

Innovation in systematic review methods: successive developments in framework synthesis

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Thesis presented for the award of a Doctorate in Philosophy

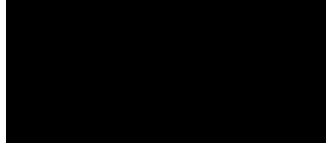
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DECLARATION PAGE

I, Virginia Brunton, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signature:



Date: 10 November 2017

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I would like to acknowledge the solid support and thoughtful guidance of my two PhD supervisors, Professor Sandy Oliver and Professor James Thomas. As always, you both provide the most inspiring examples of how to be an excellent academic.

And of course, special thanks go to my family, Jeff and Chloé Brunton, for their help, flexibility, unique insights and good humour as I worked my way through this thesis. Thanks for showing me daily that life is what we make it.

SUPPORTING STATEMENTS

Chapter 3: Brunton G, Oliver S, Oliver K, Lorenc T (2006) *A synthesis of research addressing children's, young people's and parents' views of walking and cycling for transport*. London: EPPI-Centre, Social Science Research Unit.

During this review, Ginny took part in all stages of the research process, which was led by Prof. Sandy Oliver. Ginny's role included: contributing to research question formation and protocol development; designing and leading on searches for and inclusion screening of relevant citations; retrieval, coding and analysis of studies; contributing to thematic synthesis; and conducting cross-study synthesis comparing findings from qualitative framework synthesis with findings from meta-analysis of interventions. Ginny facilitated communication with stakeholders and through writing during the review: for example, by: identifying and liaising with advisory group members; organising stakeholder consultation meetings and agenda and presenting findings for discussion; and in leading on writing draft reports of the research project required by funders. Ginny led on day-to-day management of the research process and project team members.

Chapter 4: O'Mara-Eves A, Brunton G, McDaid D, Oliver S, Kavanagh J, Jamal F, Matosevic T, Harden A, Thomas J (2013) *Community engagement to reduce inequalities in health: a systematic review, meta-analysis and economic analysis*. *Public Health Research*. 1(4): 1-548.

This was a large project involving four distinct but related pieces of work, each of which was led by one or two researchers. Ginny was involved in all stages of the project but specifically co-led (with James Thomas) two of the sub-projects: the synthesis of theoretical papers and the synthesis of process evaluation papers. Prof. James Thomas was primary investigator on the project. Alison O'Mara-Eves led on the synthesis of intervention evaluations and much of the overall day-to-day management of the project, to which Ginny contributed extensively.

Most relevant in the context of this thesis, Ginny conducted coding and analysis of studies, including descriptive analysis of studies included in meta-analysis, and was responsible for

integrating findings from research and stakeholder consultations into subsequent iterations of the conceptual framework. Ginny also contributed to research question formation and protocol development, and co-designed search, retrieval, and inclusion screening of relevant citations. Ginny co-led with Alison O'Mara-Eves on stakeholder consultations during the review, which involved identifying and liaising with diverse stakeholder groups during independent stakeholder consultations and advisory group meetings, organising agendas, and leading discussions. She led the write-up of the process evaluation chapter and co-wrote the theoretical synthesis chapter of the final report to the funder.

Chapter 5: Brunton G, Caird J, Stokes G, Stansfield C, Kneale D, Richardson M, Thomas J (2015) *Community engagement for health via coalitions, collaborations and partnerships: a systematic review*. London: EPPI-Centre, Social Science Research Unit.

Ginny led on this project as a primary investigator, submitting a successful research proposal, developing research questions and a research protocol, managed the review process and multiple team members through all stages of the review. Ginny led on the framework synthesis of processes and levels of community engagement, and the integration of these findings with those from meta-analysis and QCA, presenting findings at three NICE public health guidelines committee meetings. Ginny led on writing the final report to funders, coordinating team contributions and liaising with NICE policy team members.

Chapter 6: Brunton G, Caird J, Sutcliffe K, Rees R, Stokes G, Stansfield C, Llewellyn A, Simmonds M, Thomas J (2014) *Depression, anxiety, pain and quality of life in people living with chronic hepatitis C: a systematic review and meta-analysis*. London: EPPI-Centre, Social Science Research Unit.

As lead on this project, Ginny developed research questions and a research protocol, managing the daily review process and multiple team members through all stages of the review. Ginny led on development and refinement of the framework synthesis comparing research on extrahepatic conditions with information gained from stakeholders. Ginny led on stakeholder consultation, identifying and liaising with stakeholders, coordinating

meetings where she presented findings, led consultations and followed up with stakeholders. Ginny led on writing the final report to funders, coordinating team contributions and liaising with stakeholder groups.

As co-authors on these reports, we confirm that this information is correct and represents an original contribution to these research projects.

Alison O'Mara-Eves

Date: 26 April 2016

James Thomas

Date: 26 April 2016

Sandy Oliver

Date: 26 April 2016

ABSTRACT

Background: Systematic reviews are used increasingly to synthesise research for policy and practice decisions. Systematic reviews use transparent, explicit and consistent methods to identify, appraise and integrate research evidence, to inform existing assumptions. Traditionally used to examine the effects of health care, systematic review methods have evolved to address more complex issues across health and social policy. Framework synthesis is one systematic review method employed to address such complexity. Adapted from framework analysis used in primary research, framework synthesis begins with an *a priori* conceptual framework, which develops iteratively as new data are incorporated and themes derived from the data. Framework synthesis has been used increasingly in qualitative and mixed-method systematic reviews, sometimes incorporating stakeholder consultation to describe and interpret review findings.

Aims: This thesis will demonstrate how my development of framework synthesis methods is situated within, and contributes to, wider debates about research synthesis methods in systematic reviews.

Methods: I conducted an overview of systematic reviews. A systematic review of the literature discussing or employing framework synthesis methods was conducted. Included studies were ordered and synthesised using framework synthesis methods. The resulting conceptual framework structured the assessment of each of the submitted publications (the 'thesis reviews'). Findings were subsequently incorporated into the conceptual framework and higher order themes derived using constant comparative analysis.

Results: This thesis distinguishes two key approaches to framework synthesis: one approach that constructs a framework (often in discussion with stakeholders) to accommodate research from across academic disciplines and/or policy sectors; and the 'best-fit' approach that borrows a framework from a related area to initiate synthesis within a narrower disciplinary or policy scope. Its utility is demonstrated in handling mixed methods and mixed sources reviews, using diverse data types and synthesis methods in order to generate, explore and/or test theory in collaboration with stakeholders. I conclude that framework synthesis is a very flexible research synthesis method that can meet the complex conditions and epistemology arising from public health policy issues.

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CHAPTER 1. Thesis rationale and methods

1.1 Background

Systematic reviews are widely used to synthesise research in order to inform policy and practice decisions (1-3). A systematic review is a ‘review of research literature that uses systematic and accountable methods’ (4):p4). Systematic reviews enable us to ‘take stock’ of the research in a field of inquiry by seeking out, selecting, critically assessing and synthesising the available research. They do so using transparent, rigorous and replicable methods. By identifying the most relevant research, it becomes possible to generalise research results to a wider population by looking for knowledge and findings across individual primary studies (3). Systematic reviews build on ‘previous investments in research’ (5):8) and can ‘recast our view of research by challenging existing assumptions and suggesting new areas for investigation’ (4):p4).

Systematic review methods have addressed questions about the effects of health care since the mid-1980s, where the findings from evaluations of relatively simple interventions were aggregated, often statistically (4). Since the 1990s, systematic reviews have been used increasingly to address more complex policy questions (2, 6). The literature discussing complexity in intervention research is vast (7-10). For the purposes of this thesis, ‘complexity’ refers to ‘interventions with outcomes that are likely to be contingent on variant characteristics of the intervention itself, the systems in which the intervention is implemented, and/or interactions between properties of the intervention and systems’ (11):p.1264). This recognition of complexity led to an evolution of research synthesis methods designed to address research questions examining issues which could both precede or go beyond effectiveness (12). The research team based at the Evidence for Policy and Practice Information and Coordinating (EPPI-) Centre, University College London (UCL) has conducted systematic reviews of complex issues for policy decision making since the 1990s. Examples include syntheses of research on people’s understandings of a particular phenomenon (13), and those to develop theories explaining why people participate differently in health activities (5). My own work within the team has addressed questions exploring barriers and facilitators to positive health behaviours (14, 15); and understanding the association between chronic diseases and their importance to those who are affected by the diseases (16). Such questions require different approaches to synthesise review findings appropriately (17, 18).

Framework synthesis is one method employed to address such complex issues examined in systematic reviews. Framework analysis originated as a method of analysing primary research data to address policy concerns (19) and its adaption has more recently been described as one of an array of methods for use in systematic reviews of research literature (12). Here, the research question and the background theoretical and empirical literature shape an understanding of the issue under study into an *a priori* conceptual framework, which develops iteratively as new data are incorporated and themes are derived from the data. Framework analysis presents an opportunity to use a 'scaffold against which findings from the different components of an assessment may be brought together and organised' (20):p.29). Its flexibility captures new understanding as data is incorporated into the framework. Used in the context of synthesizing the findings of prior research within a systematic review, it is described as '*framework synthesis*' (12, 20).

Framework synthesis has been used in different types of systematic reviews, ranging from those that examine qualitative studies (21) to those undertaking mixed methods syntheses (15, 22). Its utility at different stages of a review has also been demonstrated in the use of and reporting on stakeholder consultation, where researchers are often challenged to communicate clearly with stakeholders, integrate the consultation findings with those of the review, and then describe their methods of consultation (23). By incorporating stakeholders' perspectives, consultation throughout the review process can both widen the review's scope and make it a more relevant and useful product (22).

Ontology, epistemology, methodology and method

Research syntheses are undertaken within a particular (and sometimes explicit) worldview, which requires some examination of their stated or apparent epistemology (18, 24). This requires some understanding of the philosophy of social science; however, it is challenging to find commonality around the characteristics of different traditions, as authors differ in their definitions of concepts (25). Four key philosophical underpinnings of the two predominant traditions or 'paradigms' (26) are summarised in Table 1.1.

Table 1.1 Ontology, epistemology, methodology and methods

Task	Paradigms	
What do we know? <i>Ontology</i>	Objective <i>An independent external reality exists to be known, separate from ourselves</i>	Subjective <i>No independent reality out there apart from our own experiences</i>
How do we know what we know? <i>Epistemology</i>	Positivist <i>Reality can be accurately measured; facts are observable</i>	Interpretivist/Constructivist <i>Reality is socially constructed; only individual meanings and actions can be understood</i>
How do we find out? <i>Methodology</i>	Deduction <i>Apply data to test a previously existing theory ('a priori')</i> <i>Little iteration</i>	Induction <i>Use data to build a theory ('emergent')</i> <i>Highly iterative</i>
What procedures do we use? <i>Methods</i>	Quantitative Surveys Questionnaires Random sampling	Qualitative Interviews Focus groups Analytical approaches

(Adapted from Morgan 2007, Oakley 2000; Ritchie et al. 2014)

Ontology is the study of the nature of the world; it is a philosophical concept concerned with understanding our existence in the world around us (i.e. 'what is the *meaning* of life?') (27). Two schools of thought (or paradigms) compete in studying of the nature of existence: objectivism, which asserts that there is an external reality that exists independently of our understanding of it and can be known, i.e. reality exists; and subjectivism, which claims that no external reality exists beyond our own beliefs and understandings, and in essence we construct reality either individually or collectively (25, 28).

Epistemology is the study of how knowledge is acquired, or 'ways of knowing' (25). Two main schools of thought have governed the epistemology of social research in the late 20th and early 21st century: positivism, which holds that research can be undertaken in an objective, value-free manner because human behaviour is governed by 'law-like regularities' (27); and interpretivism/constructivism, which asserts that human behaviour must be comprehended by 'understanding people's perspectives within the context of their own lives' in order to determine the meaning, rather than the causes, of human

behaviour (27). This way of knowing asserts that research is completely and necessarily value-laden.

The **methodological** approaches to examining observations about the world, or 'data', have been described as inductive and deductive. Inductive approaches tend to 'build up' theory by looking for patterns in the data, whereas deductive methods are more likely to use data to confirm or reject an already existing hypothesis (27). To some degree this is related to the extent of iteration – the interplay a researcher conducts between the data and developing higher order concepts. More iteration suggests more inductive and thus a more interpretivist stance. In contrast, less iteration suggests a more deductive approach in which theory is more established (18, 24).

