Chapter 19. Introducing meta-narrative reviews, critical interpretive synthesis and meta-aggregation

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Key points

- This chapter introduces three synthesis methods described and used by the wider systematic review community that may have potential value in Cochrane and Campbell reviews.
- Two review methods (meta-narrative and critical interpretive synthesis) can accommodate the synthesis of diverse types of quantitative, qualitative and mixed-methods evidence.
- The third synthesis method - meta-aggregation – has been developed specifically for synthesising qualitative studies addressing practice-based questions.
- Further methodological evaluation is needed to demonstrate the added value of these methods in a Cochrane and Campbell context.

19.1 Introduction

This chapter introduces three methods that may have potential value in a Cochrane and Campbell context but have yet to be fully evaluated to ascertain their potential contribution. Two review methods (meta-narrative and critical interpretive synthesis) involve the synthesis of diverse types of quantitative, qualitative and mixed-methods evidence. Meta-aggregation is a method specifically for the synthesis of qualitative studies addressing practice-based questions. Meta-aggregation has not been used in a Cochrane context as other methods of synthesis offer more opportunity for interpretation and transformation of data to make best use of the available qualitative evidence and develop new understandings of the phenomena of interest (see Chapters 8-12). In Campbell, meta-aggregation has been identified as a method that could potentially be used for highly structured practice-based questions that only require an aggregation of similar practice-based findings without further interpretation in order to make practice-based recommendations.

In the following sections each method is briefly described as follows: formulation of the review, identification of evidence, appraisal of evidence, synthesis of evidence, interpretation of the evidence, and reporting the review. The chapter concludes with sections on the role of stakeholder engagement and involvement, implications for equity diversity and inclusion, and reflexivity in relation to the aforementioned methods.

This chapter is important as the work of systematic review producers, such as Cochrane and Campbell, is evolving to meet the increasing needs of stakeholders and review funders who require different types of evidence to address different types of questions for specific needs, audiences and contexts. High volume producers of systematic reviews such as Cochrane and Campbell have traditionally used a limited set of well-developed and articulated methods and applied them rigorously to produce high quality reviews and trusted evidence. As the evidence needs of decision-makers evolve, there is likely scope for methodological innovation to determine the potential value of additional methods. For example, a pressing question that emerged from the COVID-19 pandemic was ‘why do

some research traditions explain poorer outcomes and higher COVID-19 mortality in Black and ethnic minority groups in terms of genetic predisposition and risky lifestyles while other traditions talk in terms of structural racism and inequitable access to healthcare?' A meta-narrative review is designed to explore, compare and contrast these (and other relevant) traditions. Greater understanding of these issues would help develop a better understanding of the varying effect of interventions in different groups of people, which could then be addressed through additional applied research and practice development.

In Cochrane, methodological innovation typically involves incorporating a new stream of synthesis, using a specific new method, alongside an existing method and evaluating the methodological contribution. This is commonly called a study within a review (SWAR) and is actively encouraged as a way of evaluating potential new methods alongside existing methods. For example, Harris et al (2019) incorporated a qualitative comparative analysis (QCA) (chapter 18) alongside an intervention effect review to explore the utility and value of the QCA method for a Cochrane context. Both Cochrane and Campbell have processes for the adoption and disinvestment in methods. Overall, the Editor in Chief needs to balance maintaining a core level of methods expertise without diluting either the need for methodological innovation or negatively impacting on the brand of trusted evidence. No systematic review producer can have expertise in all methods and some methods are best left to other producers to use.

19.2 Meta-narrative Reviews

A meta-narrative is the over-arching 'storyline' of research in a particular disciplinary tradition, including its philosophical assumptions, research questions and how these developed over time, preferred methods and quality criteria, and key findings. Meta-narrative reviews compare and contrast the storylines about similar phenomena of interest developed by different disciplines and traditions (such as medicine, nursing and patients or government, policy-implementers and citizens).

19.2.1 Formulation of review

Meta-narrative review seeks to unpack which research traditions have studied a particular topic and compare and contrast how they did so. It is best used to review a topic that has been researched from multiple different angles. Quality in meta-narrative review rests on six principles: pragmatism, pluralism, historicity, contestation, reflexivity and peer review.

Meta-narrative review is designed for topics that have been differently conceptualized and studied by different groups of researchers. The technique was originally developed to study the topic 'diffusion of innovations', which had been researched by multiple groups in multiple ways operating largely independently of each other (Greenhalgh et al., 2004).

The inspiration for the method was Kuhn's book *The Structure of Scientific Revolutions* (1970), which showed that science progresses in *paradigms* (that is, particular ways of viewing the world, including assumptions about how the world works) and that one scientific paradigm gives way to another as scientific progress renders yesterday's assumptions and practices obsolete. Newton's theories and methods, for example, became less and less able to answer the emerging questions of particle physics, leading Einstein to develop his theory of relativity.

Meta-narrative review looks historically at how particular research traditions have unfolded over time and shaped the kind of questions being asked and the methods used to answer them. It seeks to unpick the over-arching 'storylines' of research within which particular studies were planned, undertaken, published and critiqued (Greenhalgh et al., 2005).

Review authors should start by becoming familiar with the Realist and meta-narrative evidence synthesis: evolving standards (RAMESES) for meta-narrative reviews (Table 19.1) and guiding principles (Table 19.2); these are explained in detail in a separate paper (Wong et al., 2013).

