of educational and psychological approaches.

Option 2. Implementation of a transition programme from paediatric to adult care

Transition programmes are strategies to maintain the continuity of care of adolescents with chronic diseases, in the transfer between paediatric and adult health services. Strategies can be diverse, such as maintaining lines of communication between different service providers, professionals, adolescents and their families. These programmes are usually long-term, beginning during the paediatric follow-up, and involving multidisciplinary teams and families. Preparation for the transition should begin in early adolescence, until discharge from the paediatric service, and adolescents should feel well established in the adult heath care setting. Approaches may include educational interventions targeting health care providers, the adolescents and their families, either alone or in combination. They may also involve structural changes such as the creation of specific clinics for adolescents.

Option 3. Promotion of self-care with adolescents

Self-care is referred to as 'taking care of yourself, listening to what your body and mind needs, improving lifestyle, avoiding harmful habits, developing healthy eating habits, knowing and controlling the risk factors that lead to illness, taking measures for disease prevention'.[10] In the scientific literature, other terms used are self-efficacy and self-management. Self-efficacy 'refers to individuals beliefs that they are able to control certain events and behaviours in their lives as a way to achieve specific outcomes.'[11] Self-management 'refers to the tasks a patient can perform to minimise the impact of that illness on his/her health status by him-/herself or with the support of a healthcare provider. Self-management of illness requires that a patient has the skills to self-monitor the symptoms and clinical markers of that condition, to understand the associated implications and to adjust medication, treatment or behaviour accordingly.'[12] The term self-management has also been used in publications interchangeably with patient education, patient empowerment, patient coaching, motivational interviewing, integrated disease management, and others. [13]

Table 1. Summary of key findings relevant to the options for health care policies for adolescents with sickle cell disease.

	Option 1 – Use of psychological interventions to improve adherence to treatment	**Option 2 – Implementation of a transition programme from paediatric to adult care**	**Option 3 – Promotion of self-care with adolescents**
Benefícios	Promoting educational[14, 16-18] and multi-component[18] interventions may improve adherence to treatment. Programmes with multiple sessions or familial/parent involvement are more likely to be successful.[16]	Specific educational programmes for chronic disease,[19] transition programmes for patients with sickle cell disease (SCD),[20] and educational intervention to improve the transition[21] may improve the transition readiness of the young people, contributing to improve self-management and to adjust the adult health services.	Positive relation between self-efficacy and health outcomes in young people with SCD.[11] Mobile phone messaging has a positive impact on the health status and self-management of people with chronic diseases.[12] The Stanford chronic disease self-management programme, in the United States, shows better health status outcomes.[13]
Potential harm	Not reported.	Not reported.	Mobile phone messaging may contain inaccurate data. Messages may be misinterpreted or misunderstood, including reading difficulties. There may be lack of understanding or misinterpretation of the information, and difficulties in reading. There is a risk of replacing care delivery with mobile phone messaging.[12]
Costs and/or cost-effectiveness	Necessary costs should be considered when involving different professional categories and digital information technologies.	Programmes involving a multidisciplinary team, what may have economic implications on their implementation.	There may be additional costs, as backup systems may be needed.[12]

	Option 1 – Use of psychological interventions to improve adherence to treatment	**Option 2 – Implementation of a transition programme from paediatric to adult care**	**Option 3 – Promotion of self-care with adolescents**
Uncertainty and knowledge gaps	Most of the studies included in the systematic reviews reported positive results of interventions focusing on individuals with chronic diseases in general, not specifically SCD, and these results may be useful in supporting adolescents with SCD, since these diseases have in common the beginning in early childhood.	Not all studies have shown successful results in every intervention.[19] Educational programmes may be insufficient to improve transitional readiness.[21]	Limitations in studies regarding small sample size and loss to follow-up.[11] Individuals may lose interest in these interventions in the long-term. Some gaps were found in the studies, such as a period not adequate to assess the interventions and lack of information on costs, risks and long-term effects of such interventions.[12] The Stanford Programme have not improved some aspects, such as number of doctor visits, emergency department visits and hospitalizations.[13]
Applicability in different contexts	Not reported.	A wide range of transition programmes being developed in different countries.[21]	Mobile phone messaging, which is better used by adolescents and young people, to support the treatment of different diseases has been studied in different locations.[12] The Stanford Programme, since the 1990s, has been providing care for people in 36 countries.[22]
Perceptions and experiences	Not reported.	Not reported.	Diabetic patients were satisfied with mobile messaging to improve self-management.[12]

Considerations on barriers for implementation of the options

Treatment non-adherence in chronic diseases is a well-studied phenomenon, and it can be as high as 75% for adolescents and young adults.[14]

Implementation of options should take into consideration that patients with genetic diseases generally go to specialty care services at the expense of primary care. In this sense, it may be difficult for adolescents with sickle cell disease and their families to bond with primary care professionals. Therefore, primary care professionals need to be adequately trained to embrace, support and guide these adolescents and their families. Adult health care services should count with haematologists trained to meet the specific needs of young people with sickle cell disease. The proper functioning of the referral and counter-referral system between services with different levels of complexity is also of paramount importance to achieve comprehensive care for adolescents with sickle cell disease.

Table 2 shows some barriers and possibilities to overcome them. Brazilian studies on health services and professionals, and on adolescents with chronic diseases, in particular those with sickle cell disease, are considered important references to understand the barriers to the implementation of these options.

Table 2. Considerations on implementation of the options for health care policies for adolescents with sickle-cell disease.

	Option 1 – Use of psychological interventions to improve adherence to treatment	**Option 2 – Implementation of a transition programme from paediatric to adult care**	**Option 3 – Promotion of self-care with adolescents**
Patients / individuals / decision makers	It is important that children and adolescents become a part of the conversation with parents and doctors, not only a listener.[23] Psychotherapeutic support can be helpful and has been well accepted in dealing with school and work difficulties,[24] but it is important to ensure that psychotherapy would not be an excuse to stop medical treatment.[17]	The challenges faced by these programmes are related to determining the readiness of adolescents with chronic diseases to transfer to adult services, and tailoring services to their needs. Primary health care services could be the place for embracement and support, but they are not yet recognized as health care providers for people with sickle cell disease (SCD).[25]	Lack of psychosocial preparation of young people to cope with the disease can lead to serious health complications, resulting in hospitalizations and loss of confidence in the health system.[11] Interventions with mobile phones are best used by adolescents and young people, contributing to self-care. Especially for men, mobile phones can improve access to health professionals.[12]
Health workers	Education through verbal or written information can contributed to the understanding of the nature of the disease, the reasons and benefits of adhering to the proposed treatment.[17] It is recommended to establish routines that facilitate introducing the user into communicative acts since the beginning of treatment.[23] Primary health care teams are a point of reference for families of individuals with chronic diseases, but they are not prepared to support people with SCD.[25] Nursing staff need to be properly guided and informed about the disease.[26] Education, counselling and the establishment of mutual trust between the health care providers and the individual are crucial, thus, health professionals need specialized training.[27]	A study conducted in the State of Minas Gerais, Brazil, indicated that the primary health care team does not feel responsible for coordinating the health care of people with SCD, delegating to the family the search for the best healthcare services.[28] One of the main challenges reported in the transition programmes was the lack of haematologists specializing in SCD in adult clinics.[29]	Lack of knowledge on the part of doctors and health professionals about SCD, associated with stigmatization, can affect self-care of people with SCD.[11] Professional teams understand the basic principle of self-care theory, but they need to improve their knowledge on SCD.[31]
Organization of health care services	The Family Health Strategy is considered the primary access to health care services for people with chronic diseases (including SCD), their families and caregivers. However, the primary health care team is only sought in case of examinations, consultations and clinical complications.[28]	In order to achieve an optimal model of chronic disease care, it is necessary to: Reduce the waiting lists; provide care at the right time; improve both communication and collaboration among professionals; strive for consistency in the messages conveyed to individuals; provide easier access to medical records; rely on dedicated professionals who know how to listen and know the contexts in which patients live; consider medication costs and other expenses; discuss with users the different treatment options and support services available.[30]	The Stanford chronic disease self-management programme has been successful and shown great expansion both inside and outside the United States. However, these evidence-based programmes have the challenge of maintaining their standardization when used on a large scale.[22]

	Option 1 – Use of psychological interventions to improve adherence to treatment	Option 2 – Implementation of a transition programme from paediatric to adult care	Option 3 – Promotion of self-care with adolescents
Health systems	Treatment of SCD should not be provided only at a blood centre.28 After diagnosis in the neonatal period, children are referred to specialized centres, making it difficult for families to bond with primary health care services.	Genetic diseases, in general, do not have defined guidelines in primary care. The focus has been on specialized care, and there are few guidelines on how to structure primary care to deal with these diseases. There are no care protocols, and this may result in access difficulties, as professionals do not know their role in the delivery of care for people with SCD.[28]	Life expectancy of people with SCD has increased and, consequently, more individuals experience the transition from child to adult treatment.[11] The United Kingdom government, in 2005, recognized support for self-care as one of the three pillars of the National Health Service (NHS).[12]

Considerations on equity of the options

Decision on the options to be implemented should take into account how much they may or may not negatively affect inequalities between population groups. In implementing Option 1, some groups may be disadvantaged due to the difficulty in accessing services that offer educational or behavioural activities, particularly in towns with a smaller health care structure that do not count with a multiprofessional team.

The organization of the health system in the city of São Paulo to assist people with sickle cell disease is likely to increase inequalities in the transition from paediatric to adult care, as only five of the eight centres provide adult care. Therefore, the implementation of a transition programme, as presented in Option 2, could contribute to a comprehensive care for adolescents with sickle cell disease if the transition programme would promote a growing approach between blood centres and the Basic Health Units, through an adequate referral and counter-referral system. However, in order for this to take place, it is necessary to make arrangements to train primary care teams on the embracement, follow-up and support of these people, as well as to monitor and evaluate on an ongoing basis, in accordance with indicators, the implementation of this type of health care network.

As for the promotion of self-care addressed in Option 3, this depends on the embracement by health professionals, the offer of multiprofessional care that includes emotional care, as well as the skills to communicate effectively with adolescents. It would be necessary to think about the organization of these services in a way that does not make difficult the access to guidance and support for the development of self-care.

In any of the options proposed to improve the care of adolescents with sickle cell disease, the phenomenon of institutional racism is an important barrier to achieving equity. Institutional racism has been defined by the Programme to Combat Institutional Racism as 'the failure of institutions and organizations to provide a professional and adequate service to people due to their colour, culture, racial or ethnic origin. It is manifested in discriminatory norms, practices and behaviours adopted in the everyday work routine, which result from ignorance, lack of attention, prejudice or racist stereotypes. In whatever case, institutional racism always places people from racially or ethnically discriminated groups in a situation of disadvantage in access to the benefits generated by the State and by other institutions and organizations.'[15]

Contributions of the deliberative dialogue

A deliberative dialogue meeting was carried out after the preparation of the evidence brief for policy, with the participation of different stakeholders who may be involved in or affected by future decisions on the implementation of the options proposed. This meeting contributed to better understanding of the complexity of the problem, the lack of research on the issue, and the importance of including the user in the process, to address the options from different perspectives and experiences, and to think how Primary Health Care could be more effective and promote a growing approach to the referral centres. This problem should be analysed in the context of multiple causes, considering the following aspects: transition process; primary care, specialty care and their collaboration; quality of care in health services; communication and specificity of the care of adolescents; and issues on race / colour and institutional racism.

Regarding the options developed, stress was laid on the need for intersectoriality and the importance of considering the specificities of adolescents; the relationship with the health professionals and services, family, school and user networks, and advocacy. Barriers to the implementation of the options were addressed, and possibilities to overcome them were suggested, such as to improve quality of care in health services; to invest in primary care as a fundamental component of the care of people with sickle cell disease; to train health professionals; to promote articulation, especially the relationship of Primary Health Care to the management levels of SUS, for the implementation of public policy; and to overcome the difficulties caused by institutional racism. The structured dialogue on the issue made it possible to learn more about sickle cell disease, and contribution to the evidence brief would not have been possible without a methodology that includes different perspectives.

Conclusions:

Transition in the follow-up from child to adult health care services is a major problem for young people with sickle cell disease, and a great challenge for managers and health professionals. Policy options that are effective involve educational processes, psychological and social support, gradual transition from paediatric to adult care services, and investment in promotion of self-care. An extensive health care network, with integrated primary and specialty care services, is fundamental to ensure the continuity of care and support received during childhood. Investing to disseminate knowledge to the community at large and to train primary health care professionals is a necessary and urgent task, including to reduce prejudice against the disease and the people directly and indirectly affected by it. Important steps have already been taken within the limits of knowledge, such as early diagnosis and recommended medicines and procedures for prevention and treatment of sickle cell disease available at SUS. In conclusion, it has been found that research on this topic are scarce, and, on the other hand, there is a need to improve the methodological quality of studies on it.

Appendix – Characteristics of systematic reviews and their contribution in formulating policy options for adolescents with sickle cell disease.

Author, year	Aim	Key components of interventions	Number and type of primary studies included	AMSTAR
Systematic reviews for option 1 – Use of psychological interventions to improve adherence to treatment				
De Jongh et al., (2012)[12]	To check the characteristics of psychological interventions, effective interventions, and the common characteristics of effective interventions.	Psychological interventions for adolescents and young adults with chronic diseases.	Of 25 studies included, 13 referred to diabetes, seven to cancer, two to juvenile rheumatoid arthritis, one to asthma, one to sickle cell disease (SCD), one to asthma and SCD.	4/11
Pai et al., (2014)[14]	To analyse the effectiveness of psychological interventions.	Psychological interventions to promote treatment adherence in children, adolescents and young adults.	Of 26 studies included, 23 were included in the meta-analysis (n = 3,898 participants).	7/11
Dean et al., (2010)[17]	To analyse interventions that can improve adherence to medication.	Psychological interventions to improve adherence to medication.	Of 17 trials included, three involved children, five adolescents and nine children and adolescents. The conditions studied were tuberculosis, asthma, diabetes, HIV, oral contraception, epilepsy, juvenile rheumatoid arthritis, SCD, kidney transplantation and depression.	5/11
Kahana et al., (2008)[18]	To estimate the effectiveness of psychological interventions to promote treatment adherence for chronic health conditions in children.	Psychological interventions to promote treatment adherence for chronic health conditions in children.	Of 70 studies included, 29 were randomized controlled trials; 32 studies were on asthma, 16 on diabetes, 10 on cystic fibrosis, two on juvenile rheumatoid arthritis, two on obesity, and one on haemodialysis, haemophilia, HIV, irritable bladder, phenylketonuria, seizure disorders, SCD and tuberculosis. The age of participants in these studies ranged from 2 to 15 years old.	3/11
Systematic reviews for option 2 – Implementation of transition programmes from paediatric to adult care				
Crowley et al., (2011)[19]	To analyse the effectiveness of transitions-of-care interventions in improving health outcomes across a wide range of conditions (any chronic physical or mental illness or disability).	Transition Programme.	Ten studies were included, showing a great variability in populations, interventions and outcomes. Eight studies on diabetes mellitus, one cystic fibrosis and one kidney transplantation.	4/11

Author, year	Aim	Key components of interventions	Number and type of primary studies included	AMSTAR
Systematic reviews for option 2 – Implementation of transition programmes from paediatric to adult care				
Jordan et al., (2013)[20]	Analisar abordagens bem-sucedidas de transição.	Programa de transição.	Selecionados 14 artigos sobre processo de transição para doença falciforme; e seis artigos sobre doenças crônicas.	5/11
Campbell et al., 2016[21]	To assess the effectiveness of interventions to improve the transition of adolescent from paediatrics to adult health care services.	Interventions to improve the transition of care for adolescents with chronic diseases.	Four studies were included, with 238 participants, all adolescents aged 16 to 18 years, who had cystic fibrosis, inflammatory bowel disease, type 1 diabetes, heart disease or spina bifida.	11/11
Systematic reviews for option 3 – Promotion of self-care with adolescents				
Molter e Abrahamson, (2015)[11]	To investigate how self-efficacy during the transition period impact on health outcomes of the population living with SCD.	Relation among self-efficacy, transition and health outcomes.	20 studies included. Most involved adolescents, but in general the age of participants in these studies ranged from 6 to 56 years old. The themes vary from SCD to cancer to other chronic diseases.	5/11
De Jongh et al., (2012)[12]	To assess the effects of mobile phone messaging interventions, designed to facilitate self-management of chronic diseases.	Interventions with mobile phone messaging in support to the treatment of chronic diseases.	Four studies included involving 182 participants, including adolescents and young adults, two on diabetes, one on hypertension, and one on asthma.	11/11
Franek, (2013)[13]	To identify clinical effectiveness, cost-effectiveness, and evidence-based guidelines associated with self-management programmes and interventions to treat patients with chronic diseases.	Self-management programmes and interventions for patients with chronic diseases.	Review of systematic reviews and primary studies; a total of 22 studies included. Different chronic diseases were addressed, such as osteoarthritis and pulmonary diseases.	Not applicable

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References

1. *Ministério da Saúde (BR). Secretaria de Atenção à Saúde. Departamento de Atenção Hospitalar e de Urgência. Doença falciforme: diretrizes básicas da linha de cuidado* [Internet]. 2015 [accessed 31 March 2016]. Available at: http://bvsms.saude.gov.br/bvs/publicacoes/doenca_falciforme_diretrizes_basicas_linha_cuidado.pdf.
2. Cançado, RD, Jesus JA. *A doença falciforme no Brasil. Revista Brasileira de Hematologia e Hemoterapia* [Internet]. 2007 [accessed 8 Setember 2016]; 29(3). Available at: http://www.scielo.br/pdf/rbhh/v29n3/v29n3a02.pdf.
3. *Ministério da Saúde (BR). Secretaria de Ciência, Tecnologia e Insumos Estratégicos. Departamento de Gestão e Incorporação de Tecnologias em Saúde. Hidroxiureia para crianças com doença falciforme - Relatório n° 57* [Internet]. 2013 [accessed 5 December 2013]. Available at: http://portal.saude.gov.br/portal/saude/Gestor/area.cfm?id_area=1611.
4. *Ministério da Saúde (BR). Secretaria de Atenção à Saúde. Secretaria de Ciência, Tecnologia e Insumos Estratégicos. Portaria Conjunta nº 05 de fevereiro de 2018. Aprova o Protocolo Clínico e Diretrizes Terapêuticas da Doença Falciforme* [Internet]. 2018 [accessed 16 April 2019]. Available at: http://portalarquivos2.saude.gov.br/images/pdf/2018/fevereiro/22/Portaria-Conjunta-PCDT-Doenca-Falciforme.fev.2018.pdf.
5. *Batista TF. Con[vivendo] com a anemia falciforme: o olhar da enfermagem para o cotidiano de adolescentes [dissertação]. Salvador: Universidade Federal da Bahia*, 2008 [accessed 8 September 2016]. Available at: http://www.ccadoentescola.faced.ufba.br/arquivos/tatianafancobatista.pdf.
6. Bryant R, Walsh T. Transition of the chronically ill youth with hemoglobinopathy to adult health care: an integrative review of the literature. J of Pediatric Health Care. 2009; 23(1):37-48.
7. Fegran L. et al. Adolescents' and young adults' transition experiences when transferring from paediatric to adult care: a qualitative metasynthesis. Int J of Nursing Studies, Waltham. 2014; 51(1):123-135.
8. Lavis JN, Oxman AD, Lewin S, Fretheim A. *Ferramentas SUPPORT para a elaboração de políticas de saúde baseadas em evidências (STP)* [Internet]. 2009 [accessed 16 April 2019]. Available at: http://sintese.evipnet.net/livro/ferramentas-support-para-a-elaboracao-de-politicas-de-saude-baseadas-em-evidencias-stp/.
9. Shea BJ, Grimshaw JM, Wells GA, Boers M, Andersson N, Hamel C, et al. Development of AMSTAR: a measurement tool to assess the methodological quality of systematic reviews. BMC Medical Research Methodology [Internet]. 2007 [accessed 18 December 2015]; 7(10). Available at: http://www.ncbi.nlm.nih.gov/pmc/articles/PMC1810543/pdf/1471-2288-7-10.pdf.
10. *Ministério da Saúde (BR). Secretaria de Atenção à Saúde. Departamento de Atenção Especializada. Manual de Educação em Saúde / Ministério da Saúde, Secretaria de Atenção à Saúde, Departamento de Atenção Especializada* [Internet]. 2008 [accessed 8 September 2016]. Available at: http://bvsms.saude.gov.br/bvs/publicacoes/manual_educacao_saude_volume1.
11. Molter BL, Abrahamson K. Self-efficacy, transition, and patient outcomes in the sickle cell disease population. Pain Manag Nurs. 2015; 16(3):418-424.
12. De Jongh T. et al. Mobile phone messaging for facilitating self-management of long-term illnesses. Cochrane Database of Systematic Reviews. 2012.
13. Franek J. Self-management support interventions for persons with chronic disease: an evidence-based analysis. Ont Health Technol Assess Ser [Internet]. 2013 [accessed 6 July 2016]; 13(9). Available at https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3814807/pdf/ohtas-13-60.pdf.
14. Pai AL, Mcgrady M. Systematic review and meta-analysis of psychological interventions to promote treatment adherence in children, adolescents, and young adults with chronic illness. Journal of Pediatric Psychology [Internet]. 2014 [accessed 6 September 2016]; 39(8). Available at: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4192048/pdf/jsu038.pdf.
15. López LC. *O conceito de racismo institucional: aplicações no campo da saúde. Interface - Comunic. Saude, Educ* [Internet]. 2012 [accessed 8 September 2016]; 16(40). Available at: http://www.scielo.br/pdf/icse/v16n40/aop0412.pdf.