Finally, the **methods** utilised to gather observations vary between paradigms: quantitative (e.g. surveys, questionnaires and random sampling) and qualitative (e.g. interviews, focus groups).

Notions of theory, conceptual frameworks, theories of change and logic models

Definitions of theories, conceptual frameworks, mechanisms, theories of change, and logic models abound and are often used interchangeably. Most simply, a **theory** is a statement of ideas that together explain a phenomenon (29). **Grand theory** which describes overarching ideas about wide-ranging social issues (e.g. Marxism or feminism) is distinguished from **low-level theory** which describe aspects of a phenomenon without demonstrating their relationship (30). A **conceptual framework**, similar to theory, can be considered a working hypothesis of key concepts, constructs and the potential interactions between them (31). **Mechanisms** or **theories of change** are synonymous and thought to provide a more complete account of individual hypothesized processes or indicators which might explain how one phenomenon influences another (32). **Logic models** have been described as visual representations of a system, its elements and their interrelationships (33). These posit a 'chain of activities needed to achieve an outcome' (32):p.3); however, they may imply a single pathway to a single outcome. It is obvious that these definitions overlap, particularly in whether the interrelationships of concepts are examined or not. For example, theories or conceptual frameworks can describe multiple mechanisms or multiple logic models. For the purposes of this thesis, 'conceptual frameworks' will be

considered a type of theory; and ‘mechanisms’ used to denote single hypothesised processes between a phenomenon and outcome.

Systematic reviews describe varying methodologies for framework synthesis, suggesting some parallel variation in the underlying epistemology. For example, some reviews describe or suggest purely deductive (or *a priori*) approaches (20), whilst others describe methods indicative of more inductive (or emergent) approaches (22). At the onset of this project, no in-depth consideration had been undertaken of the epistemological foundations of framework synthesis, when to use the method, the different ways it can be used, and what it can provide.

Framework synthesis is a research approach, or method, employed increasingly in EPPI-Centre systematic reviews over the past decade. With each application, we have used the framework synthesis approach innovatively to address different research questions required for diverse health and social care policy needs. I have been involved in several systematic reviews utilising this approach, in each case leading or co-leading on review planning, conceptualisation, synthesis, report writing and project managing large research teams; most recently leading as a primary investigator. This thesis offered for consideration as a PhD by publication has consolidated the methodological literature on framework synthesis, situated my research within it, addressed the gaps identified, and developed guidance for future use of the method. This constitutes a substantive contribution to knowledge about the methods and applicability of the framework synthesis approach within systematic reviews.

[Aims and research questions](#)

The aim of this thesis is to demonstrate how my development of framework synthesis methods is situated within and contributes to wider debates about the research synthesis methods used in systematic reviews. The thesis addresses the following research questions:

1. What do we know about framework analysis methods in health research synthesis (i.e. systematic reviews)? In particular:
 - a. How do the methods compare within research synthesis?

- b. How is the framework synthesis approach located within the context of a range of research synthesis methods?
 - c. What problems does the framework synthesis approach address that other methods do not?
- 2. From examining the applicant's publications that have used this approach, what is gained from the different ways that framework synthesis methods can be applied in research synthesis? In particular, when applied in:
 - a. Qualitative synthesis and interrogating evidence of impact of interventions?
 - b. Theory development and testing?
 - c. Knowledge exchange/priority setting/public involvement?
- 3. How does the use of framework synthesis methods in these publications compare with those utilised in other systematic reviews?
- 4. What areas for the future development of framework synthesis methods for systematic review research synthesis deserve further attention?

1.2 Methods

Thesis design

The research questions posed by this thesis will be addressed using an 'overview of reviews' design. Overviews of reviews compile evidence from multiple systematic reviews in order to provide a comprehensive review of an area, sometimes including studies not included in systematic reviews (34). While they can present challenges where included reviews are disparate in terms of their aims and measured outcomes, they can support decision making (35).

Synthesis

In order to understand the previous uses of and discussions about the framework synthesis approach, and then place my submitted publications within that academic literature, I used framework synthesis methods. An initial conceptual framework of framework synthesis is presented in Chapter 2, and a resulting framework synthesis of themes developed. Next, supporting publications were analysed and reported (Chapters 3 to 6) using the framework developed in Chapter 2; and a new framework synthesis developed to integrate previous literature on framework synthesis with my submitted publications. Note that, throughout the thesis, an *initial conceptual framework* refers to the pre-existing conceptual or

theoretical framework selected to which included data are examined; a *final conceptual framework* refers to the conceptual/theoretical framework after it has been synthesised; and *framework synthesis* refers to the overall methodological approach.

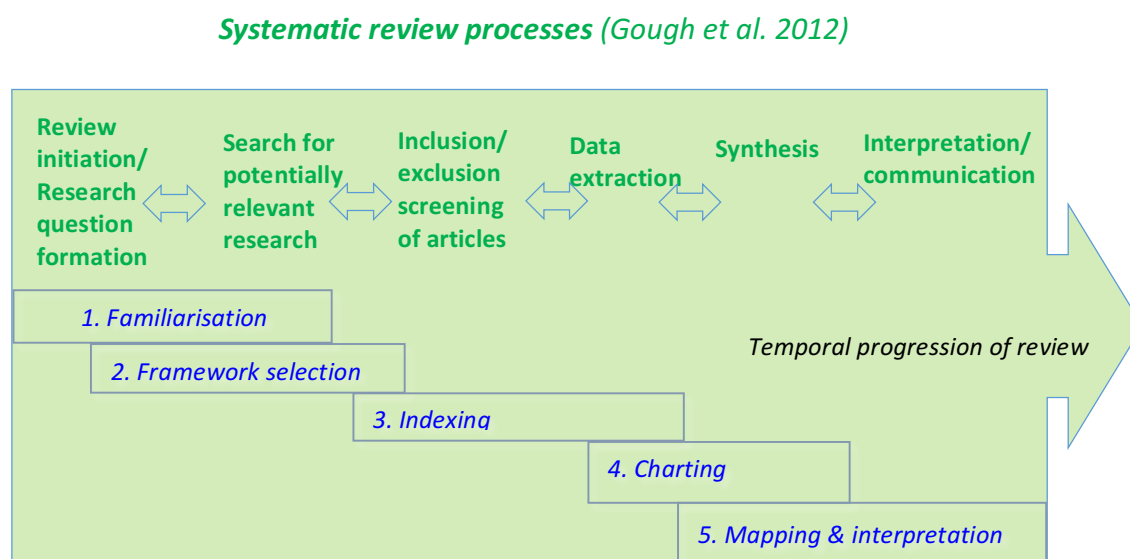
Communication of findings

The second chapter aims to understand and critically reflect on current discussions and applications of framework synthesis methods. Subsequent Chapters 3 to 6 address each of my systematic reviews that employ framework synthesis methods. These findings are narratively synthesised and discussed in Chapter 7, and in Chapter 8, conclusions from the work are drawn in relation to the thesis research questions, with implications made for future research synthesis.

CHAPTER 2. Locating framework synthesis: a systematic review of relevant research

As discussed in Chapter 1, framework synthesis has been used increasingly as a method of research synthesis in systematic reviews. In order to understand how the publications supporting this thesis fit into current academic thinking about the framework synthesis method, a systematic review of the relevant literature was conducted. According to Ritchie and Spencer (1994:174), the questions to be addressed are contextual: they seek to 'identify the nature and form of what exists'. Thus the purpose of this systematic review was to gain contextual understanding of the methods of framework synthesis. I thus followed six key systematic review stages (4), each of which corresponded to the stages of framework synthesis as described by Ritchie and Spencer (27).

Figure 2.1 **Framework synthesis stages corresponding to the systematic review process**



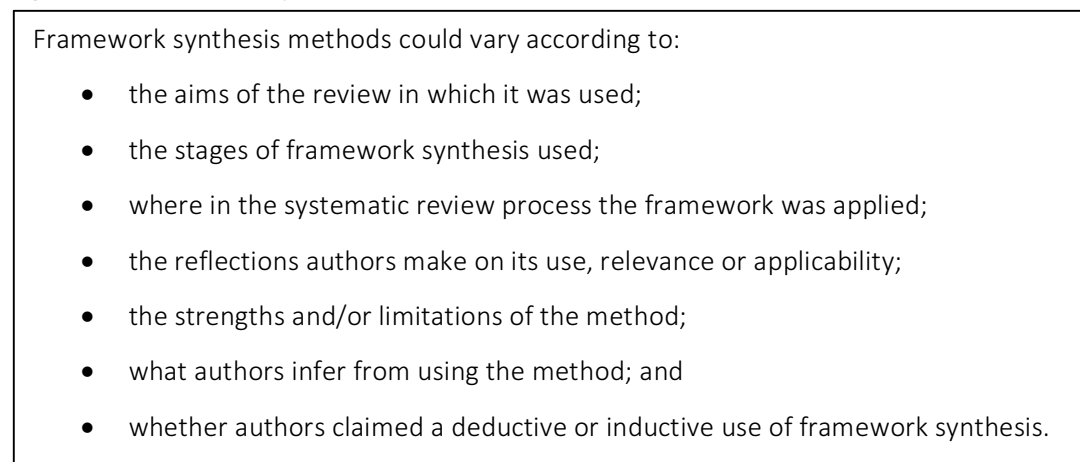
Stages of Framework synthesis method (Ritchie et al. 2014)

As shown in Figure 2.1, the stages of framework synthesis correspond to the systematic review process, but there is some overlap between stages and processes. For example, the familiarisation stage of framework synthesis occurs from a systematic review's initiation until well into searching for potentially relevant research.

2.1 Review initiation

In the **Familiarisation** stage of framework synthesis, I became acquainted with current issues and ideas about the topic under study, by drawing on a variety of sources (19, 27). These sources included previous research, discussion pieces, and my own prior knowledge of the area. This step corresponds to the background scoping conducted to determine the review's boundaries and set the research question(s). This resulted in an initial conceptual framework of factors thought to influence the methods of framework synthesis, illustrated in Figure 2.2 below.

Figure 2.2 **Initial conceptual framework**



The development of review questions reflected the fit between the PhD requirements, my previous publications and the background literature.

Aims and research questions

This chapter aims to address the overarching research question: *What do we know about framework analysis methods in health research synthesis (i.e. systematic reviews)?*

To address this, three specific sub-questions are posed:

1. *How do methods of framework synthesis compare within research synthesis?*
2. *Where is framework synthesis located within the context of a range of research synthesis methods?*
3. *What problems are addressed by framework synthesis and not by other methods?*

2.2 Methods

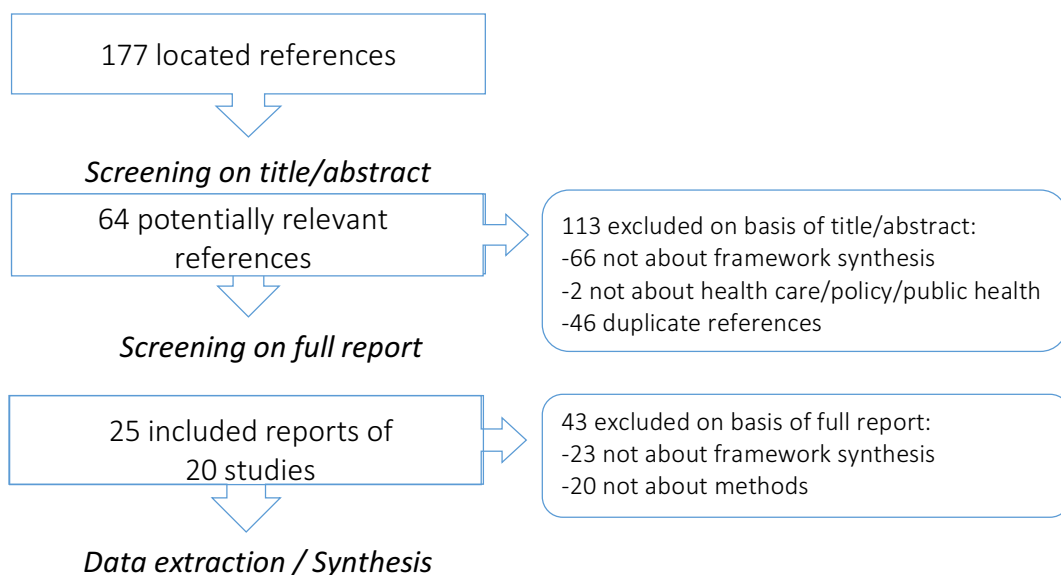
Key papers were identified from content experts, electronic databases, Google Scholar searching, other systematic reviews and reference lists of included reports. No suitable conceptual framework was identified; instead, I developed an initial conceptual framework based on prior research, stakeholder input and researcher knowledge. Reports were screened for inclusion using previously developed criteria, and data from included reports extracted using the conceptual framework as characteristics of framework synthesis methods. Thematic synthesis of framework codes was conducted, and studies were considered critically in the context of the background review's research questions. More detailed methods of conducting this background review of framework synthesis literature are described in Appendix 2.