Table 19.1: Publication standards for meta-narrative review

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Item	Standard
TITLE	
1	In the title, identify the document as a meta-narrative review or synthesis
ABSTRACT	
2	While acknowledging publication requirements and house style, abstracts should ideally contain brief details of: the study's background, review question or objectives; search strategy; methods of selection, appraisal, analysis and synthesis of sources; main results; and implications for practice.
INTRODUCTION	
3 Rationale for review	Explain why the review is needed and what it is likely to contribute to existing understanding of the topic area.
4 Objectives and focus of review	State the objective(s) of the review and/or the review question(s). Define and provide a rationale for the focus of the review.
METHODS	
5 Changes in the review process	Any changes made to the review process that was initially planned should be briefly described and justified.
6 Rationale for using meta-narrative review	Explain why meta-narrative review was considered the most appropriate method to use.

7 Evidence of adherence to guiding principles of meta-narrative review	Where appropriate show how each of the six guiding principles (pragmatism, pluralism, historicity, contestation, reflexivity and peer review) have been followed.
8 Scoping the literature	Describe and justify the initial process of exploratory scoping of literature.
9 Searching processes	While considering specific requirements of the journal or other publication outlet, state and provide a rationale for how the iterative searching was done. Provide details on all the sources accessed for information in the review. Where searching in electronic databases has taken place, the details should include (for example) name of database, search terms, dates of coverage and date last searched. If individuals familiar with the relevant literature and/or topic area were contacted, indicate how they were identified and selected.
10 Selection and appraisal of documents	Explain how judgements were made about including and excluding data from documents, and justify these.
11 Data extraction	Describe and explain which data or information were extracted from the included documents and justify this selection.
12 Analysis and synthesis processes	Describe the analysis and synthesis processes in detail. This section should include information on the constructs analysed and describe the analytic process.
RESULTS	
13 Document flow diagram	Provide details on the number of documents assessed for eligibility and included in the review with reasons for exclusion at

	each stage as well as an indication of their source of origin (for example, from searching databases, reference lists and so on). Consider using the example templates (which are likely to need modification to suit the data) that are provided.
14 Document characteristics	Provide information on the characteristics of the documents included in the review.
15 Main findings	Present the key findings with a specific focus on theory building and testing.
DISCUSSION	
16 Summary of findings	Summarise the main findings, taking into account the review's objective(s), research question(s), focus and intended audience(s).
17 Strengths, limitations and future research	Discuss both the strengths of the review and its limitations. These should include (but need not be restricted to) (a) consideration of all the steps in the review process and (b) comment on the overall strength of evidence supporting the explanatory insights which emerged. The limitations identified may point to areas where further work is needed.
18 Comparison with existing literature	Where applicable, compare and contrast the review's findings with the existing literature (for example, other reviews) on the same topic.
19 Conclusion and Recommendations	List the main implications of the findings and place these in the context of other relevant literature. If appropriate, offer recommendations for policy and practice.

20 Funding	Provide details of funding source (if any) for the review, the role played by the funder (if any) and any conflicts of interests of the reviewers.

Table 19.2: Guiding principles of meta-narrative review

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Principle	Explanation
Pragmatism	What to include is not self-evident. The reviewer must be guided by what will be most useful to the intended audience(s), for example, what is likely to promote sense making.
Pluralism	The topic should be illuminated from multiple angles and perspectives, using the established quality criteria appropriate to each tradition. For example, reviewers should avoid beginning with a single 'preferred' perspective or methodological hierarchy and proceed to judge work in other traditions using these external benchmarks. Research that lacks rigor must be rejected, but the grounds for rejection should be intrinsic to the relevant tradition, not imposed on it.
Historicity	Research traditions are often best described as they unfolded over time, highlighting significant individual scientists, events and discoveries which shaped the tradition.
Contestation	'Conflicting data' from different research traditions should be examined to generate higher-order insights (for example, about how different research teams framed the issue differently or made different assumptions about the nature of reality).
Reflexivity	Throughout the review, reviewers must continually reflect, individually and as a team, on the emerging findings.
Peer review	Emerging findings should be presented to an external audience—for example, the people who seek to use the review or whose care

	will be affected by its findings—and their feedback used to guide further reflection and analysis.

The most important step is making the commitment to studying the chosen topic through multiple different disciplinary lenses. For example: "*this* group of researchers studied topic X in *this* way, making *these* assumptions, whereas *that* group defined the topic rather differently and studied it in *that* way, making *these* very different assumptions".

Examples of complex review questions illuminated by a meta-narrative approach include:

- How have electronic patient records been researched in different disciplines, including medicine, computer science, sociology, and business and management (Greenhalgh et al., 2009)?;
- How has gratitude in health care been studied in different literatures, including healthcare (medicine, nursing), science (e.g. psychology), and the humanities (e.g. philosophy, anthropology) (Day et al., 2020);
- What can health policymakers learn about public accountability from the non-healthcare literature, including political science, organisational sociology, development studies, and ethics (Van Belle & Mayhew, 2016)?

In all these examples, the authors approached disciplines beyond their own with a very broad, open-ended question which began *"What can we learn from…?"*.

More specifically, a meta-narrative review asks some or all of the following questions:

1. Which research (or epistemic) traditions have considered this broad topic area?
2. How has each tradition conceptualised the topic—including researchers' assumptions about the nature of reality (ontology), preferred ways of knowing (epistemology) and study designs (methodology)?
3. What theoretical approaches and methods did the researchers use?