16. Sansom-Daly UM. et al. A systematic review of psychological interventions for adolescents and young adults living with chronic illness. Health Psychology. 2012; 31(3):380-393.
17. Dean AJ, Walters J, Hall A. A systematic review of interventions to enhance medication adherence in children and adolescents with chronic illness. Archives of Disease in Childhood [Internet]. 2010 [accessed 8 September 2016]; 95(9). Available at: http://adc.bmj.com/content/95/9/717.full.pdf+html.
18. Kahana S, Drotar D, Frazier T. Meta-analysis of psychological interventions to promote adherence to treatment in pediatric chronic health conditions. Journal of Pediatric Psychology [Internet]. 2008 [accessed 6 July 2016]; 33(6). Available at: http://jpepsy.oxfordjournals.org/content/33/6/590.full.pdf+html.
19. Crowley R. et al. Improving the transition between paediatric and adult healthcare: a systematic review. Archives of Disease in Childhood. 2011; 96(6): 548-553.
20. Jordan L, Swerdlow P, Coates TD. Systematic review of transition from adolescent to adult care in patients with sickle cell disease. Journal of Pediatric Hematology/Oncology. 2013; 35(3):165-169.
21. Campbell F. et al. Transition of care for adolescents from paediatric services to adult health services. Cochrane Database of Systematic Reviews. 2016.
22. Lorig K. Chronic Disease Self-Management Program: Insights from the Eye of the Storm. Frontiers in Public Health [Internet]. 2015 [accessed 6 September 2016]; 2(253). Available at: http://www.ncbi.nlm.nih.gov/pmc/articles/PMC4410327/pdf/fpubh-02-00253.pdf.
23. Oliveira VZ, Gomes WB. *Comunicação médico-paciente e adesão ao tratamento em adolescentes portadores de doenças orgânicas crônicas. Estudos de Psicologia* [Internet]. 2004 [accessed 6 September 2016]; 9(3). Available at: http://www.scielo.br/pdf/epsic/v9n3/a08v09n3.pdf.
24. Silva RBP, Ramalho AS, Cassorla RMS. *A anemia falciforme como problema de Saúde Pública no Brasil. Revista de Saúde Pública* [Internet]. 1993 [accessed 8 September 2016]; 27(1). Available at: http://www.scielo.br/pdf/rsp/v27n1/09.pdf.
25. Gomes LMX, Barbosa TLA, Vieira EDS, Caldeira AP, Torres HC, Viana MB. Perception of primary care doctors and nurses about care provided to sickle cell disease patients. *Revista Brasileira de Hematologia e Hemoterapia* [Internet]. 2015 [accessed 6 July 2016]; 37(4). Available at: http://www.scielo.br/pdf/rbhh/v37n4/1516-8484-rbhh-37-04-00247.pdf.
26. Kikuchi BA. *Assistência de enfermagem na doença falciforme nos serviços de atenção básica. Revista Brasileira de Hematologia e Hemoterapia* [Internet]. 2007 [accessed 8 September 2016]; 29(3). Available at: http://www.scielo.br/pdf/rbhh/v29n3/v29n3a27.pdf.
27. Lobo C, Marra VN, Silva RMG. *Crises dolorosas na doença falciforme. Revista Brasileira de Hematologia e Hemoterapia* [Internet]. 2007 [accessed 8 September 2016]; 29(3). Available at: http://www.scielo.br/pdf/rbhh/v29n3/v29n3a11.pdf.
28. Gomes LMX, Pereira IA, Torres HC, Caldeira AP, Viana MB. *Acesso e assistência à pessoa com anemia falciforme na Atenção Primária. Acta Paulista de Enfermagem* [Internet]. 2014 [accessed 6 July 2016]; 27(4). Available at: http://www.scielo.br/pdf/ape/v27n4/1982-0194-ape-027-004-0348.pdf.
29. Sobota A, Neufeld EJ, Sprinz P, Heeney MM. Transition from pediatric to adult care for sickle cell disease: results of a survey of pediatric providers. Am J Hematol [Internet]. 2011 [accessed 8 September 2016]; 86(6). Available at: http://onlinelibrary.wiley.com/doi/10.1002/ajh.22016/epdf.
30. Sav A, McMillan SS, Kelly F, King MA, Whitty JÁ, Kendall E, et al. The ideal healthcare: priorities of people with chronic conditions and their carers. BMC Health Services Research [Internet]. 2015 [accessed 31 March 2016]; 15. Available at: http://bmchealthservres.biomedcentral.com/articles/10.1186/s12913-015-1215-3.
31. Santana CA, Cordeiro RC, Ferreira SL. *Conhecimento de enfermeiras sobre educação para o autocuidado na anemia falciforme. Rev Baiana de Enfermagem* [Internet]. 2013 [accessed 8 September 2016]; 27(1). Available at: http://www.portalseer.ufba.br/index.php/enfermagem/article/view/6742/6783.

Evidence brief for health policy-making: prevention and control of diabetic foot in primary health care

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Abstract

This is an Evidence Brief whose priority problem was: prevention and control of Diabetic Foot (DF) in Primary Health Care (PHC). We used the SUPPORT tool (Supporting Policy Relevant Reviews and Trials) of the EVIPNet (Evidence-Informed Policy Network). The search for options occurred in February 2016 and was updated in October 2018, in the Virtual Health Library, Health Systems Evidence, PubMed and Cochane Library. The search strategy used the terms 'diabetic foot' and 'prevention'. The search resulted in 41 texts and 16 persisted after deletion of duplicates. After reading in full, 11 remained. Four options for coping with the problem were identified: a) to train PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus; b) to stimulate self-care by means of daily temperature measurement of the foot; c) to identify and perform the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network; d) to offer packages of complex interventions. The barriers to the strategies were: non-adherence of patients to self-care, overload and lack of motivation of professionals, and weaknesses of the care network. DF is an important public health problem that must be prioritized on the agenda of decision makers.

Keywords: Primary Health Care; Diabetic Foot; Prevention; Evidence-Informed Policy.

Introduction

Diabetic foot is characterized by a multifactorial syndrome in people with diabetes, which has a negative impact on the quality of life, and can cause from chronic ulcerations to lower limb amputations.[1,2]

It has been demonstrated that 50% of amputations and ulcerations may be prevented through early diagnosis, adequate treatment, and comprehensive management of diabetes.[1,3] However, mortality among diabetics related to these amputations is estimated to be 19%, and the survival is 65% in three years and 41% in five years.

Diabetic foot ulcer is one of the major health problems in many countries, causing great socio-economic impact.[4] This has put pressure on the health system by increasing demand for care and rising costs,[1] and also on the social security system due to loss of working days and early retirement.[3]

Overall, the risk of lower limb amputation in patients with diabetes mellitus (DM) is approximately 40 times higher than in the general population, and it is more common among populations with lower socio-economic status, living under poor hygiene conditions and having poor access to health services, indicating iniquities.[5,6] Another iniquity that can be pointed out is the fact that while in high-income countries peripheral arterial disease (PAD) is the leading cause of amputation in diabetics, in low- and middle-income countries it is infection.[7]

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In Brazil, the prevalence of amputation is 0.7% and 2.4%. The Brazilian National Health Survey (PNS, in Portuguese) in 2013 showed that among diabetic patients with foot ulcers, only 29.1% underwent a foot examination in the last twelve months. Therefore, as it is the case worldwide, with disparities between low- and high-income countries, there are also internal disparities between regions of Brazil, where 33.8% of diabetic patients in South-East stated that they had their feet examined by health professionals in last twelve months, while only 22.1% in North-East.[2,8]

Consequently, diabetic foot remains under-diagnosed and undertreated, when not forgotten, because of an ineffective longitudinal care of patients with DM, even with the expansion of Primary Health Care (PHC) and the emphasis on clinical management of patients with chronic diseases.

Moreover, this means that people have insufficient access to primary health care, as well as non-adherence to continuity of care throughout life. These factors affect negatively the treatment for DM, including prevention and early detection of ulcers, and their appropriate management.[4]

In this context, we considered relevant to address this issue as a priority problem of an evidence brief for policy, which was developed within the scope of the Professional Master's Degree Programme in Family Health of the North-East Network of Training in Family Health, Regional University of Cariri, Brazil. This article aims to present some results of the evidence brief, including the policy options identified, and possible barriers and strategies for their implementation, in order to support managers and professionals in the development of evidence-informed policy for prevention and control of diabetic foot in PHC.

Method

Evidence-Informed Policy Network (EVIPNet) methodology was used, based on the SUPPORT tool (Supporting Policy Relevant Reviews and Trials)[9] for systematising the search, and evaluating, adapting and applying the results of scientific research in the formulation of health policies. The following steps were taken: Defining the problem / issue; searching for evidence; delineating the options; identifying barriers and strategies for the implementation of the options; and making considerations on equity.

The search for studies occurred in February 2016 and was updated in October 2018, in the following databases: Virtual Health Library (VHL), Health Systems Evidence (HSE), PubMed and Cochane Library.

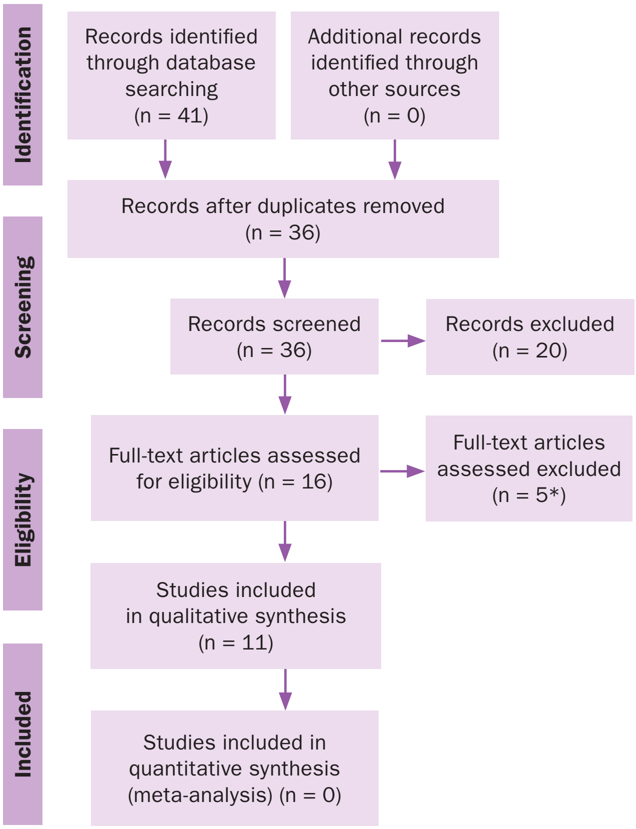
For the search strategy, we used the terms 'diabetic foot' and 'prevention', in Portuguese and English, according to the specificity of each database and filters for systematic review and full texts. The selection, quality assessment and data extraction were carried out by two reviewers.

The quality of systematic reviews was assessed using the AMSTAR (Assessment of Multiple Systematic Reviews) tool,[11] and there was no threshold score for study exclusion.

After full-text reading, possible interventions for the problem of prevention and control of diabetic foot in PHC were identified, as well as potential barriers and strategies for implementing these interventions.

Results and discussion

In total, 41 studies were identified, of which 16 were eligible, and 11 were included for the policy options. Five studies were excluded for addressing treatment, but not prevention of diabetic foot. The selection process is presented in Figure 1. Of the included studies, seven were systematic reviews and four economic evaluation studies. Concerning the methodological quality of systematic reviews, two were rated as moderate quality (AMSTAR score 6/11) and five as high quality (AMSTAR score 9-11/11).



* Articles excluded for addressing only treatment, but not prevention of diabetic foot.

Four key strategies for prevention and control of diabetic foot in PHC were identified: 1) to train PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus; 2) to stimulate self-care by means of daily temperature measurement of the foot; 3) to identify and perform the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network; 4) to offer packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems.

Table 1 provides a detailed description of each policy option regarding benefits, potential costs and uncertainties. Table 2 provides potential barriers and facilitators to the implementation of each of the four options, taking into account the dimensions of patients / individuals, health care providers, organization of services, and health system.

Table 1. Options for coping with the problem of prevention and control of diabetic foot in PHC.

Options	**Option 1: Training PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus**	**Option 2: Stimulating self-care by means of daily temperature measurement of the foot**	**Option 3: Identifying and performing the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network**	**Option 4: Offering packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems**
Benefits	The most effective preventive measure for amputation prevention is screening with referral to a foot care referral service;[11, 12] Education on foot care, as part of education for patients with diabetes, seems effective in preventing ulceration.[10,11,17]	Foot temperature monitoring provides meaningful insight into the risk of ulceration for both first and recurrent ulcers.[10,13]	Foot care alone or in a multidisciplinary context, including removal of calluses, cracks, nail trimming, treatment of onychomycosis, prescription of therapeutic footwear, and guidance on footwear, has proved effective in reducing ulceration and amputation.[10,14,11,15]	Reducing the incidence of ulcers in diabetes mellitus patients requires an intensive, integrated approach that combines more than one prevention strategy, especially in patients at high risk for foot ulceration; Individual prevention strategies alone have been ineffective in clinical practice.[16]

Options	**Option 1: Training PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus**	**Option 2: Stimulating self-care by means of daily temperature measurement of the foot**	**Option 3: Identifying and performing the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network**	**Option 4: Offering packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems**
Potential Costs	Screening of risk reduces costs related to wound care and amputations;[11,17] Diabetic patient education for self-care results in cost savings.[19.]	Daily temperature measurement of the foot by an infrared thermometer must be evaluated further through cost effectiveness and efficiency studies, as it implies individual use of specific equipment.[10]	Economic evaluation studies on foot care have shown that podiatric care to prevent lesions and amputations are clearly cost-effective or even cost-saving.[17]	Comprehensive foot care to prevent ulcers reduces cost compared to usual care with only one intervention.[18,19]
Uncertainties	It is not possible to draw firm conclusions about the effects of educational programmes for prevention of amputations because the interventions evaluated are packages of both care and education;[17] There is no consensus on the best education strategy for the diabetic foot patient; studies were rated as weak strength of recommendation, with a high risk of bias, and this intervention have always been evaluated in packages with other interventions.[10,17.]	Daily temperature measurement of the foot contribute to the general examination of the foot by the patient, possibly leading to the identification of other risk factors and, consequently, to seek health services early; this preventive strategy was not evaluated individually in the studies; the implementation of this strategy is not well known in any health care services.[10.]	Studies are quite heterogeneous concerning samples and interventions on podiatric care.	There is little evidence of effectiveness of complex interventions, as key methodological components are often lacking in systematic reviews, such as assessment of risk of bias.

Table 2. Potential barriers and facilitators to the implementation of the options, across dimension levels.

Levels	**Option 1: Training PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus**	**Option 2: Stimulating self-care by means of daily temperature measurement of the foot**	**Option 3: Identifying and performing the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network**	**Option 4: Offering packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems**
Patients / individuals / decision makers	Non-adherence to behaviour changes as part of the educational process.[10]	Non-adherence of patients to daily temperature measurement of the foot.[13,10]	There may be poor adherence of patients to adapted footwear indicated.[14]	Patients' difficulty in accessing referral services; non-adherence of patients to activities that involve lifestyle changes and specific tasks.[20] Prescription for individualized care improves patient adherence, as well as the use of rewards.

Levels	**Option 1: Training PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus**	**Option 2: Stimulating self-care by means of daily temperature measurement of the foot**	**Option 3: Identifying and performing the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network**	**Option 4: Offering packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems**
Health workers	Resistance from professionals because they see screening and guidance to self-care as additional tasks;[11] difficulty of communication among professionals from different levels of the care network.[2] Professionals should be motivated to participate in actions that involve changes in care delivery;[20] Reorganization of professionals' work in order to prevent overload;20 Financial incentives can be offered to professionals to meet their goals;[9] Communication between levels of the care network should be ensured, which could range from exchange of reports and opinions to direct communication by phone calls to case discussion.[2]	Lack of training of professionals to provide guidance on the correct use of the device, as well as lack of availability of professionals to monitor its use by patients;[10,13] Professionals should be qualified and motivated to participate in actions that involve changes in care delivery;[20] Reorganization of professionals' work in order to prevent overload.[20]	Lack of qualification of professionals for management of pre-ulcerative conditions in the feet of patients with diabetes;[16] Accessible referral services with qualified professionals in podiatric care (in Brazil, there is the estomatherapist nurse) should be ensured.[21]	Lack of qualified and motivated professionals, as well as work overload; Activities of permanent education in health should be developed at work sites; opportunities to provide care to diabetic patients, involving direct care, guidance to self-care and referrals, should be identified in existing activities such as routine consultations and collective activities; Financial incentives can be offered to professionals.
Organization of health care services	Most PHC teams do not perform diabetic foot screening, and most health care networks do not have functioning referral services for diabetic foot;[6,8] PHC should provide screening of risk; the necessary resources for its carrying out should be guaranteed by the health system; referral services should be available to receive these patients; professionals should refer patients to these services; several access opportunities for users should be offered; and there should be an effective counter-referral system.[11,18]	This device is not available in the health system; Further cost effectiveness and efficiency research is need for this option.	There should be a referral multidisciplinary team for diabetes mellitus patients and foot care referral team to support primary care teams.[11,14,15]	For the implementation of a complex intervention, there should be professionals trained in patient care and education, at all points of the continuum of care, and the health system should be organized in such a manner as to facilitate patient adherence to care.[2, 14]

Option 1: Training PHC professionals in the screening of diabetic foot risk, guidance of self-care and coordination of the care of people with diabetes mellitus

In this option, four strategies should be considered for discussion: training process of PHC professionals, patient education for self-care, diabetic foot screening, and coordination of the care of people with DM with a focus on diabetic foot.

Regarding the training process of PHC professionals, the evidence refers to permanent health education with a focus on meaningful, participatory and sustainable learning. Professionals should be motivated and understand the importance of the problem, so, based on that, strategies of permanent, sustainable and effective education should be developed.[23]

This model of professional education is even more important because, as a barrier to health professionals, they see screening and guidance to self-care as additional tasks. Therefore, it is assumed that the understanding of the magnitude of the problem through the meaningful learning improves the adherence of health professionals to this strategy.[26,27]

Regarding diabetic patient education for self-care, this should be aimed at improving the knowledge and behaviour in the diabetic foot care, providing support to the patient to adhere to the guidelines.[28]

The barrier related to the patients, then, would be non-adherence to self-care-oriented processes. Patients who do not adhere to the information received in educational programmes are at higher risk of developing foot ulcers than those who adhere to the guidelines.[30] Other barriers are related to skills, attitudes and motivation for change, adherence, and also access to care.[19,23]

Regarding screening of risk, the practice of using the best evidence in the prevention of diabetic foot is still incipient or not present in PHC services. Supporting these findings, a cross-sectional quantitative study, aiming at evaluating foot care by people with DM and the changes in their lower limbs, in a PHC service, showed that 28 patients (55%) reported that they had never performed self-care activities since the diagnosis of DM.[6]

In regard to barriers related to health services, lack of resources for screening was taken into consideration. However, diabetic foot screening requires low-cost technologies. Thus, to address this issue as a priority problem at the management level, with the redirection of existing resources, may be sufficient to finance this strategy.[23]

Finally, this first option also presents the coordination of care as an integral part of professional training. The offer to patients (especially those with more complex care needs) of necessary guidance to find their way through the health system can only occur if there is coordination of care, that is, the ability to ensure continuity of care across health care teams, with the recognition of the problems that require constant follow-up, as is the case of non-communicable diseases (NCDs) and their complications.[22,23,25]

PHC must be strengthened, so that the continuum of care can be designed according to each reality, taking into consideration offers and demands of the health care networks to assure users of actions and services in an effective, continuous, integral and humanised manner.[24]

Some challenges of the coordination of care by PHC are turnover of professionals, specialised training, low prestige of PHC workers, insufficient provision of specialised care, underfunding of the health system, among others.[22] However, improvements in coordination and quality of care can be achieved with the articulation of health actions and services, so, regardless of where they are delivered, they are synchronized and focused on achieving a common goal.[22,24]

Option 2: Stimulating self-care by means of daily temperature measurement of the foot

Temperature-guided therapy involves daily measurement of food temperature on six foot sites twice daily by the patient or caregiver. The 2°C difference between feet is a sign of inflammation or infection.[29]

This option must be evaluated carefully before being included in health policies and programmes, because one of the reported uncertainties is the fact that studies assessing the feasibility of implementation have not been identified, as the use of this strategy in health services is unknown.[10]

Moreover, economic feasibility must be further evaluated, as this strategy requires the acquisition of a device (infrared thermometer), either by the patient or the health care service.[10]

Indeed, priority interventions for NCD control must meet strict criteria, such as significant health effects; strong evidence of cost-effectiveness; as well as political and financial feasibility for a large-scale provision.

Option 3: Identifying and performing the clinical management of pre-ulcerative conditions and deformities in the feet, with referral to referral services in the care network

The skin manifestations associated with diabetes, such as dry and cracked skin, calluses, skin and nail fungal infection, should be observed and adequately treated for the prevention of ulceration.[28,29,32] Depending on the degree of complication, some of these manifestations can be managed at the level of PHC.[32]

The management of pre-ulcerative conditions in cases of feet at high risk and with deformities may require the assistance of a specialized professional. In Brazil, the professional with this specific training is referred to as the estomatherapist nurse, who works to improve the overall health of the patient, with a focus on primary prevention, diagnosis and treatment of pre-ulcerative lesions.[21]

In countries where podiatry does not exist, and there are no such professionals in the health centre, or they are not enough to provide assistance to all diabetic patients, other health professionals should provide such care through transfer of function.[31,28,32]

Option 4: Offering packages of complex interventions to patients involving strategies of at least two of the three levels: patients, health care providers and health systems

Complex or combined intervention refers to an integrated care approach that includes a combination of two or more prevention strategies in at least two different levels of treatment: patient and / or health care provider and / or health care structure.[17]

Studies have shown that the results of individual prevention strategies alone have been ineffective in clinical practice. Preventive interventions directed at patients, health professionals and / or the health care structure should be prioritized depending on the availability of resources and skills. Due to the complexity related to the problem of diabetic foot, no single strategy can be effective for long-term prevention of foot ulceration and amputation.[17]

Some interventions can directly improve health outcomes of patients, such as education, podiatry care, assessment of risk for foot ulceration, and motivational coach to strengthen foot self-care behaviour (patient-level interventions). Interventions to prevent foot ulceration can also benefit patients indirectly, by improving health professionals' ability to provide adequate care, as well as financial incentives (health care provider-level interventions).[17]

Finally, there are interventions related to the improvement of the health system (health care structure-level interventions). Structural health interventions may include a multidisciplinary approach or measures to improve regular follow-up and continuity of care.[17]

In complex interventions, barriers and strategies to cope with them cumulatively encompass the above-mentioned barriers and strategies of the other three options, regarding patients, professionals and health services.