2.3 Findings

Indexing results

Searching identified a total of 174 unique references. Screening resulted in the flow of studies shown in Figure 2.3 below, as per standards for systematic review reporting (36).

Figure 2.3 **Flow of research reports through the review process**



Of the 177 unique references identified, 157 were ultimately excluded either at title and abstract or full text screening stages: combined, over half (n=89) were not about

framework synthesis; and a further 20 did not provide data reflecting on the use of framework synthesis. Two reports did not focus on health. A total of 46 duplicate references were excluded. Citations of all excluded reports, with their reasons for exclusion, are listed in Appendix 5. Six reports were linked to other publications of the same study; and one of these was linked to a thesis candidate publication. These 'master' and 'linked' studies are listed in Appendix 6.

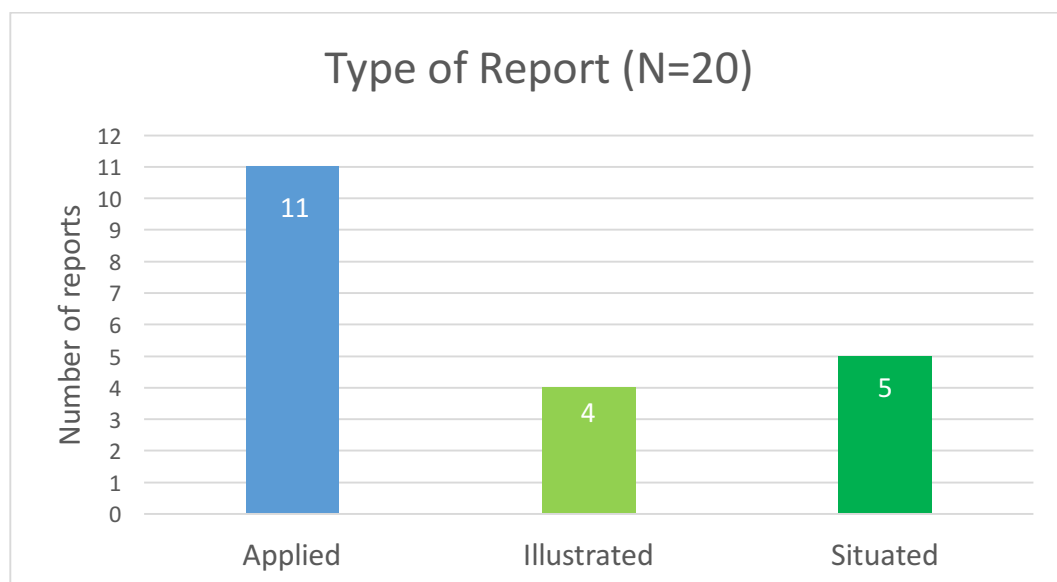
This resulted in the inclusion of 25 papers describing 20 studies. When these remaining 20 included reports were indexed using the initial conceptual framework described above, two new characteristics emerged: the extent of iteration and the epistemological approach employed. These characteristics were added to the initial conceptual framework and included reports were re-indexed.

Charting and mapping results

Aims of included studies

The included reports varied in their stated aims, as shown in Figure 2.4 below.

Figure 2.4 **Aims of included reports**



**Mutually exclusive*

Eleven reports ‘applied’ framework synthesis to understand a phenomenon (37-46); four ‘illustrated’ papers focused on describing specific stages of framework synthesis using a worked example (20, 21, 47, 48); and five papers ‘situated’ framework synthesis within a range of research synthesis methods (18, 24).

The eleven ‘applied’ reports were substantive systematic reviews. Eight of these aimed to understand stakeholder views about discipline-specific health care services (37-43, 45). One aimed to understand the conceptualisation of, measurement of and factors influencing stakeholder involvement in health research (22) and medical device design (44); and one explored influences on setting priorities in health policy across countries (46).

Four reports ‘illustrated’ the use of framework synthesis, providing: an overview of the method in conducting reviews of qualitative research (21); a worked example of methods of conceptual framework selection and development (20); and further refinements of this method, describing a more systematic method of locating, assessing, selecting and adapting an initial conceptual framework and describing its transformation into the final conceptual model (47, 48).

Five reports ‘situated’ framework synthesis within a range of research methods. Two of these examined framework synthesis within the context of qualitative synthesis methods, examining the extent of iteration, *a priori* versus emergent stance and resulting epistemological position (49). Petticrew et al. (2013) discussed and placed framework synthesis within a range of quantitative and qualitative methods used to address complexity in systematic reviews. Authors aimed in part to discuss how qualitative methods of synthesis contribute, characterising methods across a framework of whether it was quantitative or qualitative, the specific method, and its products. Finally, two papers aimed to position framework synthesis within a landscape of aggregative and configurative syntheses (please see p.22 for a further description)(12, 18). The first aimed to identify and discuss the key ‘dimensions’ on which reviews differ and ‘to examine the multitude of different combinations of those dimensions’ (18). The latter aimed to critically describe ‘the range of purposes, data types and analytical approaches underlying different forms of research synthesis’ with a view to describe how to undertake such syntheses (12).

Rationale for selecting framework synthesis method

Authors described choosing framework synthesis because it is a 'pragmatic,' time-saving method (20, 21, 39, 47, 48) that is structured enough to facilitate the organisation and analysis of data using *a priori* theory (24, 38, 43, 45, 49, 50), while also allowing an interplay between a preconceived research objective based on previous knowledge of the literature and results arising from the data (21, 40). Authors described it as suitable where the issue called for a realist perspective (described further on p.27) and analysis was suited to thematic synthesis of data (42, 44, 49, 50). Its utility was also noted in synthesising data aggregatively and configuratively most often seen with mixed methods designs (12, 18, 22, 41). Some authors also noted a wider applicability of the method in that it created a landscape wide enough to include lay perspectives; and resulted in 'data displays' that facilitated stakeholder communications (12, 21, 22).

Methods of framework synthesis

Authors of included reports cited a wide variety of overlapping methodological papers on framework synthesis. Table 2.1 illustrates the pattern of methodological citations provided within the reports.

Table 2.1 Framework synthesis methods papers cited by included reports

	Who/what was cited						
	Review(s) included in chapter	Strauss & Corbin (1990)	Miles & Huberman (1994)	Ritchie & Spencer (1994)	Pope, Mays & Ziebland (2000)	Brunton et al. (2006)*	No reference
Included reviews applying framework synthesis							
Oliver et al. (2008)				✓			
Ram et al. (2008)				✓			
Fishwick et al. (2012)	✓						
Luckett et al. (2012)	✓						
Gallacher et al. (2013)	✓			✓			
Glenton et al. (2013)	✓						
Mytton et al. (2013)	✓				✓		
Kruijsen-Terpstra et al. (2014)	✓						
Walt & Gilson (2014)	✓						
Demain et al. (2015)	✓						
Sohanpal et al. (2015)	✓						
Included reports illustrating framework synthesis							
Carroll et al. (2011)	✓			✓	✓	✓	
Dixon-Woods et al. (2011)	✓			✓			
Carroll et al. (2013)	✓						✓
Booth & Carroll (2015)				✓	✓		
Included reports situating framework synthesis							
Barnett-Page & Thomas (2009)	✓		✓	✓	✓	✓	
Gough et al. (2012)	✓	✓	✓				
Paterson et al. (2012)					✓	✓	
Thomas et al. (2012)	✓			✓	✓	✓	
Petticrew et al. (2013)	✓			✓			
Total citations	16	1	2	9	6	4	1

* – publication submitted for thesis consideration

The method of framework synthesis was described consistently across included reports, with authors often referring to each other's work: sixteen of the reports cited other authors included in this review. Nine included reports cited the method of 'framework analysis' described by Ritchie and Spencer (19) as their reference for framework synthesis. Six reports referred to framework analysis described by Pope, Ziebland and Mays (51), who themselves cited Ritchie and Spencer. Five reports cited methods described in work submitted as part of this thesis (14). Two papers referred to thematic methods outlined by Miles and Huberman (31) and one to Strauss and Corbin (52). The iterative nature of these citations suggests a building of collective knowledge of the method over time. Reports illustrating or situating framework synthesis also cited a wider variety of sources.

Within the method, the process of framework selection varied: nine of the applied reviews selected a previously developed framework during the familiarisation stage (37-41, 43-46). However, one selected a previously developed framework after studies were coded, suggesting that this would limit the risk of the framework biasing their findings, arguing that this ensured that themes were not 'unduly forced' (42). Another (44) amalgamated four existing frameworks, a method supported more recently by Booth and Carroll (2015). The review conducted by Oliver et al.(2008) did not select any pre-existing conceptual framework; instead, authors developed an *a priori* framework based on researchers' experiences of public involvement and the data identified in the systematic search for and appraisal of studies, testing that framework in subsequent stakeholder consultations. The stages of indexing and charting were similarly described across reviews. However, framework methods of mapping and interpretation varied, as described below.

Framework application

Reports were next examined to understand where in the review process framework synthesis had been applied. These suggested that the method is used both across and within stages of the systematic review process. Framework synthesis was used in all ten 'applied' systematic reviews to guide data extraction, with each using thematic synthesis methods to derive higher order concepts. Additional uses were also identified related to the mapping and interpretation stage, specific to knowledge exchange. These included utilising framework synthesis: as a communication tool to set the scope of the review (21, 22); to foster stakeholder engagement throughout the research process (22); to map and interpret findings across and for a range of stakeholders (12, 24, 43, 50)); to make recommendations for future research and policy (24, 40, 50); and to encourage future stakeholder engagement (41).

Relevance, strengths and limitations of the method

Each report reflected on the relevance, applicability, strengths and limitations of framework synthesis. Because these concepts overlap, findings for each concept were pulled together into three main dimensions: diversity; quality assessment; and theory development. In summary, the included reports described framework synthesis as a valuable method to engage stakeholders, noting that it is also a pragmatic, structured way of developing theory. These reflections suggest that framework synthesis can be used to accommodate data that is contextually, methodologically and analytically diverse. It was

described as useful for looking at inter-relationships and theorising causal processes, and suitable for a diverse literature both in terms of the type of data collected (i.e. numeric versus textual) and in combining different foci (e.g. different aspects of one situation, mixed methods synthesis). While quality assessment of the method was considered important, a consensus on how this might be undertaken was not apparent.

Diversity

Six reports discussed the method's utility in handling diverse literature (18, 22, 41, 44, 46, 50). Its value in synthesising different types of data were noted, suggesting that framework synthesis could be used to both 'aggregate' and 'configure' data (18), or integrate quantitative and qualitative research into a mixed methods synthesis (41). For instance, it was noted that this approach 'allowed comparative analysis of varied and complex methods'. Framework synthesis also appeared to address other types of complexity: others emphasised the value of framework synthesis in accommodating a 'diverse literature' encompassing many types of medical devices, multiple methods of user engagement, and varied study designs included in analysis (44). Its ability to handle literature that was diverse by its country of origin (suggesting cultural contextual differences) and methods of comparison (suggesting differences in study design) was also noted (46). Others declared framework synthesis a useful method for systematic reviews examining 'complex interventions' because the method can accommodate interactions between interventions and their geographic, social and cultural contexts, accounting for 'phase transitions' and 'evolution of theory over time' (50):p.1233). This latter idea was echoed by others who, for example, noted that framework synthesis allowed them to understand that the theory underlying parents' experiences of therapy for their children was a 'dynamic entity' that varied over time, according to parents' stage of adaptation (41).

Quality assessment

Three reports reflected on the importance of quality assessment and its impact on synthesis (20, 42, 48). All but one of the 'applied' reviews undertook quality assessment, using a range of established tools. Two of these reflected on quality assessment in relation to the method. One noted that, similar to other methods of qualitative synthesis, framework synthesis could be limited by the quality of reporting of primary research and, related to this, by a lack of rigorous standards of quality assessment for qualitative literature. Rather than providing overall study quality ratings for individual studies, authors presented percentages of the included studies across the dataset meeting each quality

assessment criterion. Authors explained that they ‘took the position that ratings of study quality had the potential to inform the development of the field as a whole but not to classify studies relative to one another’ (42). Authors also addressed assessment of study quality in part by analysing only raw data/text rather than author-derived themes and employing double-rater quality assessment techniques. Similarly, one illustrative example did not limit study inclusion in synthesis by quality ratings, arguing ‘an increasingly strong case for not excluding qualitative data studies from evidence synthesis based on quality assessment’ (20). The quality criteria were derived from ‘relevant critical appraisal checklists for qualitative studies and other systematic reviews of people’s views’. Study quality in this report was reported slightly differently: studies ‘describing clearly’ at least three of five quality criteria were included for synthesis.