4. What were the main theoretical and empirical findings? For example, how many different meta-narratives were found, and what was the over-arching research 'storyline' in each?
5. What insights can be drawn by combining and comparing findings from different traditions? Where different research traditions came up with conflicting findings, to what extent can these be explained by differences in assumptions, definitions and ways of researching the topic?

19.2.2 Identification of evidence

The search phase begins with an initial scoping step to identify in broad terms the different research traditions, situated in different literatures, which have addressed the topic of interest. This is usually best achieved by two means: informal 'browsing' of the literature and consulting with topic experts and stakeholders.

Once a preliminary map of the territory has been sketched, searching should be guided by the objectives and focus of the review and revised iteratively in the light of emerging data. The most common place for review authors to go badly wrong in a meta-narrative review is to set out a detailed search string using MeSH terms and then stick rigidly to it, looking neither right nor left, and then exhaustively plough through a long, dull and more or less homogeneous set of hits. That kind of technocratic approach to searching is poor research practice in *any* review method. In meta-narrative review, the quirky and serendipitous discoveries may be the precious breadcrumbs which, if followed, could enable the review authors to discover a whole new meta-narrative which will complement the material they were expecting to find.

By definition, a meta-narrative review seeks to identify and combine different research traditions, hence different search strategies will need to be developed as appropriate to the different literatures. This stage is likely to involve searching for different kinds of data in different ways.

Search methods using forward and backward citation tracking may be particularly valuable in finding key documents. In particular, *seminal sources* (conceptual, theoretical or empirical studies which have defined the tradition and inspired later work) may be identified from judicious searching of the reference lists of later studies. Once identified, seminal sources should be citation-tracked to identify further sources which drew on these. If you are keen on electronic tools, bibliographic network analysis (van Eck & Waltman, 2017) can aid this tracking but is not a core component of the meta-narrative method.

The meta-narrative review authors do not approach the literature with a pre-defined 'preferred' study design. Rather, study design(s) should be identified from quality standards developed within a particular research tradition. Methodological filters should be used (if at all) only when these have been designated as a quality feature by the researchers *within* that tradition.

Searching is necessarily iterative, since the reviewer must move between the seminal source(s) and papers which subsequently cited that source, so as to build a picture of how research unfolded in each tradition.

When to stop searching? As with all interpretive methodologies, the answer to that question is not "when you have chased down every last paper" but "when you have identified enough evidence to achieve understanding".

19.2.3 Appraisal of evidence

Meta-narrative review is not a technical process, so following a set protocol will not guarantee that the review will be robust. Rather, it is an interpretive process oriented to making sense of the literature. This process requires *making judgements* about what drove the unfolding of research in particular traditions, and about the relevance and robustness of particular evidence within that tradition.

Meta-narrative review takes its quality criteria from the traditions included in the review, especially from seminal papers which others within that tradition have flagged as authoritative. For example, a review might include:

- a meta-narrative from clinical epidemiology in which randomized controlled trials are particularly valued;
- a meta-narrative from critical sociology in which theory-driven qualitative studies are particularly valued;
- a meta-narrative from the participatory co-design literature, in which methods to support creative solution-finding and include seldom-heard voices are particularly valued.

Studies in these separate traditions should be appraised using the quality criteria and tools that a competent reviewer or researcher in that tradition would choose to use. See also chapter 7 and relevant chapters on appraising evidence in Higgins et al 2019.

Each meta-narrative review is very different, and as the examples above illustrate, the precise data to be extracted from sources will vary according to the traditions identified. The goal of data extraction and subsequent interpretation and synthesis is to *construct a story* of how research on a topic unfolded over time in a particular tradition. The kind of data that could contribute to such a story includes (but is not limited to):

- earlier traditions from which this tradition emerged;
- background philosophical assumptions;
- key concepts and theories;
- research questions, how these were framed and why;
- preferred methodologies, study designs and quality criteria;
- key actors (for example, leading scientists or commentators) and events (for example, conferences) in the unfolding of the tradition;
- landmark empirical or theoretical studies;
- significant findings and how these shaped subsequent work; and
- key debates and areas of dispute within the tradition, including links with or breaches from other traditions.

19.2.4 Synthesis and interpretation of evidence

A meta-narrative review has two specific stages, which may overlap to some extent.

In the analysis stage, individual meta-narratives (that is, unfolding stories of research traditions over time) are mapped out, focusing particularly on the concepts, theories, methods and instruments which have characterized the tradition, major findings in that tradition and areas of dissent and disagreement. The last of these are particularly key. As Star (2002, p. 115) put it:

> "one might … define a knowledge discipline as a commitment to engage in disagreements. Biologists do not agree on the nature of species; sociologists bicker about the nature of society; literary critics diverge on notions of genre and style. What endures, however, are debates about the categories that constitute the core knowledge of the field."

The process of building this unfolding storyline is essentially interpretive and, hence, follows the principles of interpretivist analysis, including immersion in the data by repeated reading; reflexivity and discussion among researchers; consideration of how each new data item fits with an emerging picture of the whole; and checking where appropriate that the account is considered valid by experts within the designated research tradition. Both quantitative and qualitative traditions and data may need to be incorporated in the storyline. Reviewers should explain and justify any analytic methods used to combine and summarize data within a particular tradition.

In the second (synthesis) stage, review authors should compare and contrast the meta-narratives so as to highlight how the different groups have conceptualized the topic (including differences in philosophical position), how they have theorized it, and the methodological approaches and study designs used. Differences in findings between meta-narratives are higher-order data and should be analysed interpretively to produce

further insights (for example, about differences in underlying assumptions or methodological approaches).