Conclusions:

Diabetic foot was addressed in this study as a major health problem that should be prioritized in the agenda of decision makers, among the strategies proposed for the control of non-communicable diseases.

In Brazil, there is a lack of specific actions aimed at preventing and controlling diabetic foot complications, and PHC should set the stage for these actions with the implementation of strategies based on the best scientific evidence.

The search for options to address this problem revealed the low level of existing evidence in the primary studies included in the systematic reviews. This limitation indicates a need for further primary studies with methodological rigour, reducing the risk of bias and strengthening evidence-based practice. Moreover, primary studies should be based on previous study protocols in order to reduce heterogeneity, and facilitate meta-analytic approaches in systematic reviews.

Another limitation in this study is related to the primary studies' final reports, which, in general, do not provide detailed information on the interventions implemented. Detailed descriptions of these interventions are necessary to fully understand their content, and facilitate collection of findings in the development of systematic reviews.

Despite these limitations, this study provides important evidence to support policy making for prevention and control of diabetic foot in PHC, such as:

1. Evidence on screening of risk, and patient referral and follow-up based on the screening results, has proved to be the strongest evidence for prevention and control of diabetic foot complications. Most importantly, this option has a low-cost implementation and no potential harm, and screening of diabetic foot risk can be implemented by PHC, in different care contexts;
2. Education for self-care alone will not bring results on the prevention and control of diabetic foot. Besides, there are no studies that prove the best way to provide health education, especially due to the heterogeneity of educational interventions addressed in primary studies;
3. The only scientific evidence reported for self-care of diabetic foot is the temperature-guided therapy. However, inclusion of this practice in patient care programmes needs to be analysed carefully, because cost studies on this practice are scarce and it is not well known in health services yet;
4. Foot care by podiatrists for the diagnosis and treatment of pre-ulcerative conditions can reduce the incidence of ulceration and amputation. The management of pre-ulcerative conditions in cases of feet at low risk can be performed in PHC.
5. Evidence on complex interventions for prevention and control of diabetic foot in PHC is not strong because the included studies have high risk of bias. This should be interpreted as a lack of evidence rather than evidence of no effect. Given the complexity of the condition, complex interventions should be considered in the formulation of health policies and programmes.

Competing interests

The authors declare that they have no competing interests.

References

1. *Federação Internacional de Diabetes*. Clinical Practice Recommendations on the Diabetic Foot [Internet]. 2017 [accessed 3 March 2018]. Available at: file:///C:/Users/Milena3/Downloads/IDF_DF_Foot_CPR_2017_Final.pdf/.
2. *Ministério da Saúde (BR). Secretaria de Atenção à Saúde. Departamento de Atenção Básica. Manual do Pé Diabético: estratégias para o cuidado da pessoa com doença crônica / Ministério da Saúde, Secretaria de Atenção à Saúde, Departamento de Atenção Básica*. 2016:62.
3. Amaral Júnior LAH, et al. *Prevenção de lesões de membros inferiores e redução da morbidade em pacientes diabéticos. Revista Brasileira de Ortopedia*. 2014; 49(5):482–487.
4. *Sociedade Brasileira de Diabetes. Diabetes na Prática Clínica* [Internet]. s/d [accessed 4 April 2016]. Available at: http://www.diabetes.org.br/ebook/component/k2/item/42.
5. Assumpção EC, et al. *Comparação dos fatores de risco para amputações maiores e menores em pacientes diabéticos de um Programa de Saúde da Família. Jornal Vascular Brasileiro*. 2009; 8(2):133-8.
6. Andrade NHS, et al. *Pacientes com diabetes mellitus: cuidados e prevenção do Pé Diabético em atenção primária à saúde. Revista de enfermagem* UERJ, Rio de Janeiro. 2010; 18(4):616-21.
7. *Sociedade Brasileira de Diabetes (SBD). Diretrizes da sociedade brasileira de diabetes*: 2014-2015. 2015:377.
8. *Pesquisa Nacional de Saúde. Acesso e utilização dos serviços de saúde, acidentes e violências: Brasil, grandes regiões e unidades da federação / IBGE, Coordenação de Trabalho e Rendimento*; 2015.
9. Lavis JN, Oxman AD, Lewin S, Fretheim A: SUPPORT Tools for evidence-informed health Policymaking (STP). Introduction. Health Research Policy and Systems [periódico na internet]. 2009 [accessed 10 October 2014]; 7(1). Available at: http://www.health-policy-systems.com/ content/pdf/1478-4505-7-S1-I1.pdf.
10. Netten VJJ, et al. Prevention of foot ulcers in the at-risk patient with diabetes: a systematic review. Diabetes/Metabolism Research and Reviews. 2016; 32(1):84–98.
11. Hunt DL. Diabetes: foot ulcers and amputations. Clinical Evidence. 2012; 8(602):1-44.
12. O'Meara S, et al. Systematic reviews of wound care management: (3) antimicrobial agents for chronic wounds; (4) diabetic foot ulceration. Health Technology Assessment. 2000; 4(21):1-233.
13. Arad Y. Beyond the Monofilament for the Insensate Diabetic Foot: a systematic review of randomized trials to prevent the occurrence of plantar foot ulcers in patients with diabetes. Diabetes Care. 2011:34.
14. Buckley CM, et al. Does contact with a podiatrist prevent the occurrence of a lower extremity amputation in people with diabetes? A systematic review and meta-analysis. British Medical Journal. 2013; 3:1-9.
15. Bertoldi AD, et al. Epidemiology, management, complications and costs associated with type 2 diabetes in Brazil: a comprehensive literature review. Globalization and Health. 2013; 9(62):1-12.
16. Dorresteijn JAN, Kriegsman DMW, Valk GD. Complex interventions for preventing diabetic foot ulceration. Cochrane Database of Systematic Reviews; 2010.
17. Dorresteijn JAN, et al. Patient education for preventing diabetic foot ulceration. Cochrane Database of Systematic Reviews; 2014.
18. Klonoff DC, Schwartz DM. An Economic Analysis of Interventions for Diabetes. Diabetes Care. 2000; 23(3).
19. Drivier VR, et al. The costs of diabetic foot: The economic case for the limb salvage team. Journal of Vascular Surgery. 2010; 52(12):17-22.
20. Rui L, et al. Cost-effectiveness of interventions to prevent and control diabetes mellitus: a systematic review. Diabetes Care. 2010; 33(8):1872-1894.
21. *Ministério da Saúde (BR). Portaria N.0 483, de 2014. Redefine a Rede de Atenção à Saúde das Pessoas com Doenças Crônicas no âmbito do Sistema Único de Saúde (SUS) e estabelece diretrizes para a organização das suas linhas de cuidado. Ministério da Saúde. Gabinete do Ministro*; 2014.
22. *Associação Brasileira de Estomaterapia* [Internet]. s/d [accessed 1 January 2019]. Available at: http://www.sobest.org.br/texto/5.
23. Silocchi C, Junges J. *Equipes de atenção primária: dificuldades no cuidado de pessoas com doenças crônicas não transmissíveis. Trabalho, Educação e Saúde*. 2017; 15(10).
24. Jardim, LV, Navarro, D. *Contribuição da ESF no controle de doenças crônicas não transmissíveis*. J Health Sci Inst. 2017; 35(2):122-6.
25. Schenker, M, Costa, D. *Avanços e desafios da atenção à saúde da população idosa com doenças crônicas na Atenção Primária à Saúde. Ciência & Saúde Coletiva*. 2019 [accessed 20 June 2019]; 24(4):1369-1380. Available at: https://doi.org/10.1590/1413-81232018244.01222019.

26. Peiter CC, Santos JLG, Lanzoni GMM, Mello ALSF, Costa MFBNA, Andrade SR. *Redes de atenção à saúde: tendências da produção de conhecimento no Brasil*
27. Esc Anna Nery. 2019; 23(1).
28. EVIPNet *BRASIL*. *Rede para Políticas Informadas por Evidências* [Internet]. s/d [accessed 1 June 2019]. Available at: http://brasil.evipnet.org/tipo/capacitacao/.
29. Goulart FAA. *Doenças crônicas não transmissíveis: estratégias de controle e desafios e para os sistemas de saúde. Organização pan-americana da saúde / organização mundial da saúde. Ministério da Saúde*. 2011:96
30. International Working Group on The Diabetic Foot. IWGDT Guidelines on the prevention and management of diabetic foot disease; 2019.
31. American Diabetes Association. American Diabetes Association: Standards of medical care in diabetes - 2019. Diabetes Care. 2019; 40(1).
32. Fretheim A, et al. SUPPORT: Tools for evidence-informed health Policymaking (STP). 6. Using research evidence to address how an option will be implemented. Health Research Policy and Systems. 2009; 7(1).
33. *Ministério da Saúde (BR). Secretaria de Atenção à Saúde. Departamento de Atenção Básica. Manual do Pé Diabético: estratégias para o cuidado da pessoa com doença crônica / Ministério da Saúde, Secretaria de Atenção à Saúde, Departamento de Atenção Básica*. 2016:62.

Evidence brief for health policy-making: facing congenital syphilis in primary health care

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Abstract

Introduction: Control of congenital syphilis (CS) is a challenge for professionals and managers of Primary Health Care (PHC). **Objective**: To present the results of the synthesis of scientific evidences by which options for coping with CS were identified. **Method**: The SUPPORT tool (Supporting Policy Relevant Reviews and Trials) suggested by the Evidence-Informed Policy Network (EVIPNet) was used to search for systematic reviews, with the identification of options to confront the problem. The searches were conducted in Virtual Health Library, Cochrane Library, Embase, Health Evidence, Health Systems Evidence, Nice Evidence, Pubmed, Scopus. **Results**: There were 310 studies identified. After selection, we analysed 20 systematic reviews that resulted in a set of options: 1) To expand the offer of screening test for all pregnant women and sexual partners; 2) to perform benzathine penicillin treatment for pregnant women and sexual partners with positive screening test for syphilis; 3) to develop strategies to notify sexual partners of patients with a diagnosis of sexually transmitted infections (STIs); 4) to carry out health campaigns to inform the ways of preventing congenital syphilis; promote educational interventions on condom use, STI prevention, and teenage pregnancy. **Conclusion**: It is expected that the scientific evidence presented in this article may support managers and PHC professionals in the construction of health policies to confront CS.

Keywords: Congenital Syphilis; Primary Health Care; Evidence-Informed Policy

Facing congenital syphilis in Primary Health Care

Congenital syphilis is a disease caused by the bacterium *Treponema pallidum*, which is transmitted from the infected pregnant woman to the foetus because of its transplacental passage, or during the passage of the foetus through the birth canal, when there is an active lesion.[1] Screening and diagnosis of syphilis are based on two main types of serological tests: non-treponemal and treponemal. Non-treponemal tests are relatively inexpensive, easy to perform, sensitive and quantitative. A disadvantage of non-treponemal tests is that false-positive results can be caused by acute viral infections and autoimmune diseases. The diagnosis of syphilis should be confirmed in people with a positive non-treponemal test result by testing with treponemal tests.[2]

In 2011, the Ministry of Health of Brazil determined that the screening of syphilis, especially in pregnant women, should begin with the use of rapid treponemal tests, which provide results within 30 minutes.[3] This strategy can be very useful in supporting PHC professionals to perform early diagnosis and treatment of infected pregnant women in time to prevent congenital syphilis.[4]

In Brazil, all pregnant women should be tested for syphilis at least three times: At the first antenatal visit (ideally in the first trimester of pregnancy); in the third trimester (from week

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28 of gestation); and at the time of delivery or abortion, regardless of previous exams. Sexual partners should also be tested for syphilis and other sexually transmitted infections (STIs).[5]

In regard to treatment, benzathine penicillin was recognized by the 69th World Health Assembly as an essential medicine to control vertical transmission of syphilis, through transplacental transfer. Sexual partners of pregnant women with syphilis may be infected even if they have non-reactive immunological test result; therefore, they should also be treated. In case of child exposed to syphilis during pregnancy, treatment should take into account the mother's clinical and laboratory status.[5]

Although there are simple and low-cost diagnostic and therapeutic resources for syphilis, its control during pregnancy is a challenge because pregnancy is a limited period of time to carry out diagnosis and treatment; STIs can be a difficult subject to discuss, especially during pregnancy; and a general lack of awareness on the part of both the population and health professionals of the magnitude of this health problem and the damage it can cause.[6]

The World Health Organization has launched a strategy to eliminate vertical transmission of syphilis and HIV, based on four pillars: Ensuring sustained political commitment and advocacy; increasing access to, and quality of, maternal and new-born health services; screening and treating pregnant women and their partners; establishing surveillance systems, developing indicators, and strengthening monitoring and evaluation systems.[7]

An important nationwide study from Brazil stated that a small number of congenital syphilis (CS) cases does not necessarily indicate a good programme of vertical transmission control, as cases of CS may be under-reported A large number of cases of CS possibly indicates missed opportunities for intervention due to failures in the notification, diagnosis and proper treatment of pregnant women and their partners.[8] In a meeting held in 2018, based on this study, experts and managers working in STI prevention in primary health care (PHC), in the city of São Paulo, discussed that despite the Ministry of Health's efforts and recommendations for providing screening and treatment in PHC services, CS continues to significantly increase in São Paulo.

In order to make the eradication of CS a reality in Brazil, professional qualification, sensitization, and the standardization of health professionals' conducts are necessary. Furthermore, PHC centres should undergo a supervision process to support the implementation of the recommended guidelines.[9]

We can see that the challenges to reduce the incidence of congenital syphilis cases are many. Therefore, scientific evidence to inform policy options for addressing this problem should be take into consideration.

Objective

This article aimed to present the results of the synthesis of scientific evidences by which options for coping with congenital syphilis in primary health care were identified.

Method

For the preparation of the evidence brief, we used the SUPPORT tool (Supporting Policy Relevant Reviews and Trials) suggested by the EVIPNet (Evidence-Informed Policy Network).[10] This tool aims to reduce the gap between scientific evidence and practices in the health services and policy, through knowledge translation.[11]

The identification of evidence that may contribute to addressing congenital syphilis was carried out by analysing a selection of systematic reviews of effective interventions on this issue.

Systematic reviews are considered as one of the major sources of information for policy-making.[10]

The following databases were searched: Virtual Health Library, Cochrane Library, Embase, Health Evidence, Health Systems Evidence, Nice Evidence, Pubmed, Scopus. The descriptors used were 'syphilis', 'congenital syphilis', 'vertical transmission of infectious disease', 'pregnancy', in Portuguese and English.

After defining the search strategies, the selection process was carried out by two researchers (MSD and EMG), independently, on different dates. Both identified the same studies. First, duplicate studies were excluded, followed by screening of titles and abstracts and selection for full-text review. After full-text reading, the researchers came to a consensus on which studies should be included and conducted the data extraction process. In this phase, a form was filled in for each study included. The quality of the systematic reviews was assessed by the AMSTAR tool - a measurement tool to assess the methodological quality of systematic reviews,[12] and there was no threshold score for study exclusion.

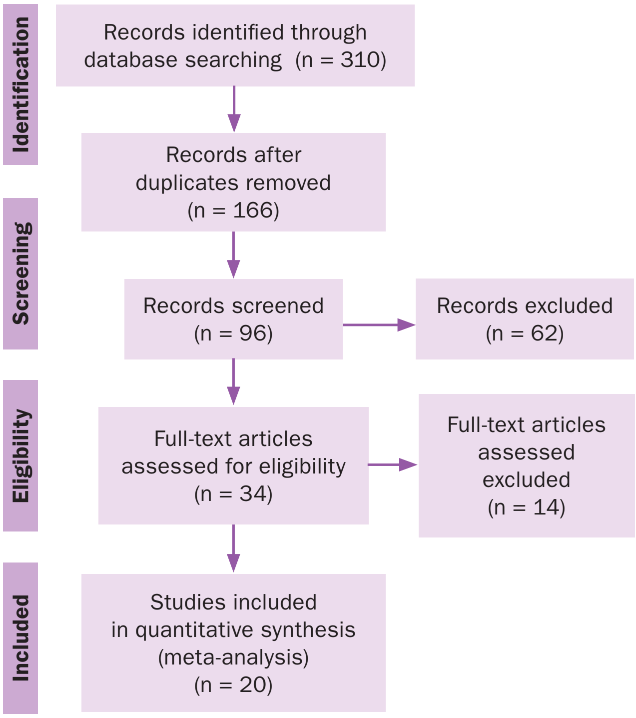
Results

In total, 310 studies were identified, of which 20 were included in this synthesis. Figure 1 shows the article selection process.

It is noteworthy to mention that none of the systematic reviews made direct reference to the term Primary Health Care. Therefore, when addressing equity and implementation of options, considerations on PHC were included. Moreover, policy options have not the same effectiveness, so considerations on risks and benefits, as well as on implementation barriers and facilitators, were also included.

This evidence brief presented the following four options for reducing CS.

Figure 1. Information flow chart of the different steps of the systematic review



1) Expanding the offer of screening test for all pregnant women.

This option focus on the importance of providing dual rapid test for the screening of HIV and syphilis, using the same device, for all pregnant women and partners, at both the first antenatal visit and during the third trimester. This strategy aims to provide immediate treatment and prevention of complications resulting from vertical transmission of syphilis and HIV co-infection.

The key elements of this option are: On-site screening for syphilis during antenatal care, at first and third trimester visits;[13] dual rapid diagnostic tests for HIV/syphilis;[14,15] and reduction of foetal death and stillbirth, as well as vertical transmission of syphilis through screening and treatment in early pregnancy.[16,17]

Evaluation of the sensitivity and specificity of rapid tests has shown that, in resource limited settings, the benefit of screening using rapid tests far outweighs the risk of a small proportion of missed cases due to false negative results or over-treatment due to a past infection.[18]

Most of the studies included in the systematic reviews evaluating the antenatal screening to reduce the incidence of congenital syphilis were conducted in low income, lower-middle income, upper-middle income countries.[13-16,17,18,19]

2) Performing benzathine penicillin treatment for pregnant women with positive screening test for syphilis.

This option is based on the need to treat pregnant women diagnosed with syphilis with benzathine penicillin, if possible, in the first trimester of pregnancy. In addition, treatment of sexual partners may prevent both reinfection of pregnant women and congenital syphilis.

Penicillin is effective in the treatment of syphilis in pregnancy and the prevention of CS. Vertical transmission of syphilis has been prevented in 95% to 98% of cases with penicillin therapy.[2] Moreover, another systematic review provided evidence supporting that at least 2.4 MU benzathine penicillin given at least 28 days prior to delivery is effective in the treatment of syphilis in pregnant women to prevent CS, with greatest effect when given early in pregnancy (before 24-28 weeks).[20]

Untreated syphilis in pregnancy leads to the increase in stillbirths in various regions of the world. In some areas of sub-Saharan Africa, about 25% to 50% of all stillbirths has been associated with syphilis.[21] Another systematic review showed that stillbirth in the Americas region associated with untreated maternal syphilis has been a serious public health problem, as pregnant women with syphilis have had a 6.8 times higher odds of stillbirth compared to women that do not have syphilis.[22]

3) Developing strategies to notify STI and treat partners.

This option addresses strategies to notify partners of STI, including syphilis, in order to prevent reinfection, dissemination and possible complications of the disease, as it is the case of CS.

The key elements of this option include four main strategies for partner notification: a) Patient referral: patient tells sexual partners they need to be treated; b) Expedited partner therapy: the patient delivers medication or a prescription for medication to their partner(s) without the need for a medical examination of the partner; c) Provider referral: health service personnel notify the partners after patient's consent; d) Contract referral: patient is encouraged to notify partner, with the understanding that the partners will be contacted if they do not visit the health service by a certain date.[23]

Partner notification strategies require patients to inform their own partners about the STI diagnosis and encourage them to consider screening or treatment. This disclosure may lead to benefits such as successful partner treatment, emotional support and protection of the health of others. But it can also lead to stigma, rejection, physical abuse and discrimination.[23]

4) Promoting educational interventions through health campaigns with emphasis on congenital syphilis, and related to condom use, STI prevention and teenage pregnancy

This option addresses the use of media campaigns for health promotion, informing the population about prevention of vertical transmission of syphilis, with long term indicators; and the implementation of interventions for increasing condom use, as well as school-based interventions for preventing STIs and pregnancy in adolescents.

Benefits of this option are: a) changes in the health status of a population through syphilis prevention campaigns; (b) reduction of STIs worldwide by promoting condom use; c) the possibility of reducing adolescent pregnancy through incentive-based interventions that focus on keeping young people in secondary school.[24-26]

It is becoming increasingly clear that addressing structural determinants of health is a key component to improve sexual and reproductive health outcomes.[24]

Considerations on equity and implementation of the options

Equity is one of the doctrinal principles of the Brazilian Unified Health System (SUS) that guide health policies. It refers to the understanding that there are needs of specific groups, and that it is possible to develop practices that recognize the differences in living and health conditions and in the needs of different groups of people, considering the right to health.[27] In regard to the options for coping with CS, this means that decisions about their implementation should take into account how much they may or may not negatively affect inequalities between population groups.

The options presented do not have necessarily to be implemented jointly, as practical implementation should consider the local viability, being inserted in the governance of decision-making, regardless of the size of the health system (national, regional or local). It is necessary to consider that the lack of infrastructure, human resources, workflow definition in the follow-up of patients with reactive test results for syphilis, and acceptability of the test may be barriers to their implementation in PHC.[4] It is also important to consider PHC weaknesses and potentialities for the implementation of these options, especially those located in the field of culture and social representations of users and health workers.

Organization of antenatal care impacts on the effectiveness of syphilis treatment during pregnancy, therefore, it demands an urgent rethinking of practices related to the opportunities for syphilis treatment, increasing the responsibility of the different points of the care network, especially the PHC units.[28]

Conclusions

Four options were identified for coping with CS in PHC based on the twenty systematic reviews selected. But none of these systematic reviews made direct reference to the term Primary Health Care, which may indicate a lack of evidence addressing CS at the PHC level.