Theory development

Seven reports reflected on framework synthesis in relation to theory development (20, 39, 40, 42, 43, 48, 50), although they varied in their descriptions of the extent and timing of theoretical development. For example, these authors noted that framework synthesis was useful where a relevant conceptual framework already existed, or to explore findings from stakeholder research in a structured, explicit way (39, 43). Some described framework synthesis as a useful method of undertaking thematic analysis *without generating new theory* (by using a previously existing theory) to address specific questions about complexity (50). Some deemed it useful in developing a taxonomy and conceptual framework that allowed the examination of relationships and *in theorising causal processes* (40), with systematic methods of searching, assessing and potentially amalgamating theories for an *a priori* theoretical framework suggested (48). Others argued that postponing the selection of a conceptual framework until after coding may limit any biases that arising from the *a priori* choice of interpretive framework (42). These descriptions suggest that theory may or may not be developed during the use of framework synthesis.

Interpretation is part of theory development; however, three reports were contradictory in the extent of creative interpretation permitted (20, 21, 49). One suggested that the method was somewhat less interpretive, but framed this as a strength, since the method is ‘more pragmatic than other forms of qualitative data synthesis’ but still allowed ‘context-specific insights that emerged...over the generic observations already present within the pre-existing model’ (20). Another noted that framework synthesis allowed the use of pre-specified questions could potentially ‘suppress interpretive creativity, and thus reduce

some of the vividness of insight seen in the best qualitative research' (21). Others alluded to limitations in creative interpretation, noting that framework synthesis was a 'highly structured approach' for informing practice and policy, rather than as a tool for developing middle-range theory (such as e.g. grounded theory) (24):p.5). These descriptions suggest that there is variation in the creative interpretation allowed by iteration between data and theory.

Two reports identified the benefits of framework synthesis to address substantive and theoretical gaps by suggesting methods to test the internal and external rigour of the developed theory. This was accomplished: by using 'gap analysis' to reveal substantive gaps in the developed framework that suggest areas for further research 'qualitative sensitivity analysis' (i.e. assessing the impact of study quality on 'frequency' and 'thickness' of study contribution to the framework); 'dissonance assessment' (i.e. consideration of new data that does not 'fit' the conceptual framework) (22, 48).

A priori or emergent methods

Within framework synthesis, data from included studies are applied deductively (*a priori*) to an existing theory; then inductive (emergent) themes derived from the data are developed (12). Thirteen of the nineteen included reports acknowledged that framework synthesis used both deductive and inductive processes (12, 18, 21, 22, 24, 38, 40-43, 45, 47, 48). However, within these studies it was not always clear whether the approach was being used to confirm, modify, or to build theory. Five of the included reports described or inferred a purely deductive approach to analysis; the majority of these also described its use in building theory (20, 44, 46, 49, 50). For example, one initially described framework synthesis as being 'primarily a deductive approach' (20):p.1) but also described the method in both inductive and deductive ways in a subsequent publication (48), although this may reflect the development of understanding that comes with continued use of and reflection on the method. Others also clearly stated the method was used to 'test theory deductively and build theory' (46):p.iii7), perhaps acknowledging the iteration of the coding and thematic development.

Extent of iteration

Fourteen of the twenty included reports either noted or inferred the use of iteration whilst undertaking framework synthesis (12, 18, 20-22, 24, 37, 41-43, 46, 48-50). The remaining

six reports made no reference to iteration. Where iteration was apparent, its extent was not clear and conflicted across reports: six reports either implied or stated that the use of iteration was limited (20, 24, 41, 48-50). For example, Barnett-Page and Thomas (2009) suggested that iteration in framework synthesis was limited to searching only. This contrasted with eight reports suggesting that multiple iterations took place during framework synthesis (12, 18, 21, 22, 37, 42, 43, 46).

Epistemology: generating, exploring or testing theory

It was challenging to understand whether authors were trying to test, explore, or generate theory due to the lack of clear information about the theoretical assumptions underlying each report's use of framework synthesis, the differences in how and when each *a priori* framework was selected or generated, and the apparent differences in positioning framework synthesis as a critical realist or scientific realist epistemology. But in general, it appears that the current research describes framework synthesis as a method that deductively (and to some extent inductively) tests and builds theory, based on realist assumptions that a shared understanding of an independently existing external reality is possible (53).

Across the reports, authors either clearly stated or alluded to a stance that framework synthesis was underpinned by a realist epistemology, in contrast to one based on idealism. Some authors clearly identified framework synthesis as having a critical realist approach (24, 50). Others stated that 'the objectives of the current review positioned it within the realist perspective' (42):p.232). One suggested framework synthesis as a method that held to 'a more realist stance that highlights the possibility for research to adequately represent an external reality.' (49):p.6). The remaining 16 reports did not clearly identify the epistemology underpinning framework synthesis.

2.4 Discussion

Mapping and interpretation of results

The utility and value of framework synthesis has been reflected upon in other substantive reviews and in discussion papers that apply or illustrate its use; and in those papers situating it within a range of synthesis methods. Framework synthesis has been used across a variety of health and health systems topics, employing a range of previously existing

conceptual frameworks. However, it has also been used to build theory from historical researcher knowledge and from issues identified in the literature, suggesting that the method's markers of using existing theory and *a priori* selection have been broadly interpreted. This suggests that the method itself is not prescriptive but is evolving, as other qualitative methods have done (49).

The findings from across each of the concepts assessed in the initial conceptual framework suggested four main themes that reflect the ways in which framework synthesis differs across methodological papers:

- The conditions (context, aims and diversity) of reviews;
- The methods of framing;
- The ways in which gaps are assessed across reviews; and
- The epistemological approach underpinning a review.

Theme 1: Context of the review

The included reports varied in their purposes and aims and in their rationale for selecting framework synthesis as a method. This appeared to be dependent upon the circumstances and conditions under which the review was undertaken: most often to understand participants' views of an aspect of a medical illness (e.g. treatment burden or participation in care). Two early examples sought to understand broader social issues of public involvement in research and medical device design. Differences were also apparent in the reasons for selecting framework synthesis (i.e. speed, theory development/data interpretation, nature of the data or analysis).

Theme 2: Methods of framing

Framework synthesis was applied in different ways across reports, being used to frame the topic and scope of a review, the concepts under study, the synthesis and the knowledge exchange with stakeholders. Whilst following steps outlined by Ritchie and Spencer (1994), authors varied in descriptions of how and when the *a priori* conceptual framework should be selected, and in how and when the framework informed the research process: at the beginning, during data extraction and synthesis, and/or for communication. In addition, it was described by most (although rarely evidenced) as a valuable method to privilege stakeholder views, in that it enabled stakeholder questions to be addressed, translated

research findings for use by stakeholders, and made it easier to embed stakeholders' experiences into future intervention development.

Theme 3: Assessing gaps

The included reports suggested that framework synthesis usefully assesses how well included research fits a selected theory; however, it is not always apparent whether authors assess how well a theory fits the data. For example, all the applied reviews examined the fit between their included research and the selected conceptual framework ('dissonance assessment'); however very few examined whether the chosen framework contained elements not addressed by their included research studies ('gap analysis'). To ensure that framework synthesis is robust, it was suggested that missing or 'uncomfortable' data should be assessed to evaluate potential gaps between the selected framework and the data (48). This has not been undertaken consistently across the included applied reviews, although most were published before Carroll's paper. Authors also consistently advocated consideration of the impact of gaps in study quality on review findings. Quality assessment methods varied between included papers in that every study used a different tool.

Theme 4: Epistemological approach

While researcher stance was not consistently described, the discussions and reflections across this set of 'situated', 'illustrated' and 'applied' reports about framework synthesis suggest that it is a critical realist research method.

The ontological stance taken by a realist approach is that an independent external reality exists and can be approximated (53). Developed by Bhaskar in the 1970s, critical realism is suggested as an epistemology that differentiates between the 'real' world, in which a phenomenon, such as structures or power relations between people, exists independent of our knowledge of it; the 'actual' world, in which structures or power are activated; and the 'empirical' or observable world (53, 54). Critical realists are interested in causality through examining the mechanisms which activate a structure to produce a particular event. These mechanisms are seen as having multiple conditions which may or may not activate them, suggesting that causality can be studied but that it is not linear in nature (e.g. that X produces Y). Further, because knowledge of the world is dependent on a historical context rooted in particular social discourses, understanding also requires

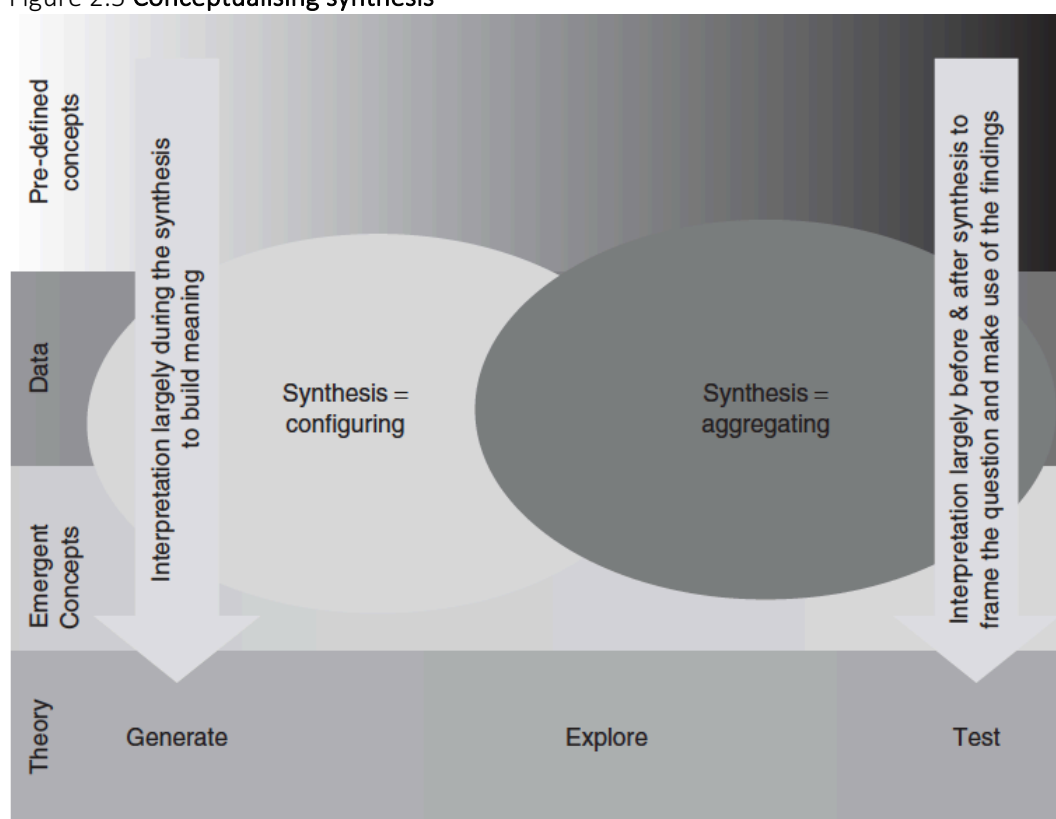
interpretation. Over time, knowledge can evolve and thus understanding is not an absolute 'truth' and is considered potentially fallible. Hence, the world can be explained in terms of cause and effect using critical realism, but for these reasons it does not predict causation. This was demonstrated across the included reports: the use of framework synthesis and descriptions of epistemological stance in some of the reports demonstrated that the knowledge derived from a framework synthesis review is mediated by people's beliefs and experiences, which are subject to what has been described as 'epistemological relativism': that reality can be known but is also a function of time and place (53). This is in contrast to positivist stances which examine cause and effect between variables in order to predict the occurrence of a phenomenon (27), and interpretivist stances which claim that the world can only be understood through meaning alone (26). Thus, critical realism has elements of both positivist and interpretivist stances embedded within it, and it has been presented as a 'third way' between positivist and interpretivist epistemologies (53); these are discussed earlier in Section 1.1.