19.2.5 Reporting the review

See Table 19 Publication standards for meta-narrative review (Wong et al 2013).
In meta-narrative review, the summary of main findings should be presented as a short description of each meta-narrative (including key qualitative and quantitative findings with it) followed by a synthesis in which the meta-narratives are compared and contrasted.

19.2.6 Examples of published meta-narrative reviews

Paparini et al (2021) used meta-narrative methods to review and synthesise the different disciplinary literatures on case study research and determine relevance to the study of contextual influences on complex interventions in health systems and public health research. They identified four broad partially overlapping research traditions that used case study in marginally different but overlapping ways with different goals: 1) developing and testing complex interventions in healthcare; 2) analysing change in organisations; 3) undertaking realist evaluations; 4) studying complex change naturalistically. Each tradition conceptualised context differently—respectively as the backdrop to, or factors impacting on, the intervention; sets of interacting conditions and relationships; circumstances triggering intervention mechanisms; and socially structured practices. Overall, these traditions drew on a small number of case study methodologists and disciplines. Few studies problematised the nature and boundaries of ‘the case’ and ‘context’ or considered the implications of such conceptualisations for methods and knowledge production.

Barry et al (2018) undertook a meta-narrative review to determine how socio-cultural influences and risk perception affected people’s behaviour (such as engagement in lifestyle interventions) after being told they had pre-diabetes. Three meta-narratives emerged. The first, which was labelled biomedical, characterised pre-diabetes as the first

stage in a recognised pathophysiological illness trajectory and sought to intervene with lifestyle changes to prevent its progression. The second, which was labelled psychological, focused on the theory-informed study of the knowledge, attitudes and behaviours in people with pre-diabetes. These studies found that participants generally had an accurate perception of their risk of developing diabetes, but this knowledge did not directly lead to behavioural change. Some psychological studies incorporated wider social factors in their theoretical models and sought to address these through action at the individual level. The third meta-narrative was labelled social realist. These studies conceptualised pre-diabetes as the product of social determinants of health and they applied sociological theories to explore the interplay between individual agency and societal influences, such as the socio-cultural context and material and economic circumstances. They recommended measures to address these structural influences on lifestyle choices.

19.3 Critical Interpretive Synthesis

The aim of a critical interpretive synthesis is to construct a critical analysis and synthesis of a complex body of literature. This synthesis method has its origins in health equity research and published examples have been increasing in number over the last few years.

19.3.1 Formulation of review

Critical interpretive synthesis is designed to incorporate diverse types of qualitative and quantitative primary research in order to develop new theory or further refine understanding of the phenomenon of interest. Similar to a meta-narrative review, it requires reviewers to take a critical orientation to the studies identified for the review which involves, for example, making explicit the traditions and their associated assumptions that studies are carried out within.

Methods for conducing critical interpretive synthesis are drawn from meta-ethnography (chapter 11) and the constant comparison method from grounded theory, that is also a feature of meta-ethnography and other methods that develop a line of argument synthesis. Like these methods, critical interpretive synthesis follows an inductive approach in order to create an overarching theory by synthesising theoretical categories extracted from the available qualitative and quantitative evidence on the topic or phenomenon of interest. An important feature of critical interpretive synthesis is that it draws on all forms of evidence in included studies and not just the 'themes' in included evidence. This is important because the development of third-order constructs (that is the new interpretations of review authors) cannot just sit on top of second-order constructs (findings reported in the primary study). The need for flexibility and iteration in critical interpretive synthesis is both a strength and a challenge. Review authors can frequently deviate too far from the central principles and core processes of critical interpretive synthesis (see box 19.2 for an overview) such that the end product no longer resembles the method as intended (Depraetere et al 2020). The degree of flexibility and iteration required when conducting a critical interpretive synthesis also makes it challenging for inexperienced review authors. Theoretical ideas and propositions are emergent throughout the review process as evidence is analysed, synthesised and interpreted. Emerging theoretical ideas and propositions subsequently guide iterative searching for evidence to substantiate or not emergent thinking and understanding about phenomena of interest and the interpretive narrative to explain them. The review team therefore needs to include review authors experienced in qualitative research paradigms, theory development and information science.

Although the popularity of critical interpretive synthesis has been increasing, there has not been much methodological development and testing reported in the literature. Dixon-Woods who pioneered critical interpretive synthesis illustrated this method of synthesis using the review in which it was developed in 2006 and this still serves as a useful and well thought through worked example. Dixon-Woods et al (2006) developed critical interpretive synthesis when trying to conduct a review of the literature on access to

healthcare by vulnerable groups in the UK. The reasons why this development was undertaken are summarised in Box 19.1

Box 19.1 Reasons why Dixon-Woods developed critical interpretive synthesis to synthesise the literature on access to healthcare by vulnerable groups in the UK.

- Literature on access to healthcare was large, diverse, and complex.
- Relevant sources of evidence included qualitative and quantitative methods; editorial comment and theoretical work; case studies; evaluative, epidemiological, trial, descriptive, sociological, psychological, management, and economics papers, as well as policy documents and political statements.
- The concept of 'access' had not been consistently defined or operationalised in the literature
- There were substantial adjunct literatures, including those on quality in healthcare, priority-setting, and patient satisfaction.
- A review of the area would be of most benefit if it were to produce a "mid-range" theoretical account of the evidence and existing theory that was neither so abstract that it lacked empirical applicability nor so specific that its explanatory scope was limited.