There are public policies for the eradication of CS in Brazil, which also addresses the options proposed in this study. Most health services, however, have presented weaknesses in the implementation of the recommended guidelines. Therefore, this synthesis of scientific evidence has noteworthy relevance in reaffirming the recommendations to face CS.

A limitation of this study was the lack of systematic reviews addressing social / family relations in the context of sexuality. There are several studies on clinical aspects of syphilis, focusing on diagnosis and therapy. However, it is also important to develop systematic reviews that address not only the biological aspects of STI, but issues related to the context of vulnerability of individuals with STI.

We expect that the scientific evidence presented in this study will support PHC managers and professionals in advancing the construction of health policies to confront CS, and that a deliberative dialogue will be developed to support the implementation of some of the options addressed in the synthesis.

Competing interests: The authors declare that they have no competing interests that might be perceived to influence the results and/or discussion reported in this article.

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References

1. Berman SM. Maternal syphilis: Pathophysiology and treatment. Bull World Health Organ. 2004; 82(6):433-38.
2. Walker GJA. Antibiotics for syphilis diagnosed during pregnancy. Cochrane Database of Systematic Reviews; 2001.
3. *Ministério da Saúde (BR). Portaria nº 3.242, de 30 de dezembro de 2011. Dispõe sobre o Fluxograma Laboratorial da Sífilis e a utilização de testes rápidos para triagem da sífilis*. Brasília; 2011.
4. Nascimento DSF, Silva RC, Tártari DO, Cardoso EK. *Relato da dificuldade na implementação de teste rápido para detecção de sífilis em gestantes na Atenção Básica do SUS em um município do Sul do Brasil. Rev Bras Med Fam Comunidade*. 2018;13(40):1-8.
5. *Ministério da Saúde (BR). Protocolo Clínico e Diretrizes Terapêuticas para Prevenção da Transmissão Vertical de HIV, Sífilis e Hepatites Virais*. Brasília; 2018.
6. Domingues RMSM, Saraceni V, Hartz ZMDA, Leal MDC. *Sífilis congênita: evento sentinela da qualidade da assistência pré-natal. Rev Nutr*. 2013; 47(1):147–57.
7. World Health Organization. The Global elimination of congenital syphilis: rationale and strategy for action; 2007.
8. Domingues RMSM, Leal MC. *Incidência de sífilis congênita e fatores associados à transmissão vertical da sífilis: dados do estudo Nascer no Brasil*; 2016.
9. Rocha AFB, Araújo MAL, Miranda AE, Leon RGP, Junior GBS, Vasconcelos LDPG. Management of sexual partners of pregnant women with syphilis in northeastern Brazil – a qualitative study. BMC Health Services Research; 2019.
10. Lavis JN, Oxman AD, Lewin S, Fretheim A. SUPPORT tooll for evidence-informed health policymaking (STP). Introduction. Health Research Policy and Systems; 2009.
11. Vettore M. *O abismo entre o saber e o fazer: a experiência do município de Piripiri com o Núcleo de Evidências em Saúde no planejamento de política em saúde*; 2011.
12. Shea BJ, Grimshaw JM, Wells GA, Boers M, Andersson N, Hamel C, et al. Development of AMSTAR: a measurement tool to assess the methodological quality of systematic reviews. BMC Medical Research Methodology. 2007; 7(10).
13. Shahrook S, Mori R, Ochirbat T, Gomi H. Strategies of testing for syphilis during pregnancy; 2014.
14. Harriet DG, Peeling RW, Kamb ML, Toskin I, Wi TE, Taylor MM. A systematic review and meta-analysis of studies evaluating the performance and operational characteristics of dual point-of-care tests for HIV and syphilis; 2016.
15. Swartzendruber A, Steiner RJ, Adler MR, Kamb ML, Newman LM. Introduction of rapid syphilis testing in antenatal care: A systematic review of the impact on HIV and syphilis testing uptake and coverage; 2015.
16. Di Mario S, Say L, Lincetto O. Risk factors for stillbirth in developing countries: a systematic review of the literature; 2007.
17. Hawkes S, Matin N, Broutet N, Low N. Effectiveness of interventions to improve screening for syphilis in pregnancy: a systematic review and meta-analysis; 2011.
18. Jafari Y, Peeling RW, Shivkumar S, Claessens C, Joseph L, Pai NP. Are Treponema pallidum Specific Rapid and Point-of-Care Tests for Syphilis Accurate Enough for Screening in Resource Limited Settings? Evidence from a Meta-Analysis; 2013.
19. Rogozinska E, Kara-Newton L, Zamora JR, Khan KS. On-site test to detect syphilis in pregnancy: a systematic review of test accuracy studies; 2017.
20. Blencowe H, Cousens S, Kamb M, Berman S, Lawn JE. Lives Saved Tool supplement detection and treatment of syphilis in pregnancy to reduce syphilis related stillbirths and neonatal mortality. BMC Public Health; 2011.
21. Bhutta et al. Effectiveness of interventions to screen and manage infections during pregnancy on reducing stillbirths: a review. BMC Public Health; 2011.
22. Lauren A, Serruya S, Duran P. Gestational syphilis and stillbirth in the Americas: a systematic review and meta-analysis; 2015.
23. Ferreira A, Young T, Mathews C, Zunza M, Low N. Strategies for partner notification for sexually transmitted infections, including HIV; 2013.
24. Mason-Jones AJ, Sinclair D, Mathews C, Kagee A, Hillman A, Lombard C. School-based interventions for preventing HIV, sexually transmitted infections, and pregnancy in adolescents; 2014.
25. Moreno R, Nababan HY, Ota E, Wariki WMV, Ezoe S, Gilmour S, Shibuya K. Structural and community-level interventions for increasing condom use to prevent the transmission of HIV and other sexually transmitted infections; 2014.
26. Saraceni V, Leal M do C, Hartz ZM de A. *Avaliação de campanhas de saúde com ênfase na sífilis congênita: uma revisão sistemática. Rev Bras Saúde Matern Infant*. 2005; 5(3):263–73.
27. Barros FPC, Sousa MF. *Equidade: seus conceitos, significações e implicações para o SUS São Paulo: Saúde Soc*. 2016; 25(1):9-18.
28. Sanine et al. *Sífilis Congênita: avaliação em serviços de Atenção Primária do estado de São Paulo, Brasil. Boletim do Instituto de Saúde*. 2016; 17(2):1809-7529.

Perceptions about the treatment of men diagnosed with syphilis: a rapid synthesis of qualitative evidence

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Abstract

Syphilis affects more than 12 million people worldwide, and men are an important source of transmission for their partnerships when they are inadequately diagnosed and treated. Individual factors related to men may be important as barriers to seeking and adhering to appropriate treatment. The objective of this study was to describe aspects related to the treatment of men with syphilis diagnosis, in their perception, of their partnerships, and health professionals. Was conducted a rapid synthesis of qualitative evidence based on the analysis of qualitative primary studies, and a subsequent description of the results. The search was done in MEDLINE (via PubMed), EMBASE (via Elsevier), LILACS (via VHL), PsycINFO, grey literature in Google Scholar, without language or time restriction. Two reviewers performed the selection and the discrepancies were resolved by consensus. The methodological quality was evaluated using the CASP instrument. From 2881 studies retrieved, five were included. The selected contents were categorized for a better presentation of the results, showing that the aspects that can influence the male population in the treatment of syphilis are associated to schooling, gender perspective, low understanding of the disease, and difficulties in accessing services, including those related to the organization of the work process.

Keywords: Syphilis, congenital syphilis, men, rapid qualitative synthesis.

Introduction

Syphilis is a sexually transmitted infection (STI), and can be transmitted in a variety of ways including via sexual encounters, parenteral and vertical transmission during pregnancy. It is characterized by active and latent periods, disseminated systemic involvement, and progression to acute complications in patients that remain untreated or have been inadequately treated.[1] Acquired syphilis has been most frequently diagnosed among younger segments of the population, especially men.[2]

When inadequately treated, men represent an important source of transmission to their partners: women, pregnant women and men who have sex

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with men (MSM). Understanding the factors that interfere with the treatment can contribute to advances in the (re)organization of work processes of health services, and formulation of public policies aimed at controlling this disease.

The objective of this study was to summarize the evidence related to the treatment of men with syphilis diagnosis, taking into consideration their perception, of their partners, and health professionals.

Ethical approval from a Human Research Ethics Committee was not sought as this was a systematic review of the published literature.

Method

A synthesis of evidence was conducted following the PRISMA[3] guidelines for reporting qualitative evidence syntheses (PRISMA stands for Preferred Reporting Items for Systematic Reviews and Meta-Analyses). The search strategy used in PubMed was: (((“Syphilis”[Mesh] or Great Pox or Pox, Great)) AND (“Syphilis, Congenital”[Mesh] OR congenital syphilis OR hutchinson’s teeth OR hutchinson teeth OR hutchinson s teeth OR teeth, hutchinson’s)) AND (“Men”[Mesh] or Boys or male). Additional search was carried out in MEDLINE (via PubMed), EMBASE (via Elsevier), LILACS (via VHL), PsycINFO, grey literature in Google Scholar with the same search terms used in PubMed , without language or time restriction, until March 2019. Studies addressing the perceptions of men, partners and health professionals about the treatment of men diagnosed with syphilis were considered eligible. Primary studies and experience reports with qualitative analyses were included. The exclusion criteria were epidemiological studies with an exclusively quantitative analysis approach.

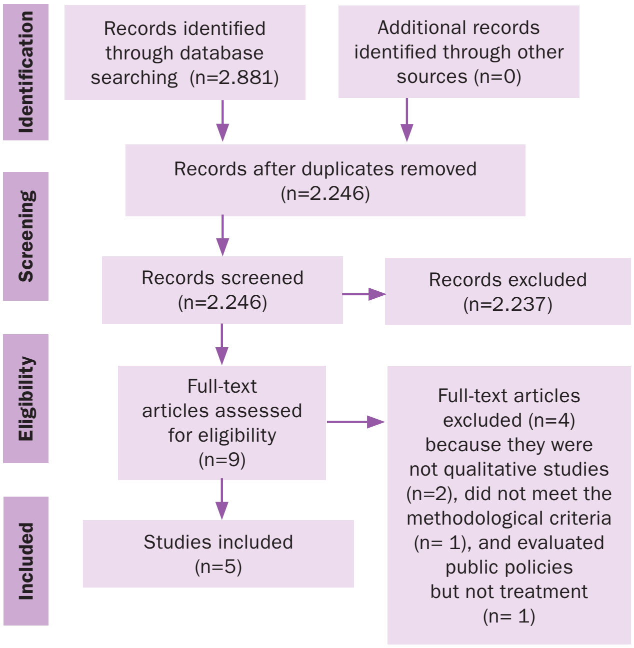
Two researchers (MLG; VCGG) performed independently the selection of studies and disagreements were resolved by a third researcher (DGMT). We used Rayyan software to perform title/abstract and full-text screening[4], with ‘Blind on’ to ensure the blinding in the study selection process. After full-text reading and data extraction, the results were submitted to individual and comparative analysis, based on grouping of categories of analysis, and interpretations were organised jointly by two researchers (MLG; VCGG) for the construction of a narrative synthesis.[5] The methodological quality of the studies was assessed using the Critical Appraisal Skills Programme (CASP) tool.[6]

Results

In total, 2,881 studies were identified, of which 635 duplicates were removed, and 2,237 were excluded after screening of titles and abstracts. Nine studies were selected for full-text reading, of which four were excluded because they were not qualitative studies, did not meet the methodological criteria, and evaluated only public policies, as Figure 1 shows.

The characteristics of the five included studies[7,8,9,10,11] are shown in the Table 1, including

Figure 1. Flow diagram of the article selection process



assessment score by CASP tool. Regarding geographic distribution, four studies were conducted in Brazil (three in the State of Ceará and one in Rio de Janeiro) and one in Lima, Peru. The articles were published from 2011 to 2019.

Four categories of analysis were defined for a better understanding of the aspects related to the treatment of syphilis: (a) social vulnerability, (b) organisation of service / work process, (c) feelings involved in treatment, (d) behaviours / attitudes of men, their partners and health professionals. The results are presented in Table 2. The findings, according to the categories of analysis, are described below.

Table 1. Characteristics of the analysed studies.

CHARACTERISTICS	STUDY S1	STUDY S2	STUDY S3	STUDY S4	STUDY S5
Author, year	Silva Júnior et al. (2017)[10]	Figueiredo et al. (2015)[11]	Williams et al. (2011)[7]	Rocha et al. (2019)[8]	Silva et al. (2017)[9]
Type of study	Descriptive, qualitative, with semi- structured interview	Qualitative, with semi-structured interview	Descriptive, quantitative and qualitative	Qualitative, with semi-structured interview	Exploratory qualitative
Participants	20 health professionals (7 physicians, 13 nurses and 6 health unit coordinators)	10 Nurses	58 professionals; 18 pregnant women	4 sexual partners, 21 professionals, 6 coordinators, 9 pregnant women diagnosed with syphilis during antenatal care	15 women diagnosed with syphilis during puerperium
Study location	City of Fortaleza, State of Ceará, Brazil	City of Crato, State of Ceará, Brazil	City of Lima, Peru	City of Fortaleza, State of Ceará, Brazil	City of Rio de Janeiro, State of Rio de Janeiro, Brazil
Period of data collection	2016	2010	Not reported	2014	2015
Aim	To investigate the perception of primary health care professionals about the treatment of sexual partners of pregnant women with syphilis.	To investigate the perception of nurses of the Family Health Strategy on the factors that influence adhesion to treatment of sexual partners of pregnant women with syphilis.	To identify factors that affect partner notification and treatment for maternal syphilis.	To evaluate the notification, testing, treatment and follow-up of sexual partners of pregnant women with syphilis in primary health care.	To understand the puerperal woman's knowledge about syphilis and treatment of gestational syphilis
Age range	Not reported	Not reported	Not reported	Professionals: 35-50 years old Women: 18-33 years old Partner: 30 years old	21-25 anos

S1 to S5: Studies included in the synthesis

Table 2. Aspects related to the treatment of men with syphilis diagnosis, in their perception, of their partners, and health professionals.

Categories	Social Vulnerability	Organisation of service/ work process	Sentimentos envolvidos no tratamento	Behaviours/attitudes of men, their partners and health professionals
STUDY S1	Use of illicit drugs	Sexual partners of pregnant women with syphilis are not from the healthcare unit area; health workers do not fell prepared to adequately manage these types of demands and require training.	Fear due to marital problems.	Chauvinism and gender issues of partners; lack of symptoms makes partners believe they have no infection; partners are drug users.
STUDY S2	Low socio-economic-cultural and educational level.	Treatment not carried out at the site of diagnosis, with referral to the hospital network. Bond can be lost; uncertainty of continuity of care; partners complaining about long waiting times in the hospital network. Lack of documentation of the treatment across the service network, depending solely on the information provided by the patient. Failure to provide guidance and health education.	Fear of anaphylactic reaction and pain. Partners' complaints of intramuscular route of administration.	Professionals do not properly communicate with patients; lack of bond between professionals and patients.
STUDY S3	Low education.	Lack of human resources; patient care only during regular clinic hours; lack of care coordination; rapid syphilis test unavailable at the time of care; partner notification by the woman was more difficult when a health professional was not present. Professionals do not feel prepared for counselling and request additional training from technical team.	Fear of blame, violence, and abandonment as women's reasons to not notify their partners. Distrust in test results.	*Machismo* of the partners. Difficulty in notifying partners due to women's lack of knowledge about the disease.
STUDY S4	Low education; unemployment; history of illicit drug abuse; prison history.	Protocols and manuals for supporting professionals not available. No clarification of doubts of professionals by the central level. No educational materials for supporting patients (restricted to the central level); training does not approach partner notification or dealing with situations resulting from revealing the diagnosis of syphilis; professional conduct varies among services: Some provide treatment prescriptions (physicians) for partners, while others request appointment for partners at the service.	Professionals' lack of confidence to deal with partners of pregnant women. Treatment not delivered in the basic health unit due to professionals' fear of anaphylactic reaction.	Professionals not able to take any actions other than continuing the pregnant woman's care, not interesting in changing these practices.

Categories	Social Vulnerability	Organisation of service/ work process	Sentimentos envolvidos no tratamento	Behaviours/attitudes of men, their partners and health professionals
STUDY S4	Low education; unemployment; history of illicit drug abuse; prison history.	Difficulties in accessing the test and delay in the delivery of results. Blood collection is not done on a daily basis, making more difficult the access to diagnosis and forcing people to return to the services several times. Incomplete health teams. Areas uncovered. Difficulty in tracking partners when patients are not from the coverage area. Patients referred for treatment in other units: Absence of financial resources; long distances to hospital services; professionals' complaints of difficulty in controlling the doses administered (lack of treatment record). Services overcrowded; validity of prescription given by primary care provider not accepted at other levels of care. Hospitals send patients back to the primary care for treatment. Nurses do not carry out rapid syphilis testing on a daily basis because it increases overload and time of consultation. Suggestion for programmatic agendas (appointments once a week). Professionals' difficulty in implementing the rapid test during antenatal care, and unawareness of the need for monthly follow-up.	Patients and partners' discomfort and embarrassment due to referrals to the hospital network (repetition of diagnosis to professionals with whom they have no bonds, and who do not recognize them or know their life histories). Partners not confident in receiving treatment, due to not feeling well informed by their women. Professionals' concern about ethical issues and the possibility of breaching the confidentiality. Professionals' fear of risk of adverse events in treatment delivered in the basic health unit. Women's discomfort to notify the partner, considering professional presence important.	Professionals not able to take any actions other than continuing the pregnant woman's care, not interesting in changing these practices.
STUDY S5	Low education	Delay in diagnosis, related to delivery of results not in a timely manner. Failure / lack of guidance on diagnosis and treatment of syphilis during antenatal care. Few patients diagnosed during antenatal care, less than a half diagnosed during puerperium.	Embarrassment in telling partner due to the possibility of judgement related to a past as a sex worker.	Lack of understanding of the disease, influencing patients' ability to notify their partners.

Social Vulnerability

The social aspects described most often were low education, unemployment, drug abuse and prison. Low education was presented in studies S3, S4 and S5 as a barrier to the treatment of partners, including lack of understanding of the disease.[7,8,9]

Abuse of licit and illicit drugs was presented in studies S1 and S4 as a vulnerability factor, as the use of alcohol, tobacco, marijuana, and other drugs may affect the individual physically and psychologically, facilitating risky behaviour such as not using condom.[10,8]

Organisation of service / work process

Institutional weaknesses that may interfere in the treatment of syphilis identified in studies S2, S3 and S4 are lack of human resources and test equipment and supplies.[11,7,8] Treatment not carried out at the site of diagnosis was reported in studies S2 and S4 as an important factor for failure to follow-up.[11,8]

Incipient counselling and pregnant women attending antenatal care without their partners were identified as weaknesses by study S5, directly related to the lack of guidelines to ensure best practices for diagnosis and treatment.[9]

Study S2 highlighted that counselling, guidance and health education are insufficient in strengthening the bond and trust between professionals and patients, and between men and women (couples)[11].

Feelings involved in treatment

Studies S3, S4 and S5 revealed, based on reports of pregnant women with syphilis, the existence of fear in communicating their partners about the infection because they do not have a correct understanding of their disease. This fear is also related to the possibility of suspicious of infidelity on the part of their partners, as well as suffering violence, and breaking off the relationship.[7,8,9]

Behaviours / attitudes of men, their partners and health professionals

Studies S1 and S2 indicated that lack of both understanding and symptoms, and also fear of medication make men less likely to seek or adhere to treatment.[10,11] In addition, other factors are related to men's personal perspective on gender, the role of men and women in society, blaming their partner for the infection and unfaithfulness In other cases, relationship is not always well established, and this may lead to distrust and difficulty in communicating the diagnosis and adhering to treatment.[7,8]

Discussion

The difficulty of treating men as sexual partners of patients with STI might be related to the historical development of health policies, which have always been exclusionary, hindering the search for care.[3]

Therefore, to address aspects of men's access to STI diagnosis and treatment, it is necessary not only to consider provision, organization, and quality of services, but also personal, social, historical, and cultural contexts. The culture of *machismo* (masculinity), typical in the Brazilian society, should also be considered by health teams in approaching patients and their partners.

As shown by this study, one of the main difficulties reported by both women and health professionals is the communication of diagnosis and the need for treatment. In this sense, health education is important, with an approach that uses appropriate language and goes beyond the signs and symptoms of the disease, ensuring understanding as well as an enjoyable learning experience.

Regarding the organization of service / work process, we found that there are a number of issues that may compromise the efficacy of beginning and continuity of the treatment of syphilis in the male population. Treatment not carried out at the site of diagnosis seems to be the most important factor. The need to go to various health services also makes the treatment more difficult because information on treatment, such as prescriptions and test results, may be lost.

According to Palácio[12], there is a great challenge in the dichotomy that comes with autonomy of the individuals to make their own choices in health care. The continuum of care for these populations should be structured to assure continuity of care. It should be taken into account that moving from the basic health unit to referral services and hospitals involves economic and social difficulties for patients.

Moreover, in MSM population, fear of disapproval and discrimination from health professionals may deter this population from seeking health services. In the study of Brignol[13], more than 60% of respondents had already suffered discrimination based on race, colour, sexual orientation, representing a real barrier to access to services.

It is important to establish continuum of care for syphilis treatment that ensures comprehensiveness, along with permanent health education of professionals of the service network, with the purpose of providing knowledge about syphilis and its treatment, developing soft technologies to promote humanised care, and, then, contributing to the (re)organization of work processes with a focus on an appropriate approach and bonding.

It also should be pointed out that professionals reported they lack confidence in administering penicillin, although the use of lidocaine hydrochloride as a diluent for benzathine penicillin G does not change the penicillin concentration in body fluids and significantly reduces the pain of injection.[14]

Finally, behaviours / attitudes of those involved in the treatment of syphilis can certainly be barriers to its beginning or continuity. Stigma, lack of knowledge about the disease, time, costs involved in treatment, and family conflicts arising from the diagnosis should always be considered in the planning of management and health teams regarding the care of people with syphilis.