Overall, it was often challenging to understand whether authors were trying to test, explore, or generate theory, potentially due to the lack of clear information about the theoretical assumptions underlying each report's use of framework synthesis, and the lack of a clear description of critical realist epistemology underpinning the framework synthesis. It was also difficult to determine whether framework synthesis was utilised to explore, test or generate theory because of the differences in the selection or generation of each report's *a priori* framework.

Framework synthesis was described as a pragmatic structured way of developing theory, and was also deemed useful for looking at inter-relationships and theorising causal processes. However, some suggest that it is an approach more suitable for exploring causality rather than addressing mid-range theory (55), and implies that framework synthesis may be less suitable for idealist review approaches (i.e. when underlying assumptions are that there is no shared reality and authors try to build up a theory of what is occurring). Further, it was suggested that it may have less utility when the conceptual understanding of a phenomenon is very clear and data do not need as much 'contextualising'. However, in some reports, the application of data about people's experiences to a previously existing theory suggests that framework synthesis may also be used to exercise judgmental rationality (i.e. the ability to decide rationally on the merit of

a theory). Thomas and colleagues (12) suggest a useful way to place framework synthesis within the range of qualitative methods and epistemologies, illustrated in Figure 2.5 below.

Figure 2.5 **Conceptualising synthesis**



(From Thomas et al. 2012: p.181)

In this model, data can be used to test, explore or generate theory, as illustrated along the bottom row. Whether data are synthesised configuratively (i.e. as in creating a 'mosaic' of ideas), aggregatively (i.e. as in pulling together similar findings) or a combination of both is dependent on, and reflected by, the research question, the extent to which concepts are pre-defined or allowed to emerge during synthesis, and the amount of interpretation between data and those concepts (12):p.181). Framework synthesis can be placed as a method with mostly pre-defined concepts, which are both aggregated and configured, in order to explore theory. However, this may stretch toward testing or toward generating theory depending on the nature of the research question under study.

In summary, discussions from included reports suggest that in thinking about the underpinning ontology and epistemology, framework synthesis sits within a critical realist approach that involves varying degrees of exploration and testing of theory, dependent on the review's context, aims and research question, which also imbue the method with some

interpretive qualities. This blurring of epistemological distinctions when categorising the method suggest that it is a method of 'selective eclecticism' (56), where synthesis methods and their underlying paradigms are mixed in order to best suit the context and research question under study.

CHAPTER 3. Framework synthesis to compare qualitative and experimental evidence

3.1 Context of the review

The systematic review considered in this chapter sought to understand the evidence base concerning the public's views of walking and cycling as modes of active transport, in order to assess specifically whether children's, young people's and parents' views of the barriers to, and facilitators of, walking and cycling matched interventions evaluated for their effects on walking and cycling. This issue arose because the Department of Health wished to understand how to facilitate more physical activity through active transport. A recent review of interventions to encourage such behaviours had determined limited effectiveness, but identified a need to understand influencing factors (57, 58).

Our research team was developing methods to integrate findings from qualitative research alongside those from intervention evaluations. This review presented an opportunity to examine the fit between research findings from qualitative studies of people's views and relevant evaluated public health interventions; in effect, exploring interventions' 'fit for purpose' (18). We were relatively inexperienced in framework synthesis: two researchers were novices and I had ten years' experience of both quantitative (statistical meta-analysis) and some qualitative synthesis (thematic synthesis), but had not used framework synthesis. However, the lead on this project had undertaken framework synthesis previously (22).

My contribution to this review came from a realist stance. At the time of undertaking this work, I was more comfortable about categorising, rather than interpreting, data. This was a result of my clinical nursing background, which was grounded in an empiricist medical model (59). Thus, I felt it would be easy to approach the first part of framework synthesis, as I was able to consider data in a categorical manner. However, I was also open to new ways of synthesising research, in that my nursing experience in hospital and community showed me that life was not always so clear cut and that understanding the world around me could be more nuanced.

The review framed two distinct phenomena: factors thought to influence walking and cycling; and the fit between the public's views and the effectiveness of evaluated

interventions to promote walking and cycling. In the synthesis of studies of people's 'views', the factors thought to influence active transport were framed into themes derived from looking separately at children's young people's and parents' views of walking and cycling, organised at the level of the individual, family, community and society. Implications for interventions derived from the main themes arising from the public's views about barriers and facilitators to walking and cycling at the community/society level then comprised a second synthesis, framing whether the intervention studies from the Ogilvie et al. (2004) review fit with what people said was important.

Stakeholder input also framed this review. Early in the process, an Advisory Group was convened consisting of key academics, policy experts and consumer representatives. They advised the research team on which populations and aspects of active transport to explore. In the first meeting, stakeholders were presented with findings from a mapping of research literature on participants' 'views': the framework of studies coded by population, barrier/facilitator type and level of operation. Ensuing discussions focused on determining the most relevant populations for more in-depth study, with children and young people's travel to school deemed most relevant for policy. Within this, specific aspects were requested, including: differences in active travel by socioeconomic status; environmental/structural factors influencing active travel; the relationships between individual behaviour change and outcomes; and the mismatch between attitudes and behaviour. At a subsequent meeting, the research team presented the synthesis of findings from views studies and the proposed methods used to frame the interventions using implications derived from the views synthesis. Advisory group stakeholders also shaped the focus of synthesis in terms of the populations of interest (children, young people and parents), the context of active travel (active travel to school), and the most policy-appropriate and efficient level of intervention to examine (societal/community). They requested that data from lower quality studies should also be included, to ensure that modifiable barriers and facilitators of active transport could be known and potentially explored further.

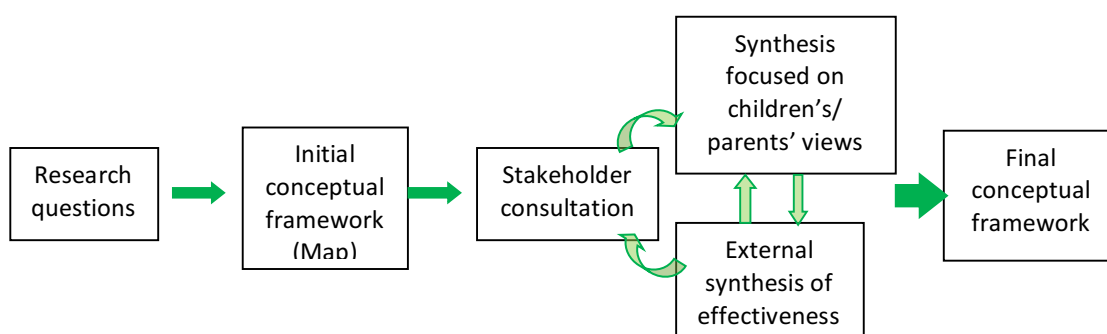
3.2 Approach

Review design

The review comprised two framework syntheses, conducted in three parts. First, we searched for and mapped the existing research literature on the general public's 'views' of

walking and cycling. Second, we conducted an in-depth review of a subset of this literature, the scope of which was selected by our Advisory Group, focusing on the views of children, young people and parents. These two parts comprised the first framework synthesis. Third, we synthesised the findings relating to these 'views studies' together with a recent review of interventions (57): the 'cross-study synthesis'. This third part comprised the second framework synthesis (although the review's reported methods do not describe it as such). Here, we used the themes derived from the findings of people's views to derive implications for future intervention development, which were assessed against the characteristics of the intervention studies, in order to both test the framework synthesis themes and the extent to which intervention design addressed themes derived from public perspectives. This is illustrated in Figure 3.1 below.

Figure 3.1 **Flow diagram of walking and cycling review**



Framework selection

While no single theory was located concerning active transport, our research team's initial scoping literature searches identified several factors, including weather, time, geographic location, and distance; which had yet to be 'pulled together' coherently. These acted as the initial 'framework' for categorising views: as recognising a barrier or facilitator, by the age or sex of persons expressing the view; and whether barriers or facilitators occurred at the level of the individual, family, community or society.

Indexing

We handled data and findings in both a configurative and aggregative manner. In the first framework synthesis, when building the framework for the views synthesis, we initially aggregated data by each factor identified in the background literature, adding new ones as they were identified. Each factor was further grouped by child, youth or parent stakeholder group, and by the individual, family, community or societal level at which it operated.

Coding into the initial conceptual framework allowed us to examine the studies' characteristics, for example by participant age (children, youth, adults) or journey type of (e.g. walking, cycling or by car). We then coded the barriers and facilitators raised by participants (e.g. 'weather', 'neighbourhood' etc.). We looked across studies to discover patterns between each characteristic and the identified barriers or facilitators (e.g. if children expressed a desire to walk more often than young people), aggregating these according to frequency. Next, looking at the most frequently occurring factors, we configured themes from data within each factor across all levels at which they operated. For example, we explored differences according to participant age, sex or the area socio-economic status; urban, suburban or rural location; and whether the view expressed was about walking or cycling.

In the second framework synthesis, we integrated two types of research: public perspectives of walking and cycling; and evidence from a review of effective interventions to promote its use (57). Here, we coded interventions by their main characteristics (target population, intervention type, setting), level of intervention (individual, family, community, society) and content. We compared these, by level, to the implications for interventions derived from the themes that arose from examination of barriers and facilitators identified by views studies.

Charting

My epistemological approach during the first framework synthesis was initially deductive (applying data to an *a priori* concepts identified in the literature), with more induction as the review progressed (requiring more interpretive creativity during the thematic development stage to allow themes to emerge). The epistemological approach in the second framework synthesis, while not reported in the review, was *a priori*, testing theory by comparing implications derived from themes against intervention characteristics. Background literature scoping conducted as part of the review revealed several factors thought to influence children and young people's walking and cycling to school. These operated at the individual, family, community and societal levels; however, no mechanisms suggesting interactions of these factors were present as would be expected in a previously existing theory.

However, there was also an emergent element to the first framework synthesis. When the findings about factors influencing people's walking/cycling were examined for recurrent themes across all age groups, overarching themes emerged. These included a culture of car use; fear and dislike of local environments; children as responsible transport users; and parental responsibility and behaviour. These suggest some mechanisms potentially underpinning a theory to explain children and young people's engagement in walking and cycling.

The second framework synthesis was *a priori*, in that intervention characteristics were compared to views-derived themes. Again, there was also iteration back and forth between the initial conceptual framework, the data and: barriers/facilitators; level of operation (individual, family, community, society). Iteration also occurred between themes and barriers/facilitators; populations; and journey type. In one sense, the concepts were pre-defined and the data were fairly fixed. Some interpretive creativity occurred as themes were derived from what was said about each concept (i.e. barrier or facilitator) according to children, youth and parents. The extent of interpretive creativity at the second framework synthesis was more limited, in that characteristics of interventions (e.g. physical environment changes) were compared to themes (e.g. fear of local environments) to derive a 'fit' between the two.

Mapping and interpretation

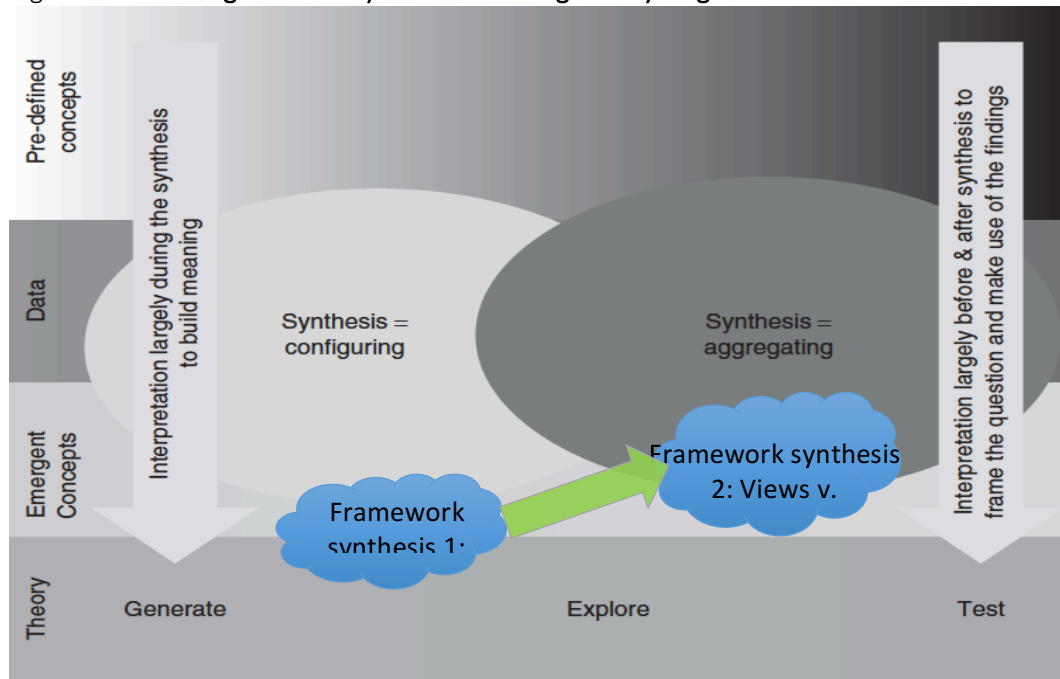
To map and interpret the data, gaps in methodological quality were assessed. In the first framework synthesis, each views study was assessed for methodological quality using a previously developed EPPI-Centre tool (60). This tool assessed sampling methods, sample description, reliability and validity of data collection tools and methods, the extent to which people's views were privileged and the usefulness of the study for the review's questions. Studies were rated on a score from zero to 12 based on the extent to which each criterion was met; those rating seven or less were not included in views synthesis. However, these were listed for reference and later used in the second framework synthesis.