An overview of the principles underpinning critical interpretive synthesis and its key processes are outlined in Box 19.2. The next sections explore these principles and processes further and draws on the methodological critique conducted by Depraetere et al 2020.

19.3.2 Formulating the review question

Box 19.2. Principles and processes in critical interpretive synthesis from Dixon-Woods et al 2006. Permission granted Dixon-Woods.

- A review question should be formulated at the outset, but should remain open to modification. Precise definitions of many constructs may be deferred until late in the review and may be a product of the review itself.
- Searching, sampling, critique and analysis proceed hand in hand, and should be seen as dynamic and mutually informative processes.
- Searching initially should use a broadly defined strategy, including purposive selection of material likely or known to be relevant.
- The analysis should be aimed towards the development of a synthesising argument: a critically informed integration of evidence from across the studies in the review. The synthesising argument takes the form of a coherent theoretical framework comprising a network of constructs and the relationships between them. The synthesising argument links synthetic constructs (new constructs generated through synthesis) and existing constructs in the literature.
- There is a need for constant reflexivity to inform the emerging theoretical notions, as these guide the other processes.
- Ongoing selection of potentially relevant literature should be informed by the emerging theoretical framework. Literatures not directly or obviously relevant to the question under review may be accessed as part of this process.
- An ongoing critical orientation to the material to be included in the review is encouraged. Some limited formal appraisal of methodological quality of individual papers is likely to be appropriate. Generally the aim will be to maximise relevance and theoretical contribution of the included papers.
- Formal data extraction procedures may be helpful, particularly at the outset of the review, but are unlikely to be an essential feature of the approach.
- The aim of critical interpretive synthesis is not to offer a series of pre-specified procedures for the conduct of the review. The method explicitly acknowledges the "authorial voice"; that some aspects of its production of the account of the evidence will not be visible or auditable; and that its account may not be strictly reproducible. Its aim is to offer a theoretically sound and useful account that is demonstrably grounded in the evidence.
- Constant reflexivity on the part of authors of reviews is demanded. Authors are charged with making conscientious and thorough searches, with making fair and appropriate selections of materials, with seeking disconfirming evidence and other challenges to the emergent theory, and with ensuring that the theory they generate is, while critically informed, plausible given the available evidence.

Critical interpretive synthesis involves the development of an open review question (also called a compass question), which is further refined as the review develops and may not be finalised until the end of the review. In their review focused on access to health care for vulnerable groups Dixon-Woods et al (2006) found that it was not desirable or possible to specify a precise review question, *a priori* definitions, or categories to group and summarise extracted data in advance because access to health care is defined and operationalised in many diverse ways across different literatures and the review team wanted to capture and describe this diversity as it emerged from the ongoing analysis of the literature. Their initial very broad fuzzy and tentative review question served as a compass and included a focus on equity and on how access, particularly for vulnerable groups of people, could best be understood in the UK National Health System, that is free at the point of delivery. Thereafter, their approach to further specifying the review question was highly iterative, and they modified the question in response to search results and findings until the final review questions became apparent towards the end of the review. The final review questions were 1. How does help-seeking behaviour affect access to health care? 2. How does provision of services affect access to health care? 3. What organisational features of health services affect access to health care? 4. How can access to health care be improved? (Dixon-Woods et al 2005).

19.3.3. Identification of evidence

Similar to searching for evidence for meta-narrative reviews, critical interpretive synthesis is a complex review methodology resulting in relatively complex processes that review authors have to operationalise, including the need for review-specific and bespoke searches for relevant evidence. Chapter 5 includes guidance on the purpose of searching for qualitative studies and how to search for evidence. Depraetere et al 2020 suggest that the search for relevant literature should involve conventional searches on databases as well as searches of websites, backwards and forwards reference chaining, and contacting experts. Dixon-Woods et al (2006) did not however find that a traditional structured search of bibliographic databases very helpful as it yielded too many potential hits but missed many key papers which provided deep insights into how vulnerable people accessed

health services. Instead, they relied on what they thought of as a more organic process that combined a number of strategies, including searching of bibliographic databases; searching websites; reference chaining; and contacts with experts. This approach needed further refining as the organic search identified a vast literature that was beyond the capacity of the review authors to manage. To progress further and get round the challenge of too much literature, Dixon-Woods et al (2006) redefined the aim of the searching phase. They no longer aimed to undertake a comprehensive search to include all relevant literature and redefined the aim of the searching phase to identifying potentially relevant papers to create a purposive sampling frame, which subsequently included around 1,200 records (see also chapter 6 on sampling). They purposively selected papers that were '*clearly concerned with aspects of access to healthcare and later used theoretical sampling to add, test and elaborate the emerging analysis*'. The entire sampling process required a constant process of examining and discussing opposing ideas in order to create an understanding that was conducted concurrently with theory generation.

Depraetere et al 2020 subsequently described the process of identifying evidence for a critical interpretive synthesis. They suggest that review teams need to select literature for inclusion informed by the emerging conceptual framework and based on the principles of theoretical saturation and likely relevance. This typically includes the use of purposive selection with flexible inclusion criteria to create a manageable sample to analyse and synthesise.