As a limitation of this study, only a small number of qualitative studies have explored the motivations and perceptions of men about their treatment, as it is more common to seek the opinion of female partners, mostly pregnant women, and health professionals, thus obtaining information indirectly. Another limitation may be the fact that in the search strategy terms such as barriers or difficulties in treatment were not used. Furthermore, an approach to assess the confidence in the findings was not used, and there was limited time to conduct this study.[15]

Conclusions

Perceptions of social vulnerability, work process, feelings, and behaviours / attitudes have been revealed based on the statements from men, their partners and health professionals, which can influence the treatment of syphilis.

This study may contribute to directing and promoting future research on the motivations, perceptions and vulnerabilities of men related to their adherence to syphilis treatment, considering that there are research gaps in this area.

It is noteworthy to mention that syphilis is a complex issue, imbued with diverse personal values and cultural interpretations. Therefore, indirect information on behaviour, such as opinion of the partners, may help address, but not determine the perceptions involved in treatment.

References

1 - Avelleira JCR, Bottino G. *Sífilis: diagnóstico, tratamento e controle. An Bras Dermatol* [Internet]. *março de 2006* [accessed 26 September 2018];81(2):111–26. Available at:

http://www.scielo.br/scielo.php?script=sci_arttext&pid=S0365-05962006000200002&lng=pt&tlng=pt.

2 - *OPAS/OMS Brasil - Início* [Internet]. 2018 [accessed 24 October 2018]. Available at: https://www.paho.org/bra/.

3- Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097.

4- Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan—a web and mobile app for systematic reviews. Systematic Reviews. 2016;5(1):210.

5- Snilstveit B, Oliver S, Vojtkova M. Narrative approaches to systematic review and synthesis of evidence for international development policy and practice. Journal of Development Effectiveness. 2016;4(3):409-429.

6 - Critical Appraisal Skills Programme (2018). CASP (Qualitative) Checklist. [Internet] [accessed 30 March 2019]. Available at: https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf.

7 - Williams B, Garcia P, Carcamo C, Chiape Guiterrez M, De la Rosa Roca S, Calderon MV, Peeling R. Partner notification and treatment for maternal syphilis in Lima, Peru: knowledge, attitudes, and practices of health providers and patients. Sex Transm Infect. July 2011, Vol 87, Supl 1.

8- Rocha AFB, Araújo MAL, Miranda AE, Ponce de Leon RG, Silva Junior GB, Vasconcelos LDPG. Management of sexual partners of pregnant women with syphilis in northeastern Brazil – a qualitative study. BMC Health Services Research (2019) 19:65.

9- Silva MRB, Silva AP, Messias CM, Silva HCDA, Silva LA, Rizzo ER. *Conhecimento das puérperas sobre a sífilis: Transmissão e tratamento*. *Revista* Nursing, 20 (224):1556-1560 (2017).

10- Silva Junior GB, Rocha AFB, Romualdo KS, Xavier PS, Araújo MAL. Primary healthcare workers' perceptions regarding calling sexual partners of pregnant women with syphilis for treatment. Sex Transm Infect 2017;93(Suppl 2):A1–A272.

11- Figueiredo MSN, Cavalcante EGR, Oliveira CJ, Monteiro MFV, Quirino GS, Oliveira DR. *Percepção de enfermeiros sobre a adesão ao tratamento dos parceiros de gestantes com sífilis*. *Rev Rene*. 2015 maio-jun; 16(3):345-54.

12- Palacio MB, Figueiredo MAC, Souza LB. *O cuidado em HIV/AIDS e a atenção primária em saúde. Psico* v. 43, n. 3, pp. 360-367, jul./set. 2012.

13- Brignol S, Dourado I, Amorim LD, Kerr LRSF. *Vulnerabilidade no contexto da infecção por HIV e sífilis numa população de homens que fazem sexo com homens (HSH) no Município de Salvador, Bahia, Brasil. Cad. Saúde Pública*, Rio de Janeiro, 31(5):1-14, mai, 2015.

14- Amir J, Ginat S, Cohen YH, Marcus TE, Keller N, Varsano I. Lidocaine as a diluent for administration of benzathine penicillin G. Pediatr Infect Dis J. outubro de 1998;17(10):890–3.

15- Taylor B, Henshall C, Kenyon S, Litchfield I, Greenfield S. Can rapid approaches to qualitative analysis deliver timely, valid findings to clinical leaders? A mixed methods study comparing rapid and thematic analysis. BMJ open. 2018, 8(10).

Motivations, thoughts and feelings associated with suicidal ideation for Brazilian adolescents: a rapid qualitative evidence synthesis

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Abstract

The objective of this study was to synthesize evidence related to the motivations, thoughts, and suicidal ideation experiences among Brazilian adolescents. The search was performed in MEDLINE, EMBASE, LILACS, PsycINFO, Google Scholar and Google. The articles selection, data extraction and methodological quality evaluation were performed by two independent researchers. Of 3,496 identified articles, four studies were included. The main motivations for suicidal ideation were fragile bonds, depression, and lack of affection. The predominant feelings were loneliness, sadness, helplessness, and hopelessness. Thoughts related to the belief that one is alone in the world and to not seeing a solution to one's sufferings and conflicts have been reported as related. Suicidal ideation may be an important phenomenon among Brazilian adolescents, requiring society's attention, especially by academics and the government, to expand the base of qualitative evidence and discussion of preventative public policy for suicide.

Keywords: adolescents, suicidal ideation, qualitative evidence synthesis.

Introduction

In Brazil, about eleven thousand people took their own lives in 2017, representing the fourth leading cause of death in young people aged 15-29. Between 2011 and 2016, 176,226 cases of self-harm were reported, of which 27.4 % were suicide attempts.[1,2,3]

One of the main triggers for suicidal behaviour in adolescents, between 10 and 19 years old, is suicidal ideation. This term refers to thoughts of serving as the agent of one's own death. Suicidal ideation varies in severity and specificity, but its presence increases the risk of suicide, especially in adolescence, when conflicts, changes and challenges are characteristic of this phase of life.[4,5,6,7]

Suicide is a complex and multifactorial phenomenon, including biological, socio-cultural

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and economic factors, aspects of personal and family history, as well as traumatic events. In adolescence, all these factors are even more stressful, due to vulnerability caused by changes in physical, sexual, cognitive and emotional aspects, as a process of emotional reorganization.[8]

Considering the social relevance, cultural aspects of the Brazilian adolescents, and the importance of strengthening evidence-based public policy in Brazil, the aim of this study was to summarize qualitative evidence that addressed motivations, thoughts and feelings related to the ideation suicidal.

Method

This study is a rapid qualitative evidence synthesis.[9] PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses)[10] was used to support the development of a transparent reporting, and the question regarding the phenomenon of interest was formulated using the acronym SPIDER.[11] The searches were carried out in February 2019, in MEDLINE (via PubMed), EMBASE (via Elsevier), LILACS (via VHL), PsycINFO, Google Scholar and Google Search (five first pages), without language or time restriction. The search strategy used was ((suicidal ideation or suicide idea or motivation or expectation) and (adolescent or youth* or child*) and (Brazil or Brazilians or Latin*)), for all the databases, except EMBASE, whose strategy was ((‘suicidal ideation’/exp OR ‘motivation*’ OR ‘expectation*’) AND (‘adolescent*’ OR ‘juvenile*’ OR ‘child*’) AND (‘Brazil’/exp OR ‘Brazilian*’ OR ‘Hispanic’/exp)). To search for additional grey literature, we carried out a new search in Google Scholar with the strategy ‘case study’ ‘suicidal ideation’ ‘adolescent’.

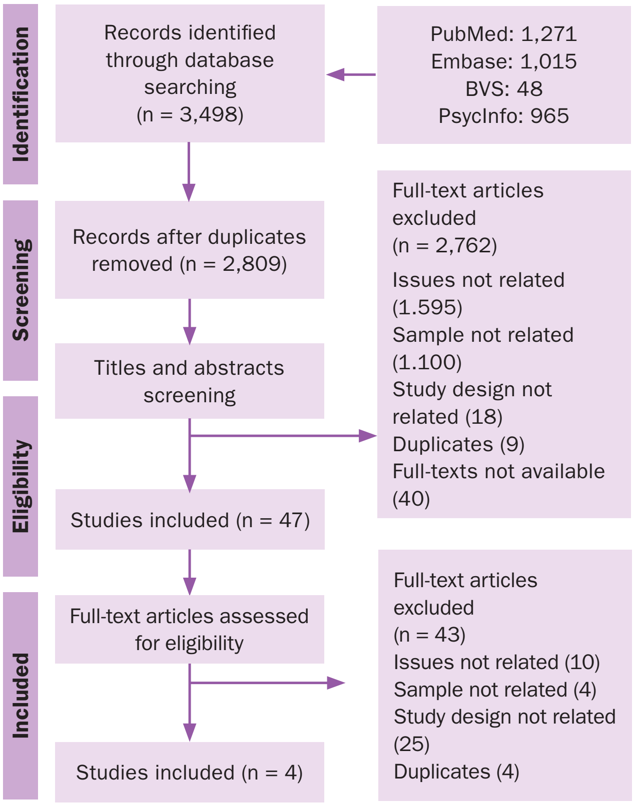
The title/abstract and full-text screening was conducted by two independent researchers (ASB and SPTO). Discrepancies were resolved by consensus. The reference management system Rayyan[12] for the selection of studies was used. Studies on adolescents living in Brazil, aged 10-19 years, who presented motivation and thoughts related to suicidal ideation were selected. Given the subjective nature of the topic and the need to understand the phenomenon in its entirety, we focused on qualitative studies.

The following information was extracted from the selected studies: Objective, study type and location, population, methodological approach, and socio-demographic characteristics of the participants (age, gender, ethnicity, socio-economic profile), findings on motivations, feelings and thoughts associated with suicidal ideation. For methodological quality assessment, the Critical Appraisal Skills Programme (CASP) tool was used.[13]

Qualitative meta-synthesis was conducted based on interpretive synthesis[14] of data whose thematic core issues were: motivations, thoughts and feelings. Ethical approval from a Human Research Ethics Committee was not sought as this was a systematic review of the published literature.

Results and discussion

Four studies were selected for analysis,[15-18] as shown in Figure 1.



In the methodological quality evaluation, S1[15] and S4[17] studies were rated as high quality, S2[18] as moderate quality, and S3[16] as low quality.

Limitations of this evidence synthesis are related to the small number of included studies, mostly case studies, indicating that the findings are insufficient to answer the research question conclusively. In addition, rapid reviews have their own methodological limitations.[9]

For a better understanding of the studies' findings, they were organized in thematic core issues that addressed motivations, thoughts and feelings of the phenomenon of interest related to suicidal ideation among adolescents (Tables 1 e 2).

Motivations

The main motivations related to suicidal ideation in S1[15] and S3[16] studies are: 'broken heart' and 'weak family ties'. Other motivations were related to family conflicts such as 'little affection', 'lack of respect among family members', 'family disagreements', and 'lack of appreciation of adolescents' physical appearance'.

Although suffering from a broken heart is not the only weaknesses that come from relationships, the love relationship takes on great importance in the adolescent's imaginary, in the expectation of filling the void of feeling helplessness, due to family conflicts.[15] Dissolution of bonds, weak family ties, along with financial difficulty are identified as triggers.[16-18]

S2[18] and S3[16] studies describe that a conflicted family life with disagreements, associated with a depressive state with symptoms such as low self-esteem, intense sadness, feelings of abandonment, hopelessness, loneliness, insomnia and worthlessness, predispose adolescents to think on suicide.

S4[17]study presents as motivations of females the absence of love, state of psychic suffering and hopelessness of the 'being suicidal'. For males, the motivations are represented as a desperate attitude, an act of weakness when facing a problem, and they are described ambiguously as 'something bad', 'something that brings relief'.

According to Borges and colleagues,[20] as well as Botega,[21] mild, moderate and severe depression is commonly associated with the presence of suicidal ideation. Adolescents with depressive disorders have an odds ratio twelve times greater of developing suicidal ideation compared to young people without depression.

Thoughts

S1[15] study describes the perception of a female adolescent, reporting that it is not enough to have economic support, because lack of emotional support and feeling of loneliness and rejection are related to suicidal ideation.

According to Minayo and colleagues[22], instead of looking at suicide as 'something isolated', we should see it as a social fact, considering the social ties extremely important, as well as the relationship conflicts and breakups.

In adolescence, suicide attempts is often triggered by difficulties in expressing one's own feelings, reported by the adolescents in S2[18] study. Many adolescents seek refuge in their inner world, but those more sensitive and emotional may develop depressive disorders with notable symptoms of discontent, confusion, loneliness and sadness.

There are numerous thoughts and circumstances that lead the individual to attempt suicide and suicidal ideation, but the studies pointed out difficulties in dealing with suffering. Silva and colleagues[23] reported that 60% of the individuals who attempted suicide had had suicidal ideation.

In S4[17]study, adolescents reported that suicidal ideation represent an attitude of despair, an act of weakness when facing a problem.

Feelings

The included studies reported feeling of loneliness,[15-18] sadness,[15,16,18] hopeless,[16-18] helplessness,[15,18] despair,[15,18] failure,[15,18] and other feelings such as anguish, anger, irritation, frustration, abandonment and suffering.[15,16,18] These are useful indicators for parents, educators, health professionals and managers in the development of policies and interventions on this issue.[24]

Loneliness[15-18] was strongly associated with social isolation and the weak emotional ties, especially family ones. Although 'feeling alone even while surrounded by people' or 'experiencing a relationship breakup' were commonly reported, breakdown of intra-family relationships has been recurrent, and most likely the main cause, as it is, in one way or another, part of all relationships. In S2[18], the female adolescent manifested feeling sadness without reason, and not knowing how to deal with it, feeling a paralysing anguish.

Sadness[16,18] was related to loss of relationships, helplessness, as well as depression.[15,16,18] Adolescence is a time for learning how to deal with personal life events, when social demands, and value for independence and autonomy contribute to increase depression.[15,16,18]

Loneliness, sadness, hopelessness and helplessness are predominant, especially in relation to relationship breakups, without the support of family ties. Poor support networks, especially at the family level, has proved to be an important factor, directly related to suicide ideation and attempt.[15,16,18]

Suicidal ideation as a response to psychological pain was reported in all the studies analysed. Despair or hopelessness regarding the cessation of suffering was presented as a motivation for suicide ideation and attempt. Impulsiveness to bring relief was indicated as the mechanism of suicide, and it is a habitual behaviour of adolescents, who then find death a quicker and easier solution to an apparently insoluble problem.[15-18]

Conclusions

The studies showed the importance of understanding suicidal behaviour not only as an individual problem, but as a phenomenon that involves social aspects, including socio-economic factors, weak family ties that can lead to psychic suffering, and depression evidenced by feelings of loneliness, sadness, helplessness and hopelessness about the future, which are key elements of suicidal ideation.

In regard to the thoughts reported by adolescents, the belief that they are alone and that there are no solution to their sufferings, associated with disturbing thoughts such as that life is not worth living, lead them to think about taking their own life.

This review has showed that there is a need for further qualitative primary studies related to motivations, thoughts and feelings associated with suicidal ideation among adolescents, in order to support mainly education and health workers to adequately manage cases of suicidal ideation at the individual, family and community level, and also to support evidence-informed decision-making.

Based on this study data, situations of weak ties or relationship breakup, and feelings of loneliness, sadness, hopelessness, helplessness, despair and failure, should be seen as signs of suicidal ideation in adolescents, therefore, should be investigated for professional guidance and intervention in a timely manner to prevent suicides.

Table 1. Description of the included studies according to author and year, research question, method and methodological quality assessment.

Study	Author, year	Method, collection	Participants, setting and location	Research Question	CASP	
					% of total score	Quality
S1[15]	Vieira; Freitas; Pordeus; Lira; Silva, 2009	Anthropological approach, documentary research, participant observation and semi-structured interview	12 adolescents, 10-19 years old, female (83%) Diagnosis: Suicide attempt. Weak family ties with both parents Location: Emergency hospital, city of Fortaleza, Ceará state, Brazil	To identify the reasons for suicide attempts in adolescents, analysing their repercussions on the family and social context	70%	High
S2[18]	Amaral; Pereira; Silva; Kuhn, 2017	Case study based on consultations	1 adolescent, 16 years old, female Diagnosis: Crying crisis, deep sadness and heart palpitation. Strong family ties with the mother and weak with the father. Location: Psychosocial Care Centre for Children and Adolescents, city of Porto Alegre, Rio Grande do Sul state, Brazil	To identify opportunities for intervention with a focus on treatment adherence, based on the investigation of consultation records of adolescents with suicidal ideation	50%	Moderate
S3[16]	Hildebrandt; Zart; Leite, 2011	Descriptive research, collection based on a semi-structured interview and thematic analysis	3 adolescents, 13-18 years old, female (67%)	To know and analyse the reasons why adolescents attempt suicide and the methods used by them	10%	Low
S4[17]	Araújo; Vieira; Coutinho, 2010	Field research, qualitative and quantitative multimethod approach. Free-Word Association Test and Bio-socio-demographic questionnaire	1 adolescent, 16 years old, female	To identify the social representations of suicidal ideation among high school adolescents	100%	High

Table 2. Description of findings on motivations, thoughts and feelings related to suicidal ideation in the included studies.

Studies	Motivations	Thoughts	Feelings
S1[15]	'Broken heart', weak family ties, little affection, lack of respect among family members, lack of appreciation of adolescents' physical appearance.	'My aunt gives me all I need, financially, for example, school; but what I need the most is affection.' 'I thought nobody wanted to see me.'	Feelings of loneliness, rejection, failure, despair in suffering (the will to end the suffering greater than the will to die), helplessness and frustration.
S2[18]	Depression.	'I watched the entire television series 13 Reasons Why, and I have identified myself with the character (...). When she is surrounded by friends, but no one seems to notice her (...). Everybody says that I am young and pretty, that I have a future ahead of me (...). Do I want a future?' 'Sometimes I walk alone down the street, when I can think of what I really want to do (...), seeking for courage.' 'I cannot express my feelings, I am not a caring person, I do not show affection (...) I am more rational, I avoid contact with people (...)' (Amaral; Pereira; Silva; Kuhn, 2017, p.198) 'I am very sad, (...) I think about killing myself, I already know how (...) I would use a knife and stab my leg (...), but I think about my mother a lot (...). (Amaral; Pereira; Silva; Kuhn, 2017, p. 199).	Feelings of sadness, hopelessness, loneliness, anguish, failure and despair.
S3[16]	Conflicted family life, family disagreements, weak family ties, becoming sick, depressive state.	'I thought I had no reason to go on. But I wanted to end that suffering. And I did not want to suffer. I felt hated by everyone, thinking no one liked me. I used to believe that everything I did was not good enough. I felt useless.' 'But sometimes I come to the conclusion that I did not want to be in this world. What I wanted was to disappear, to evaporate.'	Feeling of sadness, anger, insecurity, loneliness, helplessness, hopelessness about the future, depressed, irritable mood, abandonment and worthlessness.
S4[17]	Lack of love, lack of affection, seeking relief, an 'end' to the suffering.	Suicidal ideation represented as an attitude of 'despair', an act of 'weakness' when facing a 'problem'.	Absence of love, state of psychic suffering, hopelessness, loneliness.

Competing interests:

The authors declare that they have no competing interests.

References

1. *Organização Pan-americana da Saúde (BR). Folha informativa – Suicídio* [Internet]. Brasília: OPAS/OMS Brasil. 2018 [accessed 9 August 2019]. Available at: https://www.paho.org/bra/index.php?option=com_content&view=article&id=5671:folha-informativa-suicidio&Itemid=839.
2. *Boletim Epidemiológico (BR) - Suicídio: saber, agir e prevenir. Brasília: Ministério da Saúde* [Internet]. 2017 [accessed 11 April 2019]; 48(30):15. Available at: http://portalarquivos2.saude.gov.br/images/pdf/2017/setembro/21/2017-025-Perfil-epidemiologico-das-tentativas-e-obitos-por-suicidio-no-Brasil-e-a-rede-de-atencao-a-saude.pdf.
3. *Ministério da Saúde (BR). Setembro Amarelo - Agenda Estratégica de Prevenção do Suicídio. Brasília: Ministério da Saúde* [Internet]. 2017 [accessed 11 April 2019]. Available at: http://portalarquivos.saude.gov.br/images/pdf/2017/setembro/21/Coletiva-suicidio-21-09.pdf.
4. Abdala-Filho E, Chalub M, Telles LEB. *Psiquiatria Forense de Taborda*. 3. Porto Alegre: Artmed; 2016.
5. Carlson GA, Cantwell DP. Suicidal Behavior and Depression in Children and Adolescents. J Am Acad Child Psychiatry [Internet]. 1982 [accessed 11 April 2019]; 21(4):361–8. Available at: https://www.sciencedirect.com/science/article/pii/S0002713809609390.
6. Ruiloba JV. *Introducción a la Psicopatologia y la psiquiatria.* 7. Barcelona: Elsevier; 2011.
7. Barrios LC, Everett SA, Simon TR, Brener ND. Suicide ideation among US college students associations with other injury risk behaviors. J Am Coll Health Assoc [Internet]. 2000 [accessed 11 April 2019]; 48(5):229–33. Available at: http://www.ncbi.nlm.nih.gov/pubmed/10778023.
8. Assis SG de, Gomes R, Pires T de O. Adolescence, sexual behavior and risk factors to health. Rev Saude Publica. 2014; 48(1):43–51.
9. Taylor B, Henshall C, Kenyon S, Litchfield I, Greenfield S. Can rapid approaches to qualitative analysis deliver timely, valid findings to clinical leaders? A mixed methods study comparing rapid and thematic analysis. BMJ Open [Internet]. 2018 [accessed 9 August 2019]; 8(10):1-13. Available at: https://bmjopen.bmj.com/content/8/10/e019993#article-bottom.
10. Moher D, Liberati A, Tetzlaff J, Altman DG, Altman D, Antes G, et al. Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. PLoS Medicine. 2009; 6(7):1-10.
11. Cooke A, Smith D, Booth A. Beyond PICO: the SPIDER tool for qualitative evidence synthesis. Qual Health Res [Internet]. 2012 [accessed 11 April 2019]; 22(10):1435–43. Available at: http://journals.sagepub.com/doi/10.1177/1049732312452938.
12. Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan-a web and mobile app for systematic reviews. Syst Rev [Internet]. 2016 [accessed 9 August 2019]; 5:210. Available at: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5139140/.
13. Critical Appraisal Skills Programme. CASP Checklist: 10 questions to help you meke sense of a Qualitative research [Oxford]: CASP [Internet]. 2018 [accessed 9 August 2019]. Available at: http://www.casp-uk.net/casp-tools-checklists.
14. Zimmer L. Qualitative meta-syntesis: a question of dialoguing with texts. 2006; 52(5):546-53.
15. Vieira LJE de S, Freitas MLV, Pordeus AMJ, Lira SVG, E Silva JG. *“Amor não correspondido”: discursos de adolescentes que tentaram suicídio. Cien Saude Colet* [Internet]. 2009 [accessed 8 August 2019]; 14(5):1825–34. Available at: http://www.scielo.br/scielo.php?script=sci_arttext&pid=S1413-81232009000500024&lng=pt&tlng=pt.
16. Hildebrandt LM, Zart F, Leite MT. *A tentativa de suicídio na percepção de adolescentes: um estudo descritivo. Rev Eletr. Enf* [Internet]. 2011 [accessed 8 August 2019]; 13(2):219–26. Available at: http://dx.doi.org/10.5216/ree.v13i2.8951.
17. Araújo LC, Vieira KFL, Coutinho MPL. *Ideação suicida na adolescência : um enfoque psicossociológico no contexto do ensino médio. Psico-USF* [Internet]. São Paulo: *Universidade São Francisco*. 2010 [accessed 9 August 2019]; 15(1):47–57. Available at: https://www.redalyc.org/articulo.oa?id=401036078006.
18. Amaral C, Pereira GB, Silva KL, Kuhn SM. *Depressão na adolescência com ideação suicida: um estudo de caso. In: Anais da XI Mostra Científica do Cesuca* [Internet]. 2017 [accessed 9 August 2019]; (11):199-214. Available at: http://ojs.cesuca.edu.br/index.php/mostrac/article/view/1342.