A qualitative sensitivity analysis was also undertaken. Common contemporaneous practice was to exclude studies that were of lower methodological quality so that their findings did not bias the results of the synthesis (3). However, as Advisory Group members did not wish to lose the information contained in these studies, we examined differences in findings

when lower quality studies were included or excluded from the synthesis.

Overall, this places the framework synthesis in this review in the middle of our conceptual model of research synthesis (see Figure 3.2 below), leaning slightly more toward configuring (building/exploring) theory using pre-defined and then emergent concepts.

Figure 3.2 **Situating research synthesis: Walking and Cycling Review**



(From Thomas et al. 2012:181)

This systematic review began by taking several pre-determined concepts about walking and cycling to school that had not yet been woven together into a theory. Higher order concepts were then derived from what was said about walking and cycling by children, youth and parents. Implications for interventions were developed from these higher order concepts, and these were assessed alongside intervention characteristics to determine the fit between factors influencing walking and cycling to school and interventions that might address active travel. Looking at the fit between intervention characteristics and themes derived from views allowed us to understand whether some characteristics of interventions could be potentially related to bigger effects, suggesting a theory of 'how' an intervention may work.

Assessment of gaps in theory occurred during the second framework synthesis, where gaps between interventions and themes derived from views studies were compared (including

the methodological quality of each). These are illustrated in the final report tables on pages 117 – 122 (14). During the second framework synthesis, gap analysis was undertaken as intervention studies were examined to see whether they addressed an identified barrier or facilitator, as well as which barriers and facilitators had not been addressed in intervention studies.

All concepts had data attached to them, and newly identified concepts were added to the framework. A ‘dissonance analysis’ was undertaken, however no contradictory data were identified but there may have been in unexamined concepts operating at other levels not synthesised (e.g. individual, family levels). Further, differences in populations and ages were sought as part of the analysis.

3.3 Discussion

The synthesis of the public’s views on walking and cycling using framework synthesis was undertaken using methods similar to other framework syntheses discussed in Chapter 2 (42, 43). However, this review went beyond other systematic reviews using framework synthesis in three ways. First, it conducted a ‘two stage’ review(18) of a body of research followed by a synthesis of a subset of studies on a topic informed by our Advisory Group – the initial conceptual framework presented clearly to the Advisory Group the characteristics of the literature available for synthesis. This was a methodological innovation for systematic reviewing, which we believe allows decisions to be made on relevance that are better informed by the breadth and depth of research (18). This type of review design allowed us to employ a second innovation: we used the findings of the framework synthesis of participants’ views of walking and cycling to undertake a second framework synthesis that examined the fit between the themes derived from that views synthesis and the interventions developed to foster walking and cycling identified in the systematic review of effectiveness (57).

As with all systematic reviews undertaken by our research team, this review was undertaken with the help of an Advisory Group of key academics, policy experts and consumer groups. Gaining input from stakeholders was becoming increasingly important at the time (61, 62), particularly the inclusion of consumer representation (63-65). Here, we used the framework as an innovative mechanism to engage consumer groups and accurately capture their perspectives within synthesis.

The questionable contribution of qualitative studies of lower methodological quality provided challenges in this review, and illustrates some of the contemporaneous arguments about qualitative and quantitative research methods (3, 66). The inclusion of lower quality intervention studies to assess the fit between interventions and the public's views as potentially relevant but requiring further evaluation was seen as an appropriate way to foster knowledge transfer without advocating the implementation of potentially unreliable findings. The review process included assessment of included views studies' methodology using standard quality assessment tools. Studies rating 'low' were not included in synthesis but were listed for reference. This was appropriate given review synthesis methods at the time (67, 68). Overall, these findings suggest that framework synthesis has utility in mixed methods synthesis and in structuring stakeholder discussions.

CHAPTER 4. Iterative consultation for theory development in framework synthesis

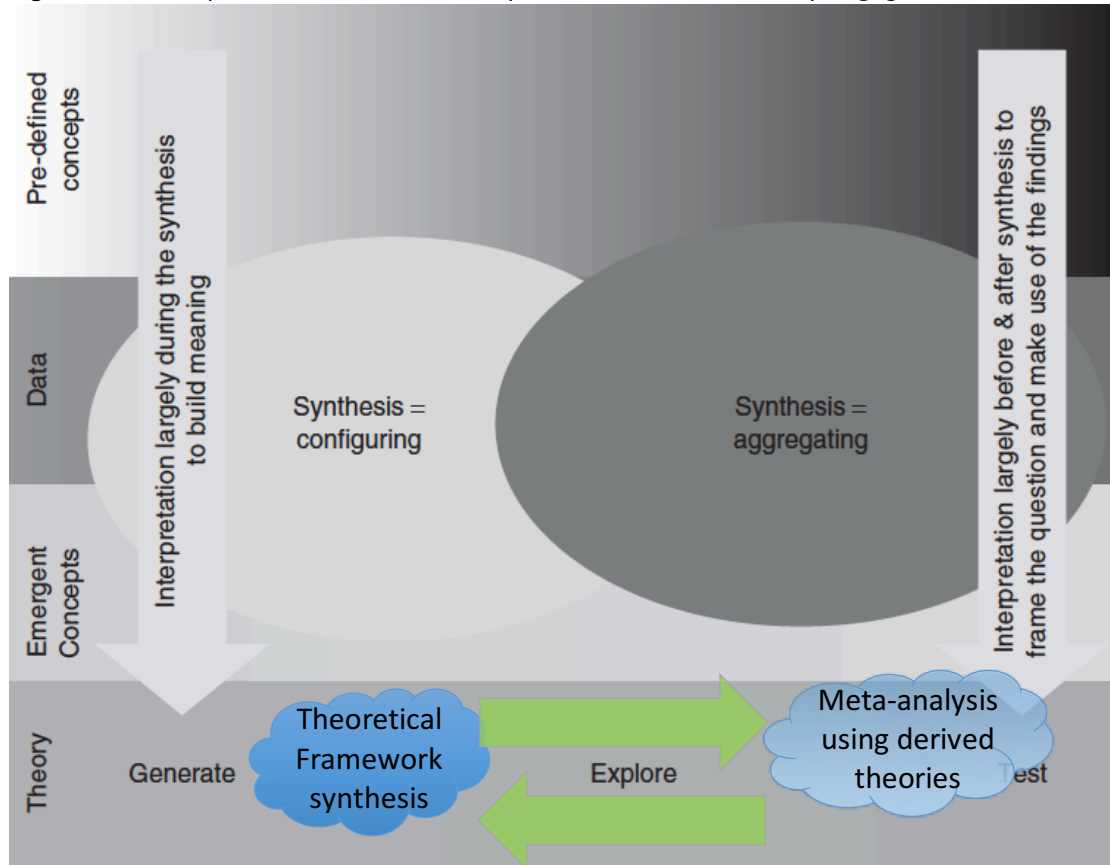
4.1 Context of the review

Recognising the importance of involving local communities in activities relating to their health, the National Institute of Health Research (NIHR) requested evidence about the effectiveness and cost-effectiveness of community engagement interventions. However, they also acknowledged a lack of understanding about the specific ways in which community engagement worked to influence health, suggesting a need to examine more closely the relationship between theory and interventions (69). Our research team wished to examine community engagement in more detail, as we consistently encountered this strategy in many intervention evaluations. As a process, community engagement involves people in any or all of the design, delivery and evaluation of interventions to improve the health of themselves and/or their communities. Such community engagement interventions are often unique, tailored to the community in question, and can be potentially multiplicative, i.e. interventions can reinforce intended health behaviours such as healthier choices; but can also improve social outcomes such as social capital (70, 71). This call for research presented an opportunity to develop methods of using theory to drive statistical analysis, which could in turn further our understanding of complexity often encountered in systematic reviews.

This review aimed to examine: the effectiveness of approaches that improve the health of disadvantaged populations; the circumstances in which such interventions work; and the costs associated with implementation. This required multiple syntheses, addressed by several research questions. The theoretical synthesis aimed to address the range of models and approaches, and mechanisms and contexts through which community engagement takes place. Meta-analyses and thematic syntheses would examine the effectiveness and cost-effectiveness of community engagement, determining for whom interventions are effective and under what circumstances. These syntheses required the configuration of data and concepts, which were subsequently aggregated to test whether some aspects of community engagement were associated with larger health effects. Because each synthesis informed the next, and all syntheses informed the final conceptual framework, this review used an approach that moved between *a priori* and emergent methods.

Iteration occurred between the framework and the data during each synthesis. The relative placement of each of these syntheses is illustrated in Figure 4.1 below.

Figure 4.1 Conceptual model of research synthesis: NIHR Community engagement review



(From Thomas et al. 2012:181)

Within the synthesis of theoretical literature, interpretation took place during the synthesis in order to build understanding about community engagement. The pre-defined concepts were those produced in the initial conceptual framework. The data were more textual than numeric in nature, requiring configuration of ideas and concepts, but also some aggregation as process data were also considered. Concepts emerged from the textual data provided by authors on the nature of community engagement. Previously described theories were integrated into synthesis and newly identified mechanisms suggesting how community engagement worked were added into the framework.

Stakeholder input during this project was expected to provide difficult-to-locate evaluations and discursive literature on community engagement, but also crucially for this

review, perspective on emerging theories and findings resulting from meta-analysis of effectiveness studies. This is described further in the approach section below.

The synthesis of effectiveness and cost-effectiveness literature was intended to test the theory of community engagement derived from the theoretical framework synthesis. In effect, interpretation took place before and after synthesis in order to frame the research question and interpret the findings. The meta-analysis used the concepts pre-defined in the conceptual framework, aggregating numeric outcome data to derive effect sizes. Three different theories of community engagement (community-identified need, peer designed and delivered, and peer delivery) drove the meta-regressions, examining the association between studies with these mechanisms and subsequent effect sizes. Findings from the meta-analysis and meta-regressions subsequently informed the final conceptual framework of community engagement.

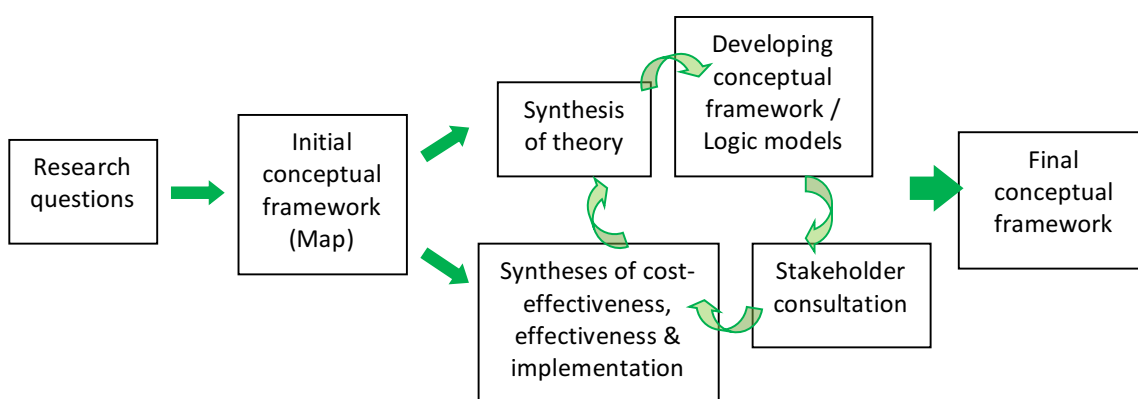
My epistemological stance during this project shifted between positivism and interpretivism (see Figure 2.4 on p.26), perhaps necessarily in response to the demands of the review. I recognised that there were many different ideas in the literature about what constituted 'community engagement', and we needed to develop a theory of how community engagement 'worked'. But my stance still remained grounded in a belief that reality is a shared experience that can be known, suggesting a realist stance.