19.3.4 Appraisal of evidence

The appraisal of evidence follows similar principles as meta-narrative reviews in that the assessment is focussed on the content of the paper, its likely relevance, and theoretical contribution to the critical interpretive synthesis rather than on methodological limitations (Depraetere et al 2020). Using the criteria in Box 3, studies which contain significant methodological limitations such that they might be considered 'fatally flawed' can be excluded from the synthesis. Each review team would need to clarify and make transparent what they considered to be 'fatal flaws' in primary qualitative studies for their

specific QES. See also chapter 7 for more information on assessing the methodological strengths and limitations of studies. Methodologically weak research that is not considered to be fatally flawed may still provide relevant insights and help develop the theoretical framework.

Dixon-Woods et al (2006) applied a low-quality threshold by only excluding fatally flawed studies to maximise the inclusion and contribution of a wide variety of papers at the level of *concepts*. They used five questions to assess methodological limitations (Box 19.3) which generally map onto the recommendations in Chapter 7. Following principles of meta-ethnography – the exclusion of studies based on methodological limitations was deferred until the synthesis phase since papers considered to be methodologically weak could still provide relevant insights regarding the emerging theoretical framework. During the synthesis phase, critical judgements and interpretations were made of a study's credibility and contribution. Those studies that did not contribute much data and were methodological weak were excluded from the sample at this stage.

Box 19.3. Five questions to assess methodological limitations (Dixon-Woods at al 2006).
Permission to use granted by Dixon-Woods.

1) Are the aims and objectives of the research clearly stated?
2) Is the research design clearly specified and appropriate for the aims and objectives of the research?
3) Do the researchers provide a clear account of the process by which their findings were produced?
4) Do the researchers display enough data to support their interpretations and conclusions?
5) Is the method of analysis appropriate and adequately explicated?

19.3.5 Synthesis and interpretation of evidence

Depraetere et al 2020 suggest that synthesis in critical interpretive synthesis, including construction of a coherent theoretical framework and formulation of a synthesising argument, follows some, but not all, of the analytical processes used in meta-ethnography (Chapter 11). Specifically, it is considered problematic to undertake an initial reciprocal translation with the large numbers of studies typically included so in a critical interpretive synthesis the interpretation of evidence tends to follow a line of argument synthesis with a synthesising argument produced as the final output. A refutational synthesis is also conceptualised more as a reflexive and critical approach to the literature in a critical interpretive synthesis.

Dixon-Woods et al (2006) included 119 papers in their critical interpretive synthesis on access to healthcare amongst vulnerable groups and developed a data extraction proforma similar to those described in chapter 8. However, they found this overly time consuming so coded larger documents with highlighter pens. They analysed extracted data and generated a synthesising argument by integrating evidence across studies into a coherent theoretical framework that depicted a network of synthesising constructs and the relationships between them. Dixon-Woods et al suggest that a synthesising construct is equivalent to a third order construct in meta ethnography (see chapter 11 for definitions of second and third order constructs) and that a synthesising argument can be made up of a combination of second and third order constructs. Their analysis process to produce a synthesising argument was similar to the analysis process in primary qualitative research. They familiarised themselves with included papers, identified recurring themes and developed a critique. They then developed themes that helped explain the phenomena described in the literature and constantly compared the developing theoretical structures against data in the papers, and specified categories of data and the relationships between categories. Software for qualitative analysis was also used to help identify patterns, themes and categories whilst at the same time taking a critical, dynamic, reflexive and transparent approach with regular team meetings and discussions.

When undertaking a refutational synthesis, Dixon-Woods et al incorporated relevant principles from the process of undertaking the line of argument synthesis to produce a synthesising argument. They went beyond appraisal of the included evidence and reflected on the credibility of the evidence-base as well as its quality and theoretical foundation. Similar to meta-narrative review, this critique involves identification of research traditions and meta-narratives that guided their particular field of research. This critique of the literature also informed sampling and selection of evidence in iterative cycles and played a key role in theory generation and seeking alternative explanations.

19.3.6 Reporting the review

There are currently no reporting guidelines for reporting a critical interpretive synthesis. Review authors should follow the guidance on selecting relevant reporting guidelines in chapter 20. Given the similarities of critical interpretive synthesis with aspects of meta-narrative reviews and meta-ethnography, review authors could also draw on the reporting tools for these methods to develop a composite reporting guideline.

19.3.7 Published example of critical interpretive synthesis

Flemming (2010) undertook a critical interpretive synthesis to synthesize quantitative research, in the form of an effectiveness review and a guideline, with qualitative research to examine the use of morphine to treat cancer-related pain. The findings of the effectiveness review were used as a framework to guide the translation of findings from qualitative research using an integrative grid (see also chapter 14). A secondary translation of findings from the qualitative research, not specifically mapped to the effectiveness literature, was guided by the framework. Nineteen qualitative papers were synthesized with the quantitative effectiveness literature, producing 14 synthetic constructs. These were developed into four synthesizing arguments which drew on patients', carers' and healthcare professionals' interpretations of the meaning and context of the use of morphine to treat cancer pain:

• Opioids and opioid concern

• Using opioids is a balancing act and a trade off

• The existential meaning of cancer and cancer pain

• The inter-subjectivity of pain

A mediating factor that was integral to all of the synthesizing arguments was the synthetic construct of 'control' and so this was presented as an over-arching part of the synthesis.

19.4 Meta-aggregation

Meta-aggregation is a highly structured method for aggregating findings from primary qualitative studies developed by the Joanna Briggs Institute, now known as JBI. Meta-aggregation was primarily developed to produce practice-based recommendations from a body of qualitative research. Detailed guidance is available on conducting meta-aggregation (Lockwood et al (2024) and the JBI website has additional resources, software and bespoke tools to conduct the review.