19. Dalgalarrondo P. *Psicopatologia e semiologia dos transtornos mentais.* 3. Porto Alegre: Artmed; 2018.
20. Borges VR, Werlang BSG, Copatti M. *Ideação suicida em adolescentes de 13 a 17 anos. Barbarói* [Internet]. 2008 [accessed 9 August 2019]; 28(1):109–23. Available at: https://online.unisc.br/seer/index.php/barbaroi/article/view/19219.
21. Botega NJ. *Crise suicida: avaliação e manejo.* Porto Alegre: Artmed; 2015.
22. Minayo MC de S, de Assis SG, de Souza ER, Correia BSC, Pacheco ML, Delgado, PGG. *Suicídios no Brasil: Mortalidade, Tentativas, Ideação, Comportamento Autopunitivo e Prevenção* [Internet]. 2010 [accessed 9 August 2019]. Available at: https://docplayer.com.br/62783655-Suicidios-no-brasil-mortalidade-tentativas-ideacao-comportamento-autopunitivo-e-prevencao.html.
23. Silva VF da, Oliveira HB de, Botega NJ, Marín-León L, Barros MBA, Dalgalarrondo P. *Fatores associados à ideação suicida na comunidade: um estudo de caso-controle. Cad Saude Publica* [Internet]. 2006 [accessed 12 April 2019]; 22(9):1835–43. Available at: http://www.scielo.br/scielo.php?script=sci_arttext&pid=S0102-311X2006000900014&lng=pt&tlng=pt.
24. Abed, A. *O desenvolvimento das habilidades socioemocionais como caminho para aprendizagem e o sucesso escolar de alunos da educação básica*. São Paulo: UNESCO/MEC; 2014.

Rapid review to inform the National Policy for Biological Medicines in SUS

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Abstract

Background: High costs with biological medicines threaten the sustainability of healthcare services. The objective of the present rapid review was to inform the discussion of the National Policy for Biological Medicines within the Brazilian Unified Health System (SUS), through the identification of barriers to the uptake of these medicines. **Methodology**: Rapid review of the data sources: Medline via PubMed, EMBASE, Cochrane Library and Centre for Reviews and Dissemination. **Results**: Nine cross-sectional studies were included. For the users, the barriers were lack of knowledge about the treatment, the distance between residence and healthcare services, the long waiting periods for care and the passivity about the treatment decisions. For the health professionals, the barriers were acceptability of treatment, interchangeability and substitution and the perception of lack on clinical data of efficacy and safety. For the payers, the barriers were high cost of medicines, reimbursement and assistance problems. No barriers were found for healthcare system in the included studies, only regarding the private sector, which was the patent protection. **Conclusion**: It is essential that decision makers use scientific evidence in healthcare policies. There is a pressing need to identify solutions to the barriers to access biologic medicines identified in this rapid review.

Keywords: Rapid review; Biological Products; Evidence-Informed Policy.

Background

Expenses with medicines pressure healthcare systems, as resources are finite and the burden of chronic conditions is growing due to the epidemiological transition.[1] Several countries face this problem, as medicines are increasingly specialized and expensive.

The Brazilian Federal Government spends approximately 15 billion Brazilian reais per year for ambulatory pharmaceutical care programmes, with an emphasis on specialized medicines, which make up about half of the budget.[2] In Brazil, public health is a constitutional right, thus, the Ministry of Health is responsible for 60% of all purchases of biological products, which raises the interest of pharmaceutical industries in having it as a buyer. Although they represent only about 2% of all medicines purchased, biological medicines constitute 40% of the pharmaceutical care budget of the Ministry of Health.[3]

Manufacturers of reference biological products have patent protection. Once the patent expires, other industries can produce similars, which are more affordable due to lower investment in research

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and development, in addition to the possible impact of market competition. Biosimilars are expected to be instrumental in reducing the cost of treatment with biologics, ensuring continuity of care, and increasing user access to treatment.[2,4,5]

The Brazilian law no. 8,666/1993 states the legal framework for public procurement: the selection of the most advantageous proposal to the administration and the promotion of sustainable national development govern the bidding process. Therefore, publicly acquired products should have the lowest price, be produced in Brazil and by Brazilians.[6] Since reference biological medicines and biosimilars have the same active ingredient, their industries bid for the same contract. Thus, if the biosimilar industry presents the proposal with the lowest price, it will win the contract, resulting in the automatic substitution for patients who use the reference biological medicine.

Brazilian medical societies and patient associations have opposed to the concept of interchangeability and the practice of automatic substitution, as they consider that changing a reference biological medicine for a biosimilar is an exclusive right of the prescribing physician, and a joint decision with the patient.[2,7,8] However, as mentioned, public procurement considers only the lowest price, not prescription.

Working group

A Working Group for the formulation of the National Policy for Biologic Medicines within the SUS was established in May 2018 to discuss these issues. The Working Group had the responsibility to develop a consolidated report on the discussions of relevant issues in research and development, production, regulation, access, and monitoring of the use of biological products supplied by SUS.[9]

The Department of Pharmaceutical Assistance – DAF (Ministry of Health of Brazil) coordinated the group, which was composed of the following institutions: Brazilian Medical Association - AMB, National Supplementary Health Agency - ANS, National Health Surveillance Agency - Anvisa, Federal Council of Pharmacy - CFF, Federal Council of Medicine - CFM, Chamber of Medicines Market Regulation - CMED, National Council of Health - CNS, National Council of Municipal Health Secretariats - CONASEMS, National Council of Health Secretaries - CONASS, National Commission for Technology Incorporation in SUS - CONITEC, Legal Advisory Service of the Ministry of Health - CONJUR / MS, Department of Industrial Complex and Innovation in Health - DECIIS, Department of Science and Technology - DECIT, Department of Management and Incorporation of Health Technology - DGITS, Department of Logistics - DLOG, Institute for Safe Medication Practices-Brazil, Brazilian Network of Drug Information Centres and Services - REBRACIM, Secretariat of Health Care - SAS, and Secretariat of Health Surveillance - SVS.[10]

The Department of Science and Technology – DECIT of the Ministry of Health of Brazil is responsible for promoting access and use of scientific evidence in a systematic and transparent manner, as well as knowledge translation. Therefore, DECIT provided methodological support to the Working Group's discussions.[11]

Evidence-informed policy-making (EIP)

The EIP approach is intended to ensure that the best available research evidence informs the decision making process, which is context specific.[12] Therefore, DECIT endeavoured to apply the EIP approach on the discussions of the Working Group for the formulation of the National Policy for Biologic Medicines within SUS. Due to the limited period of time for the group's activities, rapid products in the format of evidence brief for health policy were carried out.

Therefore, this rapid review, along with public surveys and internal consultations within the Ministry of Health of Brazil, identified problems related to the use of biological medicines. Then, the Working Group prioritized the problems identified, and decided to address the interchangeability of biological medicines in the form of a Rapid Evidence Synthesis for Policy (not published). Finally, this Rapid Synthesis informed a Dialogue for Policy. The Working Group final report was delivered to the managers of the Ministry of Health, to inform decision-making.

In order to present results of the first stage of the formulation of the National Policy for Biologic Medicines within the SUS, the aim of this rapid review was to map the main barriers related to the use of / access to biological medicines.

Rapid review

The search was carried out in August 2018, in the following databases: Medline (via PubMed), EMBASE, Cochrane Library, and Centre for Reviews and Dissemination (CRD), using a combination for the following index terms and synonyms: ‘biosimilar medicines’, ‘therapeutic equivalence’, ‘regulatory process’, switching , barriers, patient, professional, manager and ‘health system’.

Eligibility criteria included health technology assessments, systematic reviews, narrative reviews, cross-sectional studies, and position statements of organizations addressing barriers related to the use of biological medicines (innovator or biosimilar). Studies evaluating efficacy, effectiveness and safety of biological medicines were excluded.

The literature search identified 640 articles. After duplicates were removed, four independent researchers (CF, RS, BL, DG) conducted the screening of titles and abstracts, and selected 98 articles for full-text reading. In accordance with the commissioners, the review team opted to include only cross-sectional studies. The discussion explored the narrative reviews. After eligibility screening, the team selected nine cross-sectional studies.

One researcher (RS) carried out data extraction was using an electronic data collection form, including the following information: author, year and country of publication, title, target population, sample size, presence of conflict of interest, and main barriers. The team did not assess methodological quality of the included studies. Most studies conducted surveys in interest groups such as patients, health professionals and managers. In general, the studies did not describe the sampling process in detail. The analyses were descriptive, and without evaluation of outcomes. The team considered relevant the presence of conflicts of interest in the context of biological medicines. The team conducted this rapid review in one month. The methods described are in accordance with good practices for conducting rapid review of the World Health Organization.[13]

Results were presented as a narrative summary,[13] reporting the main barriers to access or use biologic medicines in the context of users, providers, management and health system. In addition, a glossary was provided to promote knowledge translation.

The included studies were published between 2013 and 2018, and were conducted in different countries in Europe, United States of America (USA), Argentina, Brazil, Mexico, and New Zealand. The most frequent contexts in the included studies were ‘professional’ and ‘patient’. This rapid review included nine cross-sectional studies, of which most used qualitative methodologies for data collection on barriers to access or use biological medicines. Structured and semi-structured interviews and questionnaires were the main instrument used in these studies. Qualitative methods were used in data analysis, such as theoretical frameworks, specific software, content analysis, and quantitative methods.

Table 1 shows the main characteristics of the studies included in the rapid review, as well as the barriers to access or use biological medicines that were identified, classified according to perspectives (of user, professional, manager and industry).[4,5,22-28]

Table 1. Main characteristics of the studies and barriers to access or use biological medicines.

Author, year, country	Study design	Participants	Perspective	Barriers to access or use
Dylst *et al.* (2014)[4] Belgium	Cross-sectional qualitative study	19	Professional	Lack of confidence in biosimilar medicines - information gap amongst clinicians; Interchangeability and substitution - batch-to-batch variations.
			User	Financial incentives - reference product manufacturers offer discount and additional benefits to hospitals (for users, clinicians, scholarships, funding), influencing professional behaviour at the time of prescription.
			Industry	Financial incentives - reference product manufacturers offer discount and additional benefits to hospitals (for users, clinicians, scholarships, funding), influencing professional behaviour at the time of prescription.
Hemmington *et al.* (2017)[5] New Zealand	Cross-sectional study	110	Professional	Lack of clinical data supporting drug efficacy; Evidence of adverse events or lack of drug efficacy (biosimilar); User is doing well on current treatment; User has a complex medical history.
Inotai *et al.* (2018)[22] Central and Eastern European (CEE) and Commonwealth of Independent States (CIS) countries	Cross-sectional study	200	Professional	Biossimilarity/ equivalence (reference and biosimilar); Possibility of immunogenic reactions.
			User	Limited number of users who can be treated with public reimbursement; Long waiting lists; Limited treatment duration with biological medicines (with reimbursement).
			Manager	Limited number of users who can be treated with public reimbursement.
Laires et al. (2013)[23] Portugal	Cross-sectional qualitative study	34	Professional	Difficulties in accessing primary healthcare services: Mainly in rural areas; limited number of professionals available; unawareness of rheumatoid arthritis by the user; Difficulties in rheumatoid arthritis diagnosis.
			User	Difficulties in accessing specialized healthcare services: Long waiting times for consults; difficult access in rural areas; hospitals are distant; deficient public transportation; Difficulties in accessing primary healthcare services: Mainly in rural areas; limited number of professionals available; unawareness of rheumatoid arthritis by the user;
			Manager	Difficulties in accessing biologics: Prescription by the hospital considered a bureaucratic process; passivity of the user in regards to treatment decisions.

Author, year, country	Study design	Participants	Perspective	Barriers to access or use
Lammers et al. (2014)[24] USA, Brazil, Mexico, Turkey, Russia	Cross-sectional study	217	Professional	Medicine not covered by health insurance; Unavailability of the biologic medicine at clinical settings; Medicine use not recommended by treatment guidelines or protocol commonly followed; User comorbidities could make it impossible to use biosimilar; High out-of-pocket treatment cost for user; Lack of clinical evidence of efficacy and safety; Reimbursement issues.
O'callaghan et al. (2017)[25] Ireland	Cross-sectional study	498	Professional	Concerns in regards to traceability, quality, safety and efficacy profile, immunogenicity, and efficacy in off-label indications.
Sandorff et al. (2015)[26] Argentina, Brazil and Mexico	Literature review and cross-sectional study	10	Professional	Impact drivers for the adoption of biosimilars: Budget, specialist acceptance, disease complexity, clarity of treatment pathway, acute versus chronic treatment duration, intravenous or subcutaneous administration.
			Manager	Impact drivers for the adoption of biosimilars: Budget, specialist acceptance, disease complexity, clarity of treatment pathway, acute versus chronic treatment duration, intravenous or subcutaneous administration.
Sullivan et al. (2017)[27] Germany	Cross-sectional study	161	Professional	Potential side effects and long-term problems; Lack of knowledge about the medicine; High cost of treatment.
			User	Potential side effects and long-term problems; Lack of knowledge about the medicine; High cost of treatment.
Waller et al. (2017)[28] Germany	Cross-sectional study	311	Professional	Potential side effects and long-term problems; Lack of knowledge about the medicine; High cost of treatment.
			User	Potential side effects and long-term problems; Lack of knowledge about the medicine; High cost of treatment.

Perspective of users

The main barriers are the cost of biological medicines and the level of knowledge about this technology in the users' perspective.[4,5,22-28] This result is consistent with narrative reviews. Users are likely to follow the advice of their physician and be 'brand-loyal', therefore, medicine manufacturers' know-how and patients' difficulty to interpret the concept of interchangeability limit access.[14,15] In addition, reference biological medicine companies have strong ties with patient associations, which may limit the uptake of biosimilars, as patients are also likely to follow the advice of associations.[15]

Clinical outcomes associated with disease progression and morbidity influence the user's decision to use a biosimilar rather than a reference biological, as they fear being affected by the switching. Moreover, there is uncertainty regarding long-term efficacy and safety data.[25-28] The level of interest for physicians to participate in clinical trials of biosimilars is lower in countries where reference biological medicines are available and accessible, because of the perceived lack of benefit to patients in this case. If a reference medication is commercially feasible to a patient, the interest to participate in these trials is also jeopardized. Thus, this constitutes a barrier to the uptake of biosimilars.[14]

Users who live in rural areas have to travel long distances to reach major centres, which may represent an important barrier to access (travel time and costs).[23] Consequently, even if biologics are fully reimbursed in many lower-income countries, the real-world utilisation of these medicines may still be low. The same applies to bureaucratic approval processes and waiting lists to access to medicines.[16]

Perspective of professionals

Another barrier was the lack of acceptability of biosimilars by health professionals.[4] In countries where the reference biologicals are available and accessible, health professional may be less interested in biosimilars due to the low number of studies.[14] Health professionals are concerned about to batch-to-batch variability, patients with different characteristics, standards of clinical care of studies comparing biologicals and long-term effects, as immunogenicity.[14]

Other concerns on the use of biosimilars are related to safety, efficacy, immunogenicity, and the ability of regulatory agencies to regulate good clinical practice in the development of these products.[17,25,26]

Physicians are hesitant to accept the benefits of biosimilars as equal to the reference drugs, and they do not directly benefit from the lower cost.[15,18,19] It is argued that incentives for healthcare payers, professionals and users would facilitate the acceptance of biosimilars, as well as the increase of prescribers' knowledge of the subject.[15]

Impossibility of substitution is also a barrier. Other studies consider that repeated switching and substitution without consent of the prescriber are not advised due to traceability.[15] Although there is a consensus that it is possible for treatment-naïve users, there are uncertainties about switching for users during their treatment. Without the possibility of substitution, biosimilars are a choice for new users or as a one-time switch for stable users only.[18] It is argued that the hypothetical risk related to the switch is communicated inappropriately, and may influence the decision of the clinical professional. However, the studies that reported this risk have a low degree of certainty, and this result has not been confirmed by new and more robust trials.[21] A study with a low degree of certainty means that the confidence in the results is limited, and new publications may alter the direction of effect.[20]

A single substitution already raises concern in many countries. In addition, multiple substitutions between reference and biosimilar are widely debated issues, especially at the pharmacy level.[21] Robust studies should be conducted to address multiple substitution, in order to demonstrate the interchangeability through clinical trials with an approach based on the real-world studies, taking into account the existence of several biosimilars in the market.[21]

Perspective of management

Management varies in different countries, through public and private health systems. In this review, managers were considered payers. In the Ministry of Health of Brazil, in the field of public health, DGITS is responsible for the management of health technologies, DAF for the budget for biological medicines, and DLOG for the execution of bidding. ANS is responsible for the management of private health insurance in the country.

In this regard, barriers in the perspective of payers include the acceptability of clinical data used for regulatory approval; payer's ability to induce price competition, extrapolation of indications, variability of efficacy between batch-to-batch and immunogenicity.[22,23,26] Payers in US and European Union markets suggest that, in the absence of a significant price discount for biosimilar, preference will be given to the reference biologic products, demonstrating payers' sensitivity to higher price (for both reference and biosimilar).[14]

Health care decision-makers aim to maximize health gain for the population by improving the allocative efficiency of limited resources. Off-patent drug policies is usually defined as reduction in health expenditures without compromising health outcomes.[16] Therefore, biosimilar policies are different in countries with significant resource constraints, where accessibility of patients to high-cost biologic medicines is limited.[16] Biosimilars may also compete with non-biologic therapies for users who do not have access to biologic medicines.[16]

Perspective of health services and system

Government level

Barriers within the health system were not found in the cross-sectional studies included. However, barriers mentioned in some narrative reviews were considered for discussion.

In an official document, the Australian government listed the following factors that influence biosimilar use: political and regulatory context that determines availability, switching and substitution; uptake of biosimilars by prescribers, pharmacists and users; outcomes resulting from biosimilars outside of clinical trials; and the perspectives of each stakeholder, including factors that influence these perceptions.[17]

A narrative review describes the regulatory process as a barrier to market access of biosimilars, for instance, the public consultation on the guideline of the European Medicines Agency that did not take into consideration the contributions of physicians.[15] This may have contributed to raise concerns and hinder the uptake by physicians, users and user organizations.[15]

The hypothetical risks of substitution between reference biologicals and biosimilar are pointed out in several studies; however, they do not outweigh the potential benefits that society would have with the substitution.[21] In middle-income countries, in the context that the reference biologic product is reimbursed without volume restrictions (amount per user) and access, the main objective of using biosimilars is to generate savings in health expenditures, treat more users and generate health gains.[16]

Private level

Regarding the private sector, specifically the manufacturers of biological products, patent protection is a barrier of legal restraint on market entry of biosimilars, as well as a tool for manufacturers to recover their expenses spent on research and development. Patents can be used to prevent manufacturers of biosimilars from using the same production processes as reference products. Therefore, adapting production processes may lead to differences in the end product, which can affect the quality, efficacy and safety of medicines.[15]

Conclusion

High cost of treatment is consistently presented as a main barrier to the use of biological medicines, and price discount is expected through competition with the uptake of biosimilars. The Working Group should contribute not only to reduce the vulnerability of SUS, but also to improve its sustainability, as well as to increase the population's access to biological medicines. Therefore, further research on potential solutions to the barriers described in this study is necessary.

Competing interests

CMF was member of the Working Group; the other authors declare that they have no competing interests.