4.2 Approach

Review design

This was a three-stage systematic review employing framework synthesis in an iterative manner: we conducted a (1) conceptual synthesis of theory to inform our (2) meta-analysis and economic analysis evaluating effectiveness and cost-effectiveness; then the findings from those informed refinements of our (3) conceptual framework developed from the first synthesis. Although the overall approach for the entire project was not described as a framework synthesis, the methods of framework selection, indexing, charting and mapping and interpretation were evident throughout the description of the review's methods. This is illustrated in Figure 4.2 below.

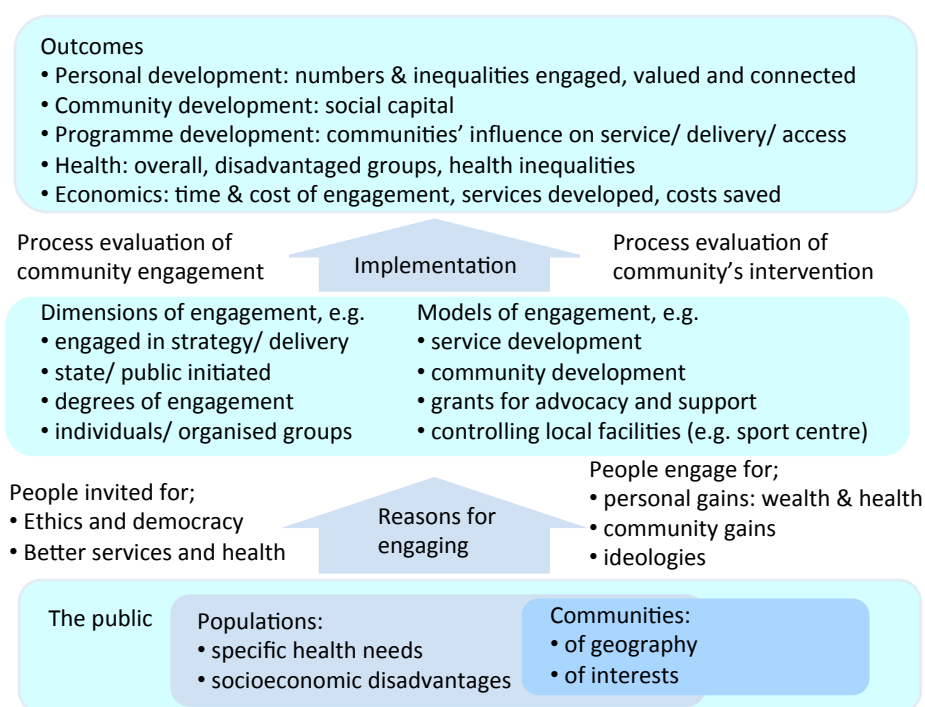
Figure 4.2 Flow diagram of NIHR Community engagement review



Framework selection

An *a priori* framework was developed from researcher expertise and initial scoping searches, organised into a map of models and approaches underpinning community engagement. The initial conceptual framework is illustrated in Figure 4.3 below.

Figure 4.3 Initial conceptual framework: NIHR community engagement review



This figure depicts community engagement beginning with a community of interest ('The public'), defined in particular ways, who were invited or took part for various reasons

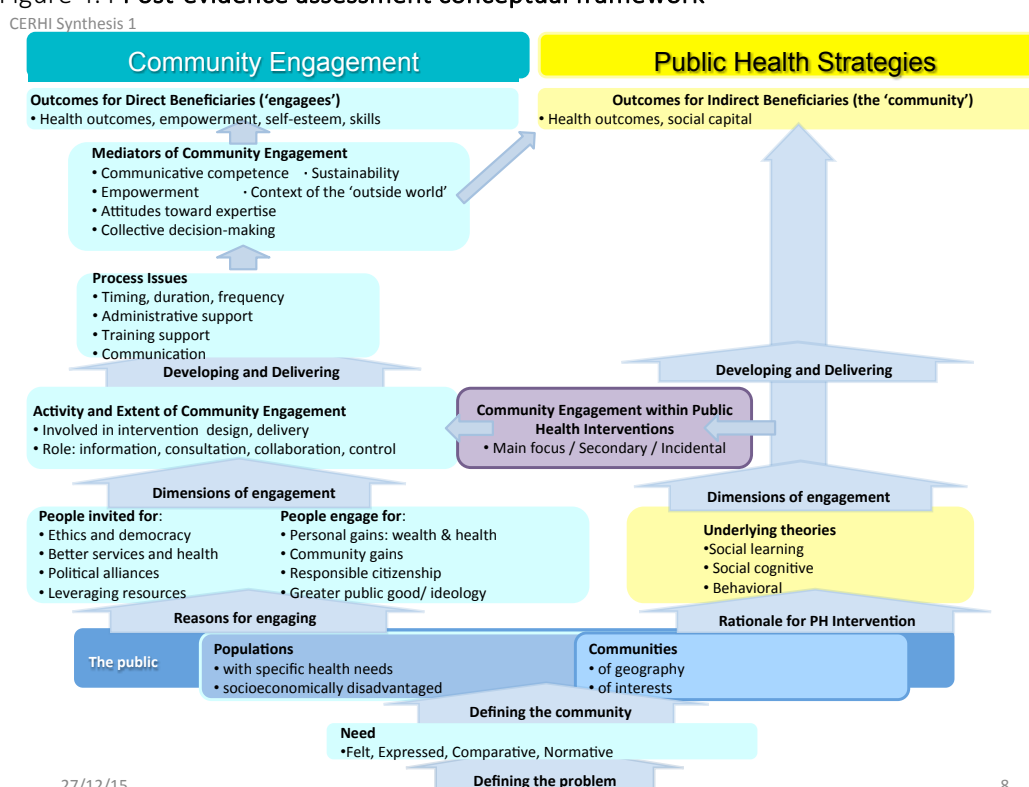
(‘Reasons for engaging’). The manner in which their engagement was undertaken (‘Implementation’) often resulted in particular types of outcomes (‘Outcomes’).

Indexing

Outcome and economic evaluations were coded using the aspects identified in the framework, and patterns of community engagement were sought by looking at combinations of codes within the intervention studies.

The conceptual framework developed appreciably during the project, changing as we moved between the data extracted from process evaluations, outcome evaluations and economic evaluations and then again once we tested it using effectiveness data. In the first stage of the review synthesis, relevant characteristics of community engagement were extracted from process evaluations and discursive studies of community engagement and added to the framework. This added new dimensions to the framework, as illustrated in Figure 4.4.

Figure 4.4 Post-evidence assessment conceptual framework

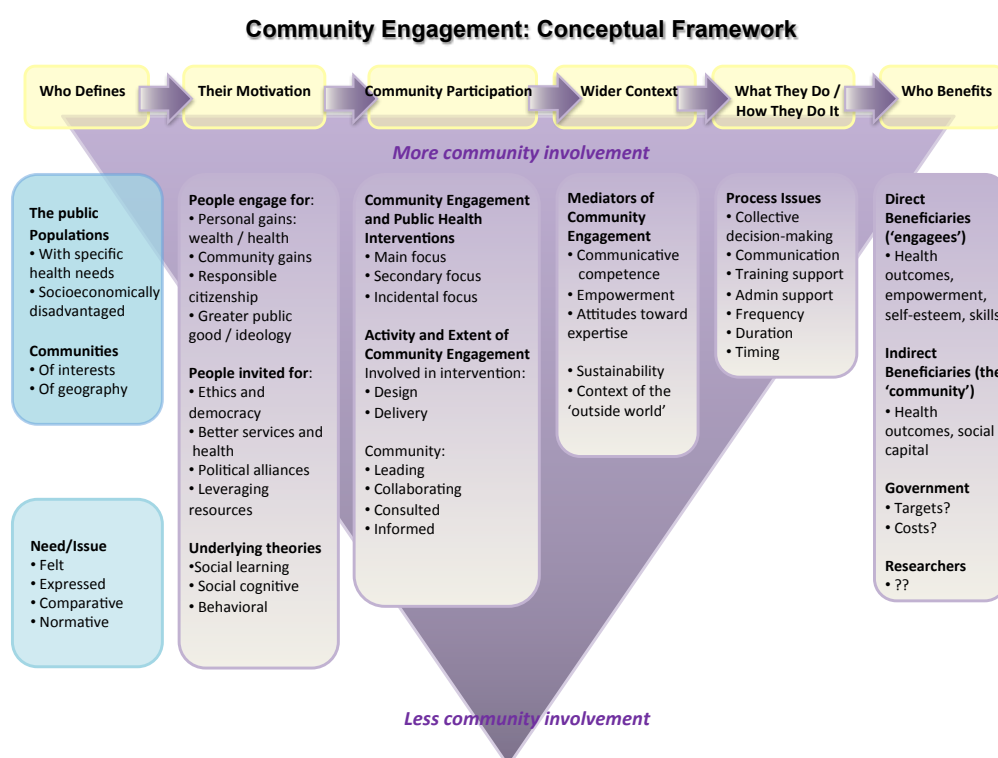


This iteration of the conceptual framework started with a health need (‘Defining the problem’), identified in various ways, prior to the identification of a community (‘Defining

the community’). It also acknowledged two distinct types of interventions to reduce health inequalities: those that employed community engagement as part of a public health strategy, and those that undertook community engagement as the strategy itself. Further, the reasons for engaging or being asked to participate were considered as part of their theoretical underpinnings, which were more evident in the public health literature as the ‘rationale’. The aspects of community engagement began to be unpacked in more detail (‘Dimensions of engagement’) and separated from aspects of implementation (‘Developing and delivering’). Finally, health outcomes were refined into those that directly benefitted engagees versus those that benefitted the community indirectly.

Next, the conceptual framework developed from the research literature was presented to an Advisory Group of key community engagement academics and practice specialists. Their feedback on key aspects of community engagement was added into the framework. At this stage, the framework was also reconfigured for clarity. These changes are illustrated in Figure 4.5 below.

Figure 4.5 Post-advisory group conceptual framework

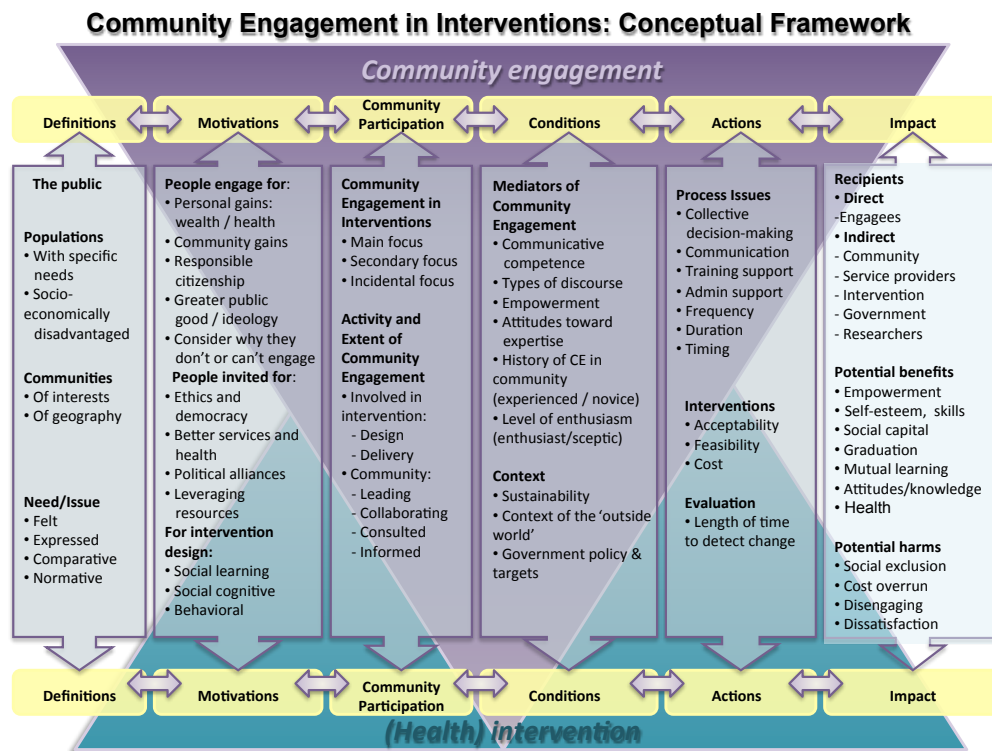


These methodological actions transformed our thinking about the framework. It revealed dimensions of community engagement that were driving our thinking about what influenced community engagement: who defines the issue, what motivates people, the variation with community engagement, what mediates it, what processes influence it, and the types of outcomes and recipients who benefit from community engagement. Dimensions appeared as the yellow boxes at the top of the figure. Characteristics within each of the dimensions lined up below. The triangle was added to represent that some interventions contained more 'community engagement' and characteristics within each dimension were lined up to demonstrate more community engagement (at the top) and less community engagement (at the bottom).