19.4.1 Formulation of the review

Meta-aggregation is viewed as being underpinned by pragmatism whereby its aim is to produce action orientated statements based on aggregated evidence from primary studies for use in practice or policy (Lockwood et al 2024). The aggregation and reporting of synthesised findings stay close to the findings reported in the primary studies with minimal (if any) re-interpretation or re-conceptualisation.

Types of questions addressed by meta-aggregation are simple rather than complex, and are geared towards exploring experiences and perceptions linked to health and care practice:

- What are the experiences of older women living with osteoporosis?
- What are the experiences of health professionals who have experienced grief as a result of a paediatric patient dying?
- What are the experiences and perceptions of adults with chronic non cancer pain of participating in a peer social support intervention?

Questions can be developed using the frameworks described in Chapter 2. One that is most commonly used for meta-aggregation is Population, Phenomena of Interest, Context (PICo), developed by JBI (Lockwood et al., 2024).

19.4.2 Identification of evidence

JBI argue for the adoption of an inclusive approach to searching and selecting studies, to ensure that all possible studies are included (Lockwood et al., 2024). Searches should be comprehensive, aiming to find most or all available studies. See Chapter 5 on comprehensive searches for identifying qualitative research studies.

19.4.3 Appraisal of evidence

All studies that meet the inclusion criteria specified in the protocol go forward for aggregation. The search for evidence is therefore comprehensive without the need to sample studies for inclusion. It is however important to identify methodological limitations in primary studies. Most QESs using meta-aggregation have used the standard JBI checklist for appraisal of qualitative evidence. The assessment is then mapped against the pre-determined cut off for methodological quality detailed in the a-priori protocol. Studies that meet the minimum methodological quality criteria are included in the aggregation.

In a Cochrane context, review authors are encouraged to use the CAMELOT tool to assess methodological limitations (chapter 7) in combination with GRADE-CERQual (chapter 13).

There is a subsequent appraisal of confidence in each extracted finding from individual studies to judge the extent to which the finding is supported by the direct quotes reporting the participants' voices. The JBI approach has three options: Unequivocal, credible and unsupported findings. Any findings that are determined to be not supported by data in the form of direct quotes are excluded from the aggregation. This means that if

none of the findings from a study are supported with direct participant quotes then the study is excluded from the aggregation, but not supported findings should still be extracted and presented separately. If some of the findings are not supported with direct participant quotes but others are, then only the unequivocal and credible findings stay in the aggregation.

JBI use the CONQual 'Summary of Findings' to rank Dependability and Credibility. In QESs conducted in a Cochrane and Campbell context, review authors should instead use GRADE-CERQual (Chapter 13) to assess the confidence in synthesised qualitative findings as Grade-CERQual alongside GRADE is already commonly built into existing guideline and decision-making processes.

19.4.4 Synthesis and interpretation of evidence

The process of aggregation involves minimal further interpretation of extracted evidence and has three steps:

1. Extraction of contextual details and findings from studies using standardised JBI tools
2. Grouping similar findings to form categories
3. Aggregation of categories to create synthesized findings.

Step 1 involves standard processes including the capturing contextual information about each of the included studies such as author details, year of publication, countries involved, and participant details. The extraction of verbatim findings from included studies is undertaken using a standardised or bespoke data collection form and then the extracted findings are listed along with the best excerpts of raw data such as participant quotes that have been reported by authors. The level of credibility is established for each finding at this stage (Figure 2).

Study: Almeida *et al.* 2016[38]	
Finding 1	It is a difficult time for the nurse (U)
Illustration	There are situations that are a little bit more difficult... such as these situations of death. It is a very difficult moment for the whole team, not just to nurses... Also because not everyone has this serenity... We were trained to take care of alive people, not dead people, right. And even more at this age... if they were dying elderly, you accept. I know this is wrong, but I accept that the elderly has already lived, had his life, was happy, got married, had a son. Now, a baby had nothing, okay ... nothing, simply nothing, okay! He did not even have time to have... p.120 We end up getting involved, creating a very great bond: family, baby and staff (...) So for us, nurses, who're closest to the family, it is clear that we suffer more than the doctor who, of course, suffers, but it's a different involvement. We're there six hours, every day, until he/she (the baby) dies, right... p.120

Figure 2 Table extract from a meta-aggregation of healthcare professionals' experiences of paediatric death by Barnes et al (2020) that shows the extraction of a finding with supporting participant quotes from a primary qualitative study that are judged to be unequivocal (U). Permissions needed. Original image required.

Step 2 involves grouping the extracted findings from step 1, based on similarity of meaning. Common findings are put together (aggregated) and then a category created that captures the meaning of these aggregated findings.

Step 3 takes the aggregation one stage further to create a synthesised finding (Figure 3).