References

1. GBD 2016 Brazil Collaborators. Burden of disease in Brazil, 1990-2016: a systematic subnational analysis for the Global Burden of Disease Study; 2016.
2. Fernandes GS SC, Lopes G, Chammas R, Gifoni MAC, Gil RA, Araujo DV. The use of biosimilar medicines in oncology - position statement of the Brazilian Society of Clinical Oncology (SBOC). Braz J Med Biol Res. 2018; 51(3).
3. Vieira, FS. *Evolução do gasto com medicamentos do Sistema Único de Saúde no período de 2010 A 2016. Texto para discussão / Instituto de Pesquisa Econômica Aplicada*. Rio de Janeiro; 2018.
4. Dylst P, Vulto A, Simoens S. Barriers to the uptake of biosimilars and possible solutions: A Belgian case study. PharmacoEconomics. 2014; 32(7):681-91.
5. Hemmington A, Dalbeth N, Jarrett P, Fraser AG, Broom R, Browett P, et al. Medical specialists' attitudes to prescribing biosimilars. Pharmacoepidemiology and Drug Safety. 2017; 26(5):570-7.
6. Presidência da República (BR). Lei nº 8.666, de 21 de junho de 1993, com alterações resultantes da Lei nº 8.883, de 08 de junho de 1994 e da Lei Nº 9.648, de 27 de maio de 1998. Regulamenta o art. 37, inciso XXI, da Constituição Federal, institui normas para licitações e contratos da Administração Pública e dá outras providências.
7. Azevedo VF, Meirelles Ede S, Kochen Jde A, Medeiros AC, Miszputen SJ, Teixeira FV, Damião AO, Kotze PG, Romiti R, Arnone M, Magalhães RF, Maia CP, de Carvalho AV. Recommendations on the use of biosimilars by the Brazilian Society of Rheumatology, Brazilian Society of Dermatology, Brazilian Federation of Gastroenterology and Brazilian Study Group on Inflammatory Bowel Disease--Focus on clinical evaluation of monoclonal antibodies and fusion proteins used in the treatment of autoimmune diseases. Autoimmun Rev. 2015; 14(9):769-73.
8. Torres P. *Consenso brasileiro multi-institucional de pacientes sobre medicamentos biossimilares. Jornal Brasileiro de Economia da Saúde*. 2017; 9(1):39-43.
9. *Ministério da Saúde (BR). Gabinete do Ministro. Portaria nº 1.160, de 3 de maio de 2018.* Brasília, DF; 2018.
10. *Ministério da Saúde (BR). Grupo de Trabalho para discussão e formulação da Política Nacional de Medicamentos Biológicos no âmbito do Sistema Único de Saúde (SUS). Regimento Interno. Brasília* [Internet] 2018 [accessed 9 August 2019]. Available at: http://portalarquivos2.saude.gov.br/images/pdf/2018/outubro/19/3-Regimento-GT-Biologicos.pdf.
11. *Ministério da Saúde (BR). Secretaria-Executiva. Subsecretaria de Assuntos Administrativos. Regimento Interno do Ministério da Saúde : Decreto nº 8.901, de 10 de novembro de 2016 / Ministério da Saúde, Secretaria-Executiva, Subsecretaria de Assuntos Administrativos. – Brasília : Ministério da Saúde*; 2017.
12. Oxman AD, Lavis JN, Lewin S, Fretheim A. SUPPORT Tools for evidence-informed health Policymaking (STP) 1: What is evidence-informed policymaking? Health Res Policy Syst. 2009; 16(7).
13. Tricco AC, Langlois EV, Straus SE, editors. Rapid reviews to strengthen health policy and systems: a practical guide. Geneva: World Health Organization; 2017.
14. Rompas SG, T; Amanuel, S; Coutinho, V; Lai, Z; Antonini, P; Murphy, MF. Demonstrating Value for Biosimilars: A Conceptual Framework. Am Health Drug Benefits. 2015; 8(3):129-39.
15. Moorkens E, Jonker-Exler C, Huys I, Declerck P, Simoens S, Vulto AG. Overcoming barriers to the market access of biosimilars in the European union: The case of biosimilar monoclonal antibodies. Frontiers in Pharmacology. 2016; 7.
16. Inotai AC, M; D, Vitezic; Francetic, I; Tesar, T; Bochenek, T; Lorenzovici, L; Dylst, P; Kalo, Z Policy Practices to Maximise Social Benefit from Biosimilars. Journal of Bioequivalence & Bioavailability. 2017; 09(04).

17. Australian Government. Department of Health. Literature Review of International Biosimilar Medicines: Update; 2017.
18. Rickwood SD, S. Searching for Terra Firma in the Biosimilars and Non-original Biologics Market. IMS Health; 2013.
19. Prescribing and Research in Medicines Management (UK and Ireland) Annual Conference 2017. Pharmacoepidemiology and Drug Safety. 2017; 26.
20. *Diretrizes metodológicas : Sistema GRADE – Manual de graduação da qualidade da evidência e força de recomendação para tomada de decisão em saúde / Ministério da Saúde, Secretaria de Ciência, Tecnologia e Insumos Estratégicos, Departamento de Ciência e Tecnologia. – Brasília : Ministério da Saúde*; 2014.
21. Inotai A, Prins CPJ, Csanádi M, Vitezic D, Codreanu C, Kaló Z. Is there a reason for concern or is it just hype?–A systematic literature review of the clinical consequences of switching from originator biologics to biosimilars. Expert Opinion on Biological Therapy. 2017; 17(8):915-26.
22. Inotai A, Csanadi M, Petrova G, Dimitrova M, Bochenek T, Tesar T, York K, Fuksa L, Kostyuk A, Lorenzovici L, Omelyanovskiy V, Egyed K, Kalo Z. Patient Access, Unmet Medical Need, Expected Benefits, and Concerns Related to the Utilisation of Biosimilars in Eastern European Countries: A Survey of Experts. Biomed Res Int; 2018.
23. Laires PA, Mesquita R, Veloso L, Martins AP, Cernadas R, Fonseca JE. Patient's access to healthcare and treatment in rheumatoid arthritis: the views of stakeholders in Portugal. BMC Musculoskelet Disord. 2013; 14:279.
24. Lammers P, Criscitiello C, Curigliano G, Jacobs I. Barriers to the Use of Trastuzumab for HER2+ Breast Cancer and the Potential Impact of Biosimilars: A Physician Survey in the United States and Emerging Markets. Pharmaceuticals (Basel). 2014;7(9):943-53.
25. O'Callaghan J, Bermingham M, Leonard M, Hallinan F, Morris JM, Moore U, Griffin BT. Assessing awareness and attitudes of healthcare professionals on the use of biosimilar medicines: A survey of physicians and pharmacists in Ireland. Regul Toxicol Pharmacol. 2017; 88:252-261.
26. Sandorff E, Pinheiro AV, Bruni DS, Halbert R, Azevedo VF. Payer and physician evidence and discount requirements for biosimilars in three Latin American countries. GaBI Journal). 2015; 4(1):11-6.
27. Sullivan E, Piercy J, Waller J, Black CM, Kachroo S. Assessing gastroenterologist and patient acceptance of biosimilars in ulcerative colitis and Crohn's disease across Germany. PLoS One. 2017; 12(4).
28. Waller J, Sullivan E, Piercy J, Black CM, Kachroo S. Assessing physician and patient acceptance of infliximab biosimilars in rheumatoid arthritis, ankylosing spondyloarthritis and psoriatic arthritis across Germany. Patient Prefer Adherence. 2017; 11:519-530.

Compulsive Hoarding: an integrative review of the potential risk of spread of vector-borne and other zoonotic diseases[I]

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Abstract

People with hoarding disorder are known to gather objects or pets excessively in a disorderly and disorganized way. In general, they suffer from lack of self-care and live in places that are not sanitary, which impact their own quality of life and that of their families and community. In the community, there is a potential risk of some endemic vector-borne diseases transmitted by unpleasant and poisonous host. The objective of the study was to carry out a literature review on hoarding of materials / objects (Diogenes syndrome) and domestic animals (Noah syndrome), and its impact on health with a focus on endemic diseases. A structured question guided the method, with a search of terms in three languages, in PubMed, Embase, Cochrane, Web of Science, VHL Portal Regional, Google Scholar, and also grey literature. The analysis included sixteen documents that pointed to the need for multiprofessional care for the treatment of these patients. With this review we hope to contribute with a better understanding of the hoarding problem, regarding the control necessary to prevent endemic diseases at different levels of health care, with a perspective to develop an effective flow of multiprofessional care.

Keywords: Hoarding disorder, Diogenes syndrome, Noah syndrome.

Introduction

People with hoarding disorder are known to gather objects or pets excessively in a disorderly and disorganized way, with no relation to their monetary value, but with great distress and resistance to let go of possessions. In general, they suffer from lack of self-care and live in places that are not sanitary, which impact their own quality of life and that of their families and community.

It is necessary to differentiate hoarding from collecting. While collecting involves the acquisition of one type of item in large quantities, stored in an organised manner, to be shown as collections to other people with pleasure, hoarding involves the acquisition of many different possessions, usually stored in a disorganised manner, and hoarders do not allow anyone to access them.[1]

These manifestations and disorders are known in medical environment as Diogenes syndrome for object hoarding, and Noah syndrome for animal hoarding.[2-4] There are other designations to hoarding disorder, including 'messy house syndrome', 'senile self-neglect', 'senile recluse syndrome', and 'severe domestic squalor'.[5] According to Khan, 'severe domestic squalor' could be a better descriptor for both Diogenes and Noah syndromes.[6]

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Diogenes syndrome was named after the Greek philosopher Diogenes of Sinope (323-404 BC) who, unlike hoarders, chose to live in poverty, in a barrel, and eschewed social norms, relying on the philosophical current of detachment from material goods, and believing that happiness is not dependent on external factors.[7,8] This syndrome was first described in 1975, characterized by people with paranoid behaviour who neglect their personal hygiene and of their housing. It may affect any social class, men and women, including children and adolescents.[8] Noah syndrome is a variant of Diogenes syndrome, and its predisposing factors are situations of psychosocial stress and loneliness, characterized by animal hoarding practices.[8]

In 2013, hoarding disorder was defined as a psychopathology and included in the Diagnostic and Statistical Manual of Mental Disorders of the American Psychiatric Association (APA).[9] There are several hypotheses for the aetiology of this psychopathology, such as: Personality disorder (isolation, distrust, aggressive behaviour, cunning, emotional lability, and tendency to distort reality), dementia of the central lobe of the brain, obsessive-compulsive disorder, psychiatric problem associated with collecting practices. Triggering factors of the pathology may be associated with biological, psychological, social or age-related stressors, or alcohol and drug abuse.[8]

Some factors and characteristics may be associated with this collecting behaviour, contributing to the diagnosis:[9]

- Persistent difficulty in discarding or parting with possessions, regardless of their true value. This difficulty is due to the strong urges to 'save' items and distress associated with discarding;
- This difficulty results in the accumulation of possessions that congest and clutter living areas to the point that the intended use of the areas is substantially compromised. When keeping a particular place organized, it is usually by pressure from family members or authorities;
- Significant suffering, because this behaviour interferes with social life;
- Difficulty in maintaining self-care, hygiene and lifestyle, such as: eating properly, maintaining personal hygiene, removing household waste, organizing and cleaning the living space;
- In the case of psychoactive substance use, difficult to organize and clean the living space due to side effects of drugs.
- Other factors associated with animal hoarding behaviour may contribute to the diagnosis:[10]
- Difficulty in providing minimum standards of space, sanitation, nutrition and care for the animals, as well as difficulty of controlling the obsession to maintain a collection of animals, denying or minimizing the problems and living conditions for people and animals;
- Inability to recognize the effects of this behaviour on the welfare of the animal, family members and environment.

Epidemiological studies have indicated a great variation on prevalence data of hoarders in the population, for example: Massachusetts, USA, with 26 hoarders per 100,000 residents, Germany with 4.6% and Italy with 6.0% of the population.[6-8,11] Age is an important factor, and hoarding disorder tends to increase where there are many individuals aged 60 years and over, such as in Sydney, Australia, where the prevalence is 1/1,000 residents in community-dwelling elderly population, according to Snowdon's study.[12]

The unhealthy environments of hoarders are favourable for the proliferation of the *Aedes* *aegypti* mosquito[13], and can influence on cases of dengue, since solid waste in the house or yard where water may be stored may provide breeding sites for mosquitoes.[14] Moreover, accumulation of materials and objects provides also favourable conditions for proliferation of insects, which are food for other animals such as scorpions and rodents. Houses of hoarders are at risk not only of vectors of urban arboviruses and other insects and animals of medical importance, but also other zoonoses such as visceral leishmaniasis and Brazilian spotted fever.

Objective

To conduct an integrative review of scientific literature, government policies and legal documents on hoarding of materials and objects (Diogenes syndrome) and domestic animals (Noah syndrome), its impact on public health, specifically for endemic vector-borne diseases transmitted by unpleasant and poisonous host.

Method

The search for evidence was based on a structured question: 'Do hoarders of objects and / or materials and / or animals increase the risk of endemic vector-borne diseases transmitted by intermediate hosts (dogs and cats) and unpleasant hosts (cockroaches, rats, scorpions, etc.)?'

The search descriptors were obtained from the reading of studies collected in a non-systematic way, which addressed hoarders, since each country defines hoarders with specific descriptors, such as: squalor, extreme squalor, self-neglect, among others. This was useful to expand the review process.

The searches were carried out in the following databases: PubMed, Embase, Web of Science, VHL Regional Portal, Cochrane, Google Scholar, as well as in grey literature resources such as government guidelines and theses.

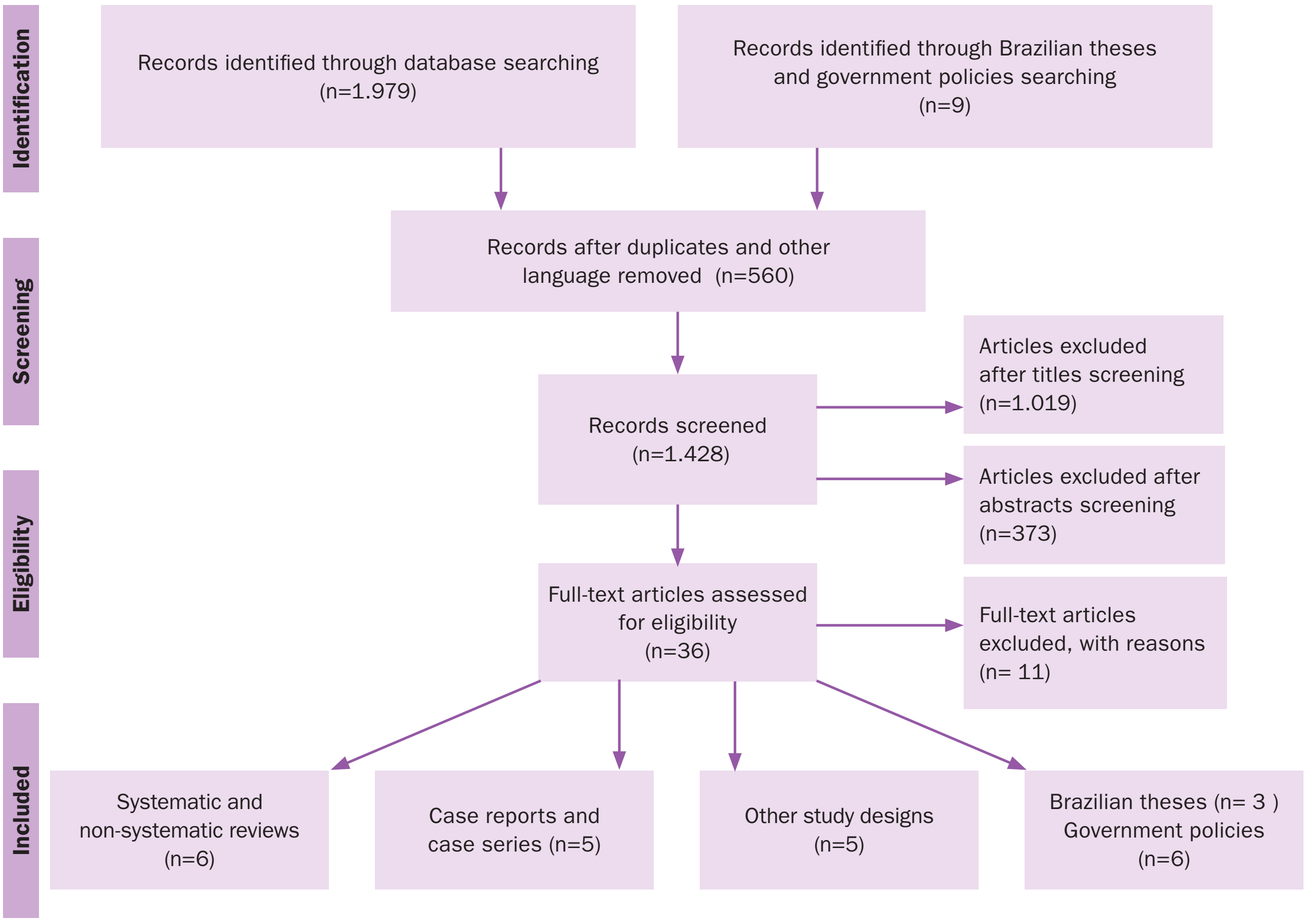
No time period restriction was applied, but the search, conducted until June 2018, was restricted to the Portuguese, Spanish and English languages. In order of priority, considering the level of evidence, searches focused on meta-analyses, systematic reviews, randomized controlled trials, and cohort studies, including Health Technology Assessment reports, government policies, theses, and other relevant documents, even with low level of evidence such as case studies.

Study selection and data extraction were conducted by the research team members independently. Disagreements were resolved by consensus. First, duplicates and articles in languages other than those mentioned above were removed, followed by titles and abstracts screening along with reasons for excluding. The systematic reviews selected were assessed using the AMSTAR tool – A Measurement Tool to Assess the Methodological Quality of Systematic Reviews.[15]

Results

It is noteworthy that there is a large scientific production on hoarding. Many of the studies have not always conformed to quality criteria, but the quantity indicates that the scientific community has been concerned with this issue and seeking for evidence to address it. The literature search showed that research has been conducted to explore this issue in-depth since the year 2000. Many articles have been focusing on clinical practice with patients with hoarding disorder: aetiology, diagnosis, treatment, evolution. However, there have been few on public health and even less on vectors, intermediate and unpleasant and poisonous hosts, which are the focus of this review.

Sixteen articles, three theses and six government policies were selected for analysis (Figure 1).



After reading and analysis of included articles, three major thematic categories were defined: *Hoarders' health; health of family members and surrounding area; and public health problems*. These findings are presented below and in more detail in Table 1.

Hoarders' health

The health of hoarders was described in the studies as very compromised, due to the exposure to unhealthy environments because of accumulation of items or animals, as well as the suffering caused by a psychiatric condition without proper treatment. Usually hoarders are seen, even by health staff, as someone with an inappropriate social behaviour, unrelated to a mental health condition which is recognized by medicine as the Diogenes syndrome or Noah syndrome.

A non-systematic review by Stumpf and colleagues,[8] conducted in Brazil, reported on the impact this psychiatric pathology has on the social and economic environment, and the insecurity and risk of infection not only to hoarders, but also their surrounding community. According to Araujo and Santos,[16] when therapeutic results are obtained, health staff recognize that there is a health problem, and not just a stereotypical behaviour of the individual. In this study, the Singular Therapeutic Project was successfully used as treatment.

In the last 10 years, there has clearly been an evolution of studies on the aetiology of these syndromes, with evidence pointing to family, genetic and cognitive factors, and traumatic experiences. In relation to associated conditions, psychiatric disorders, such as obsessive-compulsive disorder, depression and anxiety, has been the most commonly reported, [17] but rheumatic diseases with reduced mobility and substance dependence have also been associated to personal (self-neglect) and environmental neglect.[18]

In the included studies, hoarders' characteristics were well described, highlighting clutter taking over the person's life and space. Moreover, it was also presented measurement tools, such as 'rating scales' on hoarding severity, mainly in the government policy documents.[1,19,20] The highest degree of hoarding has been established as the living space becoming so cluttered that it is not possible to move around without bumping into objects and animals, at risk of being buried under piles of stuff, in the case of a patient with severe Diogenes syndrome.[8] In addition, neglect of personal hygiene (self-neglect), of the living environment, and of animals was exhaustively addressed in all included studies.

Regarding gender, the studies[18,21,23] indicated females as the most affected by these syndromes, except one study that pointed to males.[13] In regard to age, all studies that investigated this variable reported it as an important predisposing factor for triggering hoarding disorder, and the rate of hoarding in those aged over 49 and over 60 years old were greater.

Some studies reported on forms of treatment of patients with hoarding disorder, and cognitive therapy and medication were indicated to its control.[16,23,24] The removal of items or animals without medical and psychological treatment usually has higher fail and recurrence rates, soon after cleaning the environment,[1] and hoarders may be unwilling to accept treatment when they do not recognise their health problem.

Health of family members and surrounding area

All studies indicated a social isolation of hoarders related to two main reasons: First, they are embarrassed about their disorganization, unlike collectors who always like to show other people their collections;[8] and secondly, withdrawal from family members and significant others due to foul odour of the property, difficulty moving around, infestation of cockroaches and rodents, among other factors.[25,26]

One study found that there are families of hoarders, with all members hoarding objects or animals, but usually hoarders are lonely individuals who lose interaction with family and caregivers, who may require legal intervention for them.[22] The withdrawal of family members, neighbours, and significant others, although it is understandable, is not a good attitude since hoarders need their support to undergo treatment.[18,27] One of the included studies reported on the lack of love relationship as an aggravating factor for isolation and increasing hoarding.[26]

It is not only the hoarders' living spaces that are affected by the deleterious effect of accumulation of possessions and animals, but also their surrounding community, who are aware about the unhealthy and unsafe situations that result from hoarding.[17,18,22,25,28-29] All included studies reported on the risks to surrounding communities related to infestation of cockroaches, rodents, poisonous animals, snakes, insects and mosquitoes, and also to the foul odour that, according to one study, may be toxic when inhaled.[18, 21,30, 31]

At least three studies reported that, in regions endemic for visceral leishmaniasis, people are at greater risk of contracting the disease when there are hoarders in the neighbourhood, especially dog and cat hoarding.[17,32,33,34] In this case, it becomes a major public health problem, as the disease has no cure in animals, and can be fatal in humans.