Findings	Categories	Synthesized findings
Experiencing a feeling of accomplishment, despite the sadness for the loss of the newborn (C) Relief (C) Gaining perspective on life (U) Providing better care (U) Relief (U) Growing personally (C) Laughter through tears (U)	Positive outcomes	Alleviating grief: Health professionals identified factors that contribute toward improving or alleviating the experience of grief. In spite of the difficult experience, support from friends, family or colleagues; focusing on the positives; and allowing time and experience to assist with coping led to improvements in the experience of grief.
Collegial camaraderie and support (C) Personal support (C) Reconfirming (C) Talking/sharing (U) Reaching out (U) Sympathy (U) Support from colleagues (C) Emotional support could impact the quality of the nursing care (U)	Support from people	
Desensitization (U) The impact of the sudden death of the child/adolescent (C) Time (C)	The importance of time and experience	

Figure 3 Extract from a meta-aggregation of healthcare professionals experiences of paediatric death by Barnes et al (2020) that shows how the findings have been linked to

categories and synthesized findings. C= credible finding. U = unequivocal finding. Permissions needed. Original image required.

19.4.5 Reporting the review

Review teams should follow the guidance for reporting QESs in Chapter 20. JBI guidance outlines that findings from the meta-aggregation should be reported as indicatory statements, which form the basis of recommendations for policy and practice (Lockwood et al 2024). Review authors should also report what data were considered findings, how findings were grouped, and how categories were aggregated and synthesised findings created and finalised.

19.4.6 Examples of meta-aggregation

The worked example shown in this chapter is derived from a meta-aggregation of healthcare professionals' experiences of paediatric death (Barnes et al 2020). The meta-aggregation produced three findings from 12 studies that mostly included nurses working in a hospital with sample sizes ranging from six to 25 participants. The findings were related to mostly nurses experiences of 1) physical, behavioural, psychological or spiritual symptoms; 2) compounding grief; and 3) alleviating grief. Concerns about methodological quality led to synthesized findings receiving a ConQual rating of low or moderate.

The review authors concluded that findings from the meta-aggregation and the included studies highlighted the relevance of the grief experiences by health professionals when paediatric patients die. Based on the quality of evidence they made 2 conditional rather than strong recommendations for practice, including: 1) that health professionals maintain awareness of their own response to paediatric death as well as the potential response of their colleagues, and 2) that employers implement strategies and policies to support staff who are experiencing grief associated with their work environment.

Additional examples can be found in the JBI Library (https://jbi.global/).

19.5 Stakeholder involvement and engagement

The principle of stakeholder engagement and involvement is key to all the methods described in this chapter. Patient and user input is important as it helps ground researchers in the things that matter to those whose care the research is intended to inform. Meta-narrative review in particular offers a methodology for getting to first base when addressing the question "how have groups *beyond the main orthodox tradition* conceptualized and studied this topic?", and also for asking the crucial higher-order question "*why* are there such marked differences between the orthodox and heterodox traditions?". Representation from these groups as stakeholders can help with developing and interpreting the story from the perspective of different groups. The inherently pragmatist and pluralist nature of meta-narrative review and CIS can help identify, and promote valuing of, research traditions which study patients' experiences and concerns from different perspectives. Given the similarities in some data processing between critical interpretive synthesis and meta-ethnography, see also chapter 11 for involving stakeholders. All the stages of meta-aggregation can likewise be further enhanced and grounded in practice by early and continuing engagement and involvement of relevant stakeholders and people expert by experience.

19.6 Equity, diversity and inclusion

Irrespective of the method, review authors should consider equity, diversity and inclusion in all aspects of planning and conducting their review.

Meta-narrative review and critical interpretive synthesis are inherently pragmatic (i.e. use-oriented), non-hierarchical and pluralist, so in theory at least they may offer greater potential for understanding inequities and solutions for reducing inequities. Meta-narrative review in particular does not seek to produce a 'grand mean' where different perspectives are in some way averaged out or squeezed into an awkward consensus. Rather, the differences become data to be explained and even harnessed. See also chapter 11 for addressing equity, diversity and inclusion in meta-ethnography, which involves similar considerations to critical interpretive synthesis.

As a general observation, traditional forms of systematic review have been criticised for being overly focused on English-language papers describing 'ivory-tower' research produced in the Global North, based on problems, ideas and methodologies that emerged in those regions and on topics that were prioritized for funding in those regions. There are, for example, many more systematic reviews of Western medicines than reviews of non-pharmaceutical interventions or non-Western remedies. Equity, diversity and inclusion considerations will also be vital in practice-based reviews using meta-aggregation, whereby practices may be influenced by different social contexts, the belief systems of individuals and cultures, and the health, social care and education systems included in the review.

19.7 Reflexivity

The composition, disciplines, positionality and inherent biases of the review authors and stakeholders involved in conducting the review can potentially impact on choices and decisions at all stages of a review and should be made transparent.

Meta-narrative review and critical interpretive synthesis in particular demand constant reflexivity with an ongoing critical orientation to the material by placing the literature within its context.

For example, Dixon-Woods et al (2006) found that in the process of refining the critical interpretive synthesis question, they benefited from the multidisciplinary nature of the review team: this allowed a range of perspectives to be incorporated into the process, something that was also helpful and important in other elements of the review. Crucially, they also used expertise within the team to identify relevant literature from adjacent fields not immediately or obviously relevant to the question of "access". Similarly for meta-aggregative reviews that explore practice-based questions affecting people in a social context where there are inherent power imbalances between actors. These power imbalances can be further amplified if the review team largely represent a professional rather than a lay perspective and these power imbalances and their potential impact on

all review processes and decisions are not explored amongst the team and made transparent.

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19.9 Declarations of Interest

Flemming, Noyes and Harden are convenors of the Cochrane Qualitative and Implementation Methods Group. Noyes is a member of the Cochrane Methods Executive and Editorial Board. Greenhalgh is an originator of meta-narrative reviews. Flemming is the author of a cited review.

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