Public health problems

The safety issue related to the risk of fire in the homes of hoarders and their neighbours is addressed by several studies,[8,18,20,22,28,30,32] which causes concern to fire-fighters and local managers. Coverage policies usually address safety issues as fundamental, mainly related to fire. Neighbours and surrounding community members have been filing lawsuits related to the hoarding problem, and the main reason indicated has been the fire hazard.[1,21,22,25,31,34]

Landlords also bear the burden of having hoarding tenants, with substantial economic losses when they leave the property in a poor state of repair and inadequate cleaning, as well as depreciation of the commercial value.[30]

Therefore, the risk that hoarders pose to themselves, to their surrounding community and to the whole environment is evident, becoming a relevant public health problem. All included studies reported on the variety of risks, and indicated diagnosis, treatment and taking care of the mental health of the patients with hoarding as necessary to solve the problem. Specific and non-integrated interventions, such as removal of possessions by enforcement of the law and guidelines on household clean-up, are ineffective for hoarders who lack the ability to recognise their condition. Other interventions can lead to a breakdown of trust between hoarders and those who they trust (caregiver, family members, neighbours, local health staff, and significant others).[20]

Integrated treatments are fundamental, across a range of areas including health, environment, fire brigade, among others.[8,18,20-22,28,31,35,36] Social workers play an important role in bringing the family closer to the hoarders and making interventions more effective.[8,20,28,29]

Lawsuits and coverage policies

Data from the Brazilian justice, according to Jusbrasil website,[37] indicated that until 2018 there were 6,739 lawsuits focusing on compulsive hoarding, either objects or animal hoarding. Analysing some judicial decisions, it is noteworthy that the main focus is on the immediate removal of objects or animals from the property by public agencies, with little attention paid to comprehensive care for hoarders.

Six documents on public policies addressing hoarders were analysed: Two of Australia,[1,38] two of United States,[32,35] and two of United Kingdom.[20,36] In summary, according to these public policies, interdisciplinary care should be provided to hoarders. Health interventions that are fragmented and disconnected from the social network where these people live are likely to fail.

Table 1. Thematic synthesis of the main findings of the selected studies.

Authors (year)	Study design (methodological quality)	Thematic classification		
		Hoarders' health	**Health of family members and surrounding area**	**Public health problems**
		Review studies		
Frost et al. (2011)[21]	Narrative review	The main characteristics of hoarding disorder are excessive accumulation of objects or animals with significant clutter and disorganization in the living areas, which is related to health problems, especially mental health conditions.	Negative family relationships can deepen the hoarder problem. Withdrawal of family members due to the lack of hygiene conditions in which hoarders live.	The consequences for personal and public health are substantial. Animal health is also greatly compromised.

Authors (year)	Study design (methodological quality)	Thematic classification		
		Hoarders' health	**Health of family members and surrounding area**	**Public health problems**
Review studies				
Gahr et al. (2017)[17]	Narrative review	Hoarders have associated mental health condition. The following mental health conditions were reported: obsessive-compulsive disorder, personality disorder, depression and alcohol abuse.	Relevance of social isolation and break-up of family ties related to animal hoarding.	Hoarders' neglect of the hygiene conditions under which the animals are held poses a public health risk, particularly regarding diseases that affect animals and may spread to the community.
Ong et al. (2015)[27]	Systematic review (AMSTAR 9/11)	Focus on the quality of life of hoarders. Among the variables explored, the ability to perform daily activities was the most compromised.	Besides the hoarders' personal problem, their insertion in neighbourhood is also compromised. Hoarders lose control of the functional capacity to manage family and home life. They have difficulty or complete inability to receive visits form friends or to keep a love relationship.	Hoarders persist in living in dirty and cluttered environments, thus, increasing the risk of contamination due to cleaning procrastination.
Roane et al. (2017)[18]	Systematic review (AMSTAR 8/11)	The elderly individuals with hoarding have a persistent difficulty with discarding items, accumulating clutter, which congests living areas. Anxiety disorder and depression are the psychiatric conditions most commonly associated with this group of individuals, mostly females.	Ruptures in family and with significant others may leave hoarders without caregivers, causing negative impact on the environment and increasing infestation with rodents and insects.	Congested and cluttered living spaces pose a danger to the elderly due to the high risk of fall. In regard to collective health, this cluttering also poses a danger to the house and its surroundings due to the high risk of fire and infestation with insects, rodents and other animals, with potential negative impact on public health.
Tolin et al. (2015)[39]	Narrative review	Since 2000, animal and / or object hoarding disorder has become an official diagnosis in DSM-5.	Unhealthy environment in which hoarders live poses a high risk of disease. The family can trigger the problem and, then, withdraw from it.	The risk to public health is serious, mainly related to overcrowding of animals and extremely unhealthy conditions at home.
Stumpf, Rocha (2010)[8]	Case report with narrative review	Hoarding disorder has been always associated to mental health conditions, but it may also have other aetiologies: Family, genetic and cognitive factors, and traumatic experiences.	Hoarding disorder commonly leads to social isolation that feeds back into the process, as it reduces social life. The community is affected, especially when food waste that is thrown away serves as food for rodents.	It poses a significant risk when the house is filthy with faeces and urine, as well as insecurity, as cluttering represents a risk of fire or even being buried.

Authors (year)	Study design (methodological quality)	Thematic classification		
		Hoarders' health	Health of family members and surrounding area	Public health problems
Case reports and case series				
Fond et al. (2011)[26]	Report and discussion of a case	Case study of a hoarder suffering from depression whose symptoms started after her husband's suicide. She had sleep disturbance, periods of euphoria and grandiose delusions.	Her sons asked for guardianship. The offensive smell of the house and herself was unbearable and kept friends and family members away. She had never washed her two dogs.	Animal and object hoarding poses a great risk to society.
Hoarding of Animals Research Consortium – HARC (2002)[22]	Descriptive analysis of case reports	Focus on Noah syndrome and its association with psychopathologies. Identification of animal hoarding in the community may be a sentinel for a range of medical, social, and economic problems in the region.	The study highlights three cases of a whole family of hoarders, not just one member. Most of them were elderly people who had been withdrawal from family and friends.	The public health of surrounding areas is extremely compromised in many aspects. The best intervention should be a task force with the participation of many diverse civil society stakeholders and organisations.
Lee, Lo Giudice (2012)[25]	Report and discussion of three cases	Hoarders are individuals suffering from self-neglect, defined as failure to engage in activities that a given culture deems necessary to maintain a socially accepted standard of personal and household hygiene, and health status.	Hoarders do not have the ability to participate in family and community social life, taking into account that they neglect their personal hygiene (e.g., they are malodorous) and of their housing.	The public health of surrounding areas is extremely compromised
Lenders et al. (2015)[28]	Retrospective analysis of a case series	Focus on the analysis of the domestic environment of 186 patients with hoarding disorder in the city of Dortmund, in Germany. All patients suffered from psychiatric illnesses: addiction, psychosis and depression.	Many patients were unwilling to accept help from family or even from health care team. Around 50 patients refused treatment. A big problem reported was the risk of fall due to congested living spaces.	These patients compromise their surroundings with pests, bad smell and imminent risk of fire.
Raeburn et al. (2015)[29]	Report and discussion of a case	People with hoarding disorder live in squalor, have associated chronic mental disorders, and can compromise their own health and safety.	Hoarders' propensity to live in squalor becomes a public health issue and presents substantial challenges to family, carers, social service agencies and clinical mental health services.	Hoarders pose a public health problem in any community.

Authors (year)	Study design (methodological quality)	Thematic classification		
		Hoarders' health	**Health of family members and surrounding area**	**Public health problems**
Studies with different methodologies				
Araujo, Santos (2015)[16]	Participant observation	In the neighbourhood of Cocaia, in the city of Guarulhos, São Paulo State, Brazil, elderly with hoarding disorder were followed up with home visits and treated with an intervention of the Single Therapeutic Project.	Compulsive hoarding disorder poses a risk of various diseases to hoarders as well as their surrounding community. Notification is usually done by neighbours or family members of these patients.	Hoarders constantly put themselves and their surrounding community at risk of respiratory disease, leptospirosis, dengue, pest infestation, and even fires.
Caixeta et al. (2011)[13]	Descriptive analysis of the application of the questionnaire Hoarding Rating Scale-Interview	Hoarders are individuals with psychiatric disorders, mainly obsessive-compulsive disorder. Identification of compulsive hoarding may contribute to health teams identify potential risk of proliferation of vectors.	Risk of dengue due to proliferation of vectors in surrounding areas of hoarders' living spaces.	Compulsive hoarding creates favourable environmental conditions to the dengue proliferation, mainly due to the habit of accumulating garbage and keeping trash in living spaces.
Cramer e Vols (2016)[30]	Comparative analysis of legislation in two countries	Hoarders' health is compromised, and hoarding should be addressed by the legal system, taking into consideration neighbour law, housing law, as well as administrative law.	Hoarding disorder does not only have consequences for the sufferers, but affects those living around them.	Hoarding often creates unsanitary conditions that are characterised by rodents, cockroaches, and other insects' infestations. Odours are also a common complaint where hoarding occurs.
Cunha et al (2017)[40]	Cross-sectional ecological study	Hoarders have a particular mental disorder characterized by a persistent difficulty with discarding of possessions, regardless of their value, with serious hindrance to living spaces and harmful consequences for the person, their family and community. Other mental disorders may cause hoarding behaviour, such as: Neurodevelopmental disorders, schizophrenia, psychotic disorders, major depressive episodes, obsessive-compulsive disorder, and cognitive disorders.	Living spaces obstructed, unsanitary conditions, odour and odd behaviour can make social life difficult for hoarders, causing a negative impact on individual and collective health with higher risk of infections.	Garbage and other types of unsanitary material directly impact on the environment where hoarders live, posing a health risk to the community.
Day, McCarthy (2016)[31]	Analysis of community interventions	Patients with Noah syndrome usually have associated conditions, with the following manifestations: attachment, obsession and compulsion, delirium, dementia, attachment disorders, alcohol abuse, persecutory delusions, personal (self-neglect) and environmental neglect. This neglect compromises their own health as well as the health of their animals.	Self-neglect affects family and community social life.	It is a serious public health problem that can impact the health, welfare and safety of individuals, families, communities, and animals. The treatment of mental health conditions has been neglected by health care providers, and almost always patients with Noah syndrome are sued for animal hoarding.

Discussion

This review aimed to contribute to a better understanding of the behavioural dynamics of individuals with Diogenes and Noah syndromes with regard to public health, especially the control necessary to avoid endemic vector-borne diseases transmitted by intermediate and unpleasant and poisonous host. It corroborated the empirical statements of health professionals in the field of zoonoses, vector control, family health, mental health and social care, regarding the association of hoarding of objects and animals with the spread of diseases. Moreover, it showed that hoarders are becoming common in our social environment, and their unhealthy living conditions put themselves and their communities at risk of diseases.

There are many issues addressed in the included studies, such as the relation between the population ageing and the global increase in the number of hoarders, and the lack of access to medication treatment and other therapies, highlighting that further research is needed to address knowledge gaps related to patients with hoarding disorder.

Prevalence studies on Diogenes and Noah syndromes should be conducted to promote public health policies for these patients, as well as to plan the costs for their care. Another knowledge gap relates to the treatment that can be offered to these patients, who needs comprehensive health care and not the forced removal of objects and animals.

The systematic review of Thompson[23] on the interventions that can be implemented for the treatment of hoarders included twenty studies, showing that behavioural therapies, medication and support from family members resulted in a statistically significant improvement for patients with hoarding disorder. Ong and colleagues[27], in turn, showed that hoarders with diagnosis of associated obsessive disorders were less successful in medication treatment compared with those who had anxiety disorder.

Muroff and Steketee,[24] in a pilot study with seven patients with hoarding disorder receiving cognitive-behavioural therapy, reported on the need to intensify treatment with more therapies. Using a webcam for monitoring the progress of treatment, the researchers oriented the patients to present their own evolution for clinicians who were following them at a distance. They concluded that this process was successful in making patients willing to follow the guidance on the hoarding, and effective at reducing this practice. This type of virtual follow-up are lower cost; however, the consent of volunteers is a major problem to be addressed for its further use.

Kyrios and colleagues[41]examined 89 patients treated with cognitive-behavioural therapy and compared with an cohort of hoarders without any treatment. The conclusion indicated significant improvement of the patient receiving mental health treatment. Moreover, some triggers of the hoarding disorder could be identified, such as mood factors considered important in the aetiology and maintenance of accumulation, negative emotional states, anxiety, sadness, guilt and anger.

A research agenda should be created urgently, so that new studies may address the knowledge gaps about patients with hoarding disorder, highlighted in this review, for instance, prevalence studies on hoarding disorder to support decision-making by public health managers, planning the costs for their care, as well as studies on better treatments for these patients.

The included studies indicated that there is no single formula for treatment, therefore, it is necessary multidisciplinary and multi-sector teams aiming at reducing the disease burden and enhancing individual and collective health.

It is noteworthy to mention that in some countries there are established protocols of embracement and comprehensive care to these patients. In Brazil, there is a need for public policy that will promote multiprofessional and comprehensive care, and a continuum of care from primary care to specialized care.

References

1. Australia. South Australian Department for Health and Ageing. Health Protection Programs. A foot in the door: stepping towards solutions to resolve incidents of severe domestic squalor in South. 2013:1-62.
2. Irvine JDC, Kingsley N. Recognizing Diogenes syndrome: a case report. BMC Res Notes. 2014; 276(7):2-4.
3. Saldarriaga-Cantillo A, Rivas Nieto JC. Noah Syndrome: A Variant of Diogenes Syndrome Accompanied by Animal Hoarding Practices. J Elder Abuse Negl. 2015; 27(3):270-5.
4. Frank C, Misiaszek B. Approach to hoarding in family medicine Beyond reality television. Can Fam Physician. 2012; 58(10):1087–1091.
5. Almeida R, Ribeiro O. *Síndrome de Diógenes: revisão sistemática da literatura. Rev. Port Saude Publica*. 2012; 30(1):89–99.
6. Khan S. Diogenes Syndrome: A special manifestation of Hoarding Disorder. The American Journal Psychiatric. 2017; 12(8):9-11.
7. Holm M. Severe domestic squalor. Tidsskr Nor Laegeforen. 2010; 130(24):2469-71.
8. Stumpf BP, Rocha FL. *Síndrome de Diógenes. J. bras. psiquiatr*. 2010; 59(2):156-159.
9. Schmidt DR, Méa CPD, Wagner MF. *Transtorno da Acumulação: características clínicas e epidemiológicas. Revista CES Psicologia*. 2014; 7(2):27-43.
10. Büscher TP, Dyson J, Cowdell F. The effects of hoarding disorder on families: an integrative review. Journal of Psychiatric and Mental Health Nursing. 2014; 21(6):491-8.
11. Dong XQ, Simon MA, Evans DA. Prevalence of Self-Neglect across Gender, Race, and Socioeconomic Status: Findings from the Chicago Health and Aging Project. Gerontologia. 2012; 58(3):258-268.
12. Snowdon J, Halliday G. A study of severe domestic squalor: 173 cases referred to an old age psychiatry service. Int Psychogeriatr. 2011; 23(2):308-14.
13. Caixeta L, Azevedo PVB, Caixeta M, Reimer CHR. Psychiatry disorders and dengue: Is there a relationship?. Arq. Neuro-Psiquiatr. 2011; 69(6):920-923.
14. Paiva SA, Silva SCS, Aguiar VG. *Dengue Versus Lixo uma Problemática no Jardim Nova Esperança. In: III Congresso Brasileiro de Gestão Ambiental*. 2012:1-8.
15. Shea BJ, Grimshaw JM, Wells GA, Boers M, Andersson N, Hamel C, et al. Development of AMSTAR: a measurement tool to assess the methodological quality of systematic reviews. BMC Med Res Methodol [Internet]. 2007:1-7.
16. Araujo ENP de, Santos VG. *Transtorno de acumulação compulsiva de idosos: Possibilidades de cuidados e questões de saúde pública. Revista Kairós Gerontologia*. 2015; 18(4):81-100.
17. Gahr M, Connemann BJ, Freudenmann RW, Kölle MA, Schönfeldt- Lecuona CJ. Hoarding: A Mental Disorder with Implications for Public Health. Fortschr Neurol Psychiatr. 2014; 82:330–336.
18. Roane DM, Sherratt ALJ, Wilson GS. Hoarding in the elderly: a critical review of the recent literature. Int Psychogeriatr. 2017; 29(7):1077-1084.
19. Beyond Overwhelmed. Catholic Community Services NSW/ACT. Identifying pathways to deliver more effective services for people & their pets affected by hoarding and squalor across. 2012:104.
20. Lambeth. Self-Neglect and Hoarding Guidance. 2017; (1):38.
21. Frost RO, Patronek G, Rosenfield E. Comparison of object and animal hoarding. Depress Anxiety. 2011; 28(10):885–891.
22. Hoarding Animals Research Consortium (HARC). Implications of Animal Hoarding Animal. Work. 2002; 27(2):125-131.
23. Thompson C, Fernández de la Cruz L, Mataix-Cols D, Onwumere J. A systematic review and quality assessment of psychological, pharmacological, and family-based interventions for hoarding disorder. Asian J Psychiatr. 2017; 27(6):53-66.
24. Muroff J, Steketee G. Pilot trial of cognitive and behavioral treatment for hoarding disorder delivered via webcam: Feasibility and preliminary outcomes. Journal of Obsessive-Compulsive and Related Disorders. 2018; 18:18–24.
25. Lee SM, LoGiudice D. Phenomenology of squalor, hoarding and self-neglect: an Australian aged care perspective. Internal Medicine Journal Royal Australasian College of Physicians. 2012; 98-101.

26. Fond G, Jollant F, Abbar M. The need to consider mood disorders, and especially chronic mania, in cases of Diogenes syndrome (squalor syndrome). International Psychogeriatrics. 2011; 23(3):505–507.
27. Ong C, Pang S, Sagayadevan V, Chong SA, Subramaniam M. Functioning and quality of life in hoarding: A systematic review. Journal of Anxiety Disorders. 2015; 32:17-30.
28. Lenders T, Kuster J, Bispinck R. Management of Uninhabitable Homes – Investigation of 186 Cases of Hoarding, Domestic Neglect and Squalor in Dortmund. Fortschr Neurol Psychiatr. 2015; 83:695–701.
29. Raeburn T, Hungerford C, Escott P, Cleary M. Supporting recovery from hoarding and squalor: insights from a community case study. Journal of Psychiatric and Mental Health Nursing. 2015; 22:634–639.
30. Cramer R, Vols M. Hoarding disorder and the legal system: A comparative analysis of South African and Dutch law. International Journal of Law and Psychiatry. 2016; 49:114–123.
31. Day MR, McCarthy G. Animal Hoarding: A Serious Public Health Issue. Ann Nurs Pract. 2016; 3(4):1054-1057.
32. Livingston County Hoarding Task Force Guidelines for Addressing Hoarding Behavior. 2017:1- 14.
33. De Tommazzo VG. *São Paulo. Análise de denúncias de excesso de cães e gatos no munícipio de São Paulo de 2006 a 2015: Dissertação apresentada no programa de pós-graduação em Saúde Pública em ciências*; 2017.
34. Ferreira EA. *Acumuladores de animais: caracterização do perfil psicopatológico. Porto Alegre: Dissertação apresentada ao Programa de Pós-Graduação em Psicologia da Pontifícia Universidade Católica do Rio Grande do Sul*; 2016.
35. Hoarding Best Practice Committee/Massachusetts. Hoarding. Best Practices. Massachusetts. 2012:1-45.
36. National Health Service (NHS). Hoarding disorder – Guideline 2019-2021 [Internet]. 2011 [accessed 25 February 2019]:22. Available at: https://www.nhs.uk/conditions/hoarding-disorder/Guide.
37. *Ministério da Justiça (BR). Banco de ações judiciais (Jusbrasil) com termos de acumuladores compulsivos* [Internet]. 2018 [accessed 13 December 2018]. Available at: https://www.jusbrasil.com.br/topicos/26833900/acumulacao-compulsiva.
38. Australia. Government of South Australia. A Foot in the Door Stepping towards solutions to resolve incidents of severe domestic squalor in South Australia. A Guideline. 2013:1-62.
39. Tolin DF, Frost RO, Steketee G, Muroff J. Cognitive Behavirol Therapy for Hoarding Disorder: A Meta Analysis. Depression and Anxiety. 2015:1-9.
40. Cunha GR, Martins CM, Ceccon-Valente MF, Silva LL, Martins FD, Floeter D, Robertson JV, Ferreira F, Biondo AW. Frequency and spatial distribution of animal and object hoarder behavior in Curitiba, Paraná State, Brazil. *Cad. Saúde Pública*. 2017; 33(2):1-12.
41. Kyrios M, Mogan C, Moulding R. Frost OR, Yap K. Fassnacht DB. The cognitive-behavioural model of hoarding disorder: Evidence from clinical and non-clinical cohorts. Clin Psychol Psychother. 2017; 25(2):1–11.

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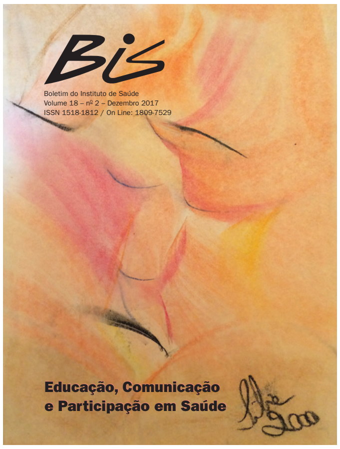

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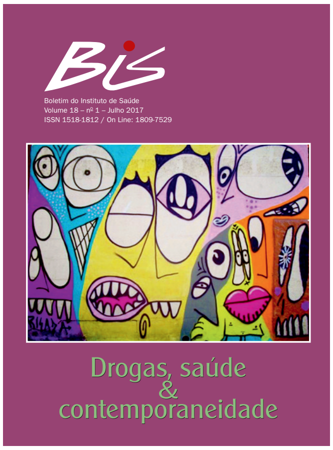

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