Charting

Examination of the patterns of community engagement revealed by the combinations of codes within the intervention studies revealed three mechanisms of community engagement, which represent the overarching themes of the final conceptual framework: (1) those where the health need is defined by the community; (2) those that involve the community in design and delivery; and (3) those that rely on community members to deliver an intervention. These themes were tested subsequently against outcome data, using meta-regression techniques. Finding suggested that no one theory of change was associated with larger effect sizes; models that were founded on public health with a little community engagement were equally as effective as those that were completely grounded in community engagement and influenced health outcomes. Thus, a second triangle representing more and less health interventionist stance was added to the framework. In addition, a final consultation with advisory group members suggested that community engagement was more likely to proceed iteratively from one dimension to another (e.g. between who defines, their motivation and participation). In addition, more mediators and actions related to implementations and specific outcomes were suggested, which were added into the framework. The final conceptual framework is shown in Figure 4.6.

Figure 4.6 Final conceptual framework: community engagement



(From O'Mara-Eves et al., 2013)

Mapping and interpretation

Gaps in quality were assessed in a variety of ways. The theoretical literature was critically assessed as it was synthesised (for more detail, please see O'Mara-Eves et al., 2013:43-56). Appropriate standard critical appraisal tools were applied to process, outcome and economic evaluations. All studies were included in synthesis regardless of study quality, and the impact of study quality on effect sizes was assessed for outcome evaluations using sensitivity analysis.

The initial conceptual framework provided concepts that were all addressed by subsequent data extracted from each type of study. In addition, newly identified concepts in the literature were added into the framework, suggesting the need for a thorough gap analysis. Qualitative sensitivity analyses were undertaken in two ways: (1) through critical assessment of theoretical literature and quality assessment of process evaluations; and (2) through discussions with Advisory Group members on the sense, ease of understanding and comprehensiveness of the conceptual framework. Dissonance assessment (i.e. looking at the studies to see whether new characteristics 'fit' with the initial conceptual

framework) was not undertaken, as any new characteristics identified were incorporated into the conceptual framework regardless of whether they fit or not.

Gaps in theory were tested to reveal any substantive gaps in the developed framework suggesting areas for further research. Gaps in mechanisms were identified (e.g. three proposed mechanisms of community-identified need, collaborating or consulting on design and implementation) and tested in the meta-analysis. A need for further testing around extent of engagement throughout intervention design, delivery and evaluation was suggested, as was a need for further evaluation of the processes of engagement.

4.3 Discussion

This review aimed to develop an understanding of what community engagement is, and how it works, by assessing and extracting quantitative and qualitative data from multiple types of literature (i.e. discursive papers and outcome, process and economic evaluations). Framework synthesis was able to handle the organisation of data and concepts arising from this complex topic to reveal several dimensions across which community engagement appeared to operate, and aspects within each dimension to illustrate the varied ways in which community engagement could be enacted, depending on the extent to which the intervention was health-related or community-engagement related.

The real innovations of this systematic review were two-fold: it utilised iterative 'checking' of the developing framework with advisory group members, relying on their substantive and theoretical expertise to further develop the framework (rather than just sense-checking it); and empirical testing of mechanisms derived from the framework on trials of community engagement, in order to get an idea of relative effectiveness of different approaches ('mechanisms') of community engagement.

The method of framework synthesis allowed the incorporation of new concepts into the framework, derivation of mechanisms of community engagement to test against the trials of interventions, and further development of the conceptual framework based on these results. This systematic review was limited with respect to generalisability in two ways. First, the conceptual framework is transferable only to studies reporting on at least one health outcome. Some community engagement literature was excluded because it did not contain health outcomes; however, it could have usefully informed a conceptual

framework of community engagement (just not as it related to health). Second, the trials on which the conceptual framework were tested were those limited to two priority areas identified by Marmot (2010), and not all health inequalities - thus it is not clear the extent to which it is generalisable to communities with other types of health inequalities, or to communities without health inequalities. However, such priority-setting decisions in systematic review are often necessary due to the breadth and scope of public health literature and related complex policy questions (18, 50). Framework synthesis offers a way to be explicit about the breadth of theoretical knowledge possible before focusing down on an area of most interest to stakeholders.

CHAPTER 5. Framework synthesis to derive and test theory

5.1 Context of the review

The review described in this chapter was conducted for the National Institute of Health and Care Excellence (NICE), who requested proposals to undertake an update of the research evidence on the effectiveness of community engagement specifically concerning 'what works, with whom, and under what circumstances' (72):p.3). NICE commissioners intended the findings from this systematic review to inform their Public Health Advisory Committee, tasked with updating the national guidance on community engagement (73).

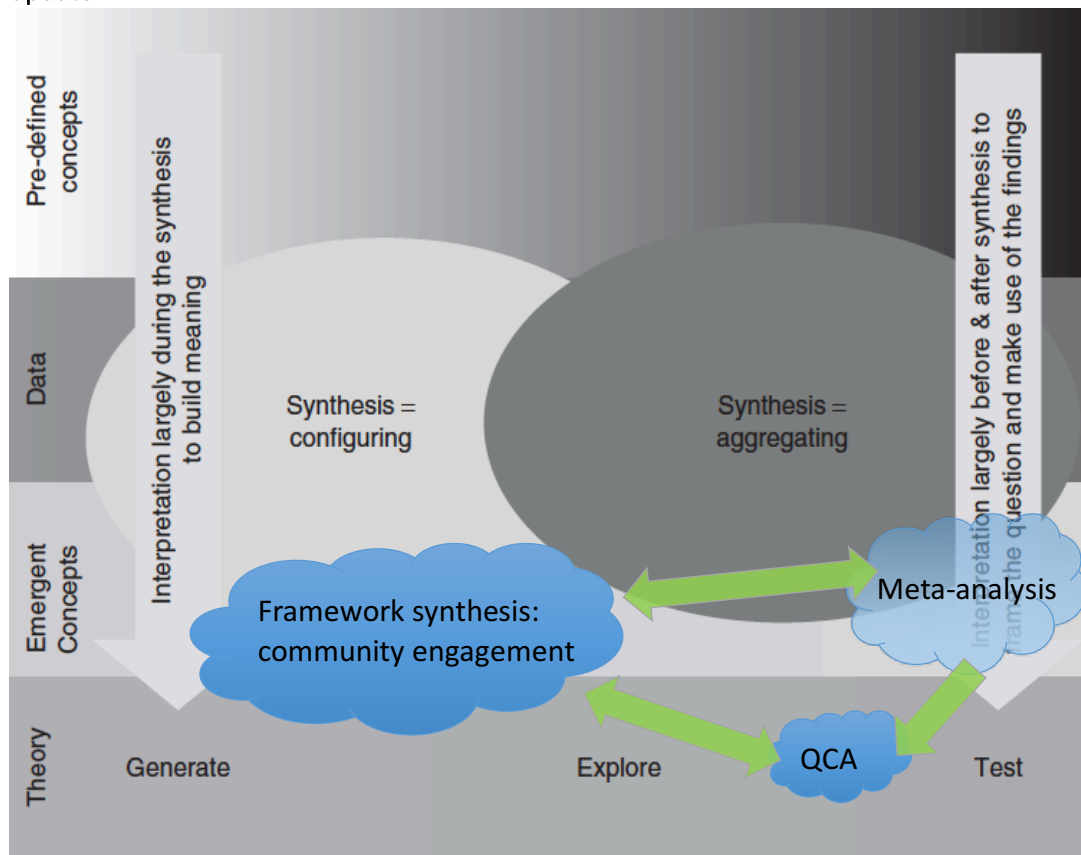
For our research team, this presented a good opportunity to update substantively the systematic review we had conducted previously (5), which I discussed in Chapter 4. This included the chance to further develop and test the conceptual framework we had developed in our earlier review. In particular, we wanted to discover more about the mechanisms of community engagement. These motivations led to the formation of research questions that included examining the processes of community engagement, the type and extent of community engagement, and their relationship to health outcomes.

As in our first community engagement review described in Chapter 3, this was considered a complex topic. There was considerable iteration between the conceptual framework and the data, although perhaps less than in the earlier review as the framework was already established. The studies were diverse in terms of their populations, the types of intervention undertaken and outcomes measured, and the type and extent of community engagement utilised. Each were examined for differences in effects by age, gender, and targeted health issue.

We undertook conceptual development in our first review that provided multiple pre-defined concepts concerning different aspects and domains of community engagement (see Figure 4.6: p.47). As before, synthesis required analysis of both textual and numeric data, where specific located processes would be added into the conceptual framework, descriptively analysed, and explored further in meta-analysis and in qualitative comparative analysis to understand their relationship (if any) with health outcome effect sizes. Newly identified processes were treated as emergent concepts and added into the conceptual framework, and the combinations of processes that were tested during

synthesis. As in the earlier community engagement review, interpretation occurred during synthesis in order to build meaning during the development of the final conceptual framework and the qualitative comparative analysis; however, the meta-analysis was built upon interpretation derived from the final conceptual framework and its results informed the qualitative comparative analysis. Both of these informed the final conceptual framework. The relative placements of the syntheses in this review are illustrated in Figure 5.1 below.

Figure 5.1 **Conceptual model of research synthesis: Community engagement review update**



(From Thomas et al. 2012:181)

Two types of stakeholder engagement were sought: those of a small Advisory Group of academics with relevant expertise; and NICE's Public Health Advisory Committee (PHAC) Community Engagement Guideline Development Group. The first group advised on our methods, including the specific area of community engagement on which to focus synthesis. The second group provided relevant literature, commenting on findings from each stage of the review; and suggested subsequent analyses that would inform guideline development. For example, the PHAC asked us critical questions related to our judgment

about the extent of engagement, arguing that since so few of the studies reported engagement in evaluation this aspect of engagement should be dropped from analysis. We re-ran the analyses according to this suggestion but found that no difference in effects.

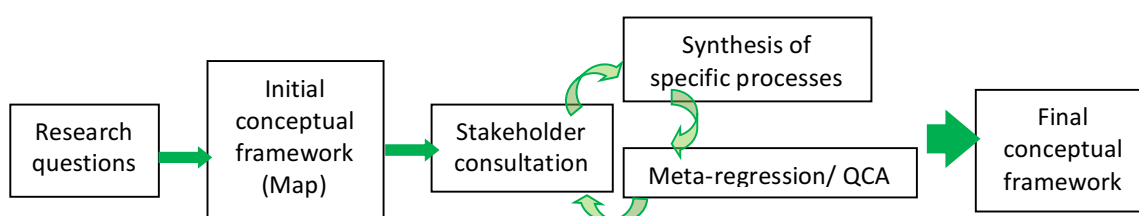
Although not explicitly stated at the time, I approached this review from a realist stance, i.e., that reality can be known and understood through shared experiences. However, the iterative nature of examining data, relating it to the conceptual framework, and looking for patterns in processes or types of community engagement meant that some interpretive work was also needed; thus, my stance was more critical realist.

5.2 Approach

Review design

This review, undertaken in two stages, aimed to understand from the most current literature whether community engagement interventions were effective, for whom, and under what circumstances. The first stage of the review was a mapping exercise, in which framework synthesis was explicitly utilised to understand how newly published studies varied with respect to the extent of engagement, the health topics and the populations under study, across age groups, gender-specific studies, disadvantaged groups, and health outcomes/effects. This stage is described more fully elsewhere (74). The second stage of the review, described in this chapter, examined specific processes of community engagement to see how they were related to both the extent of community engagement across a project and to the health outcomes/effects experienced. This is illustrated in Figure 5.2 below.

Figure 5.2 **Flow diagram of community engagement review update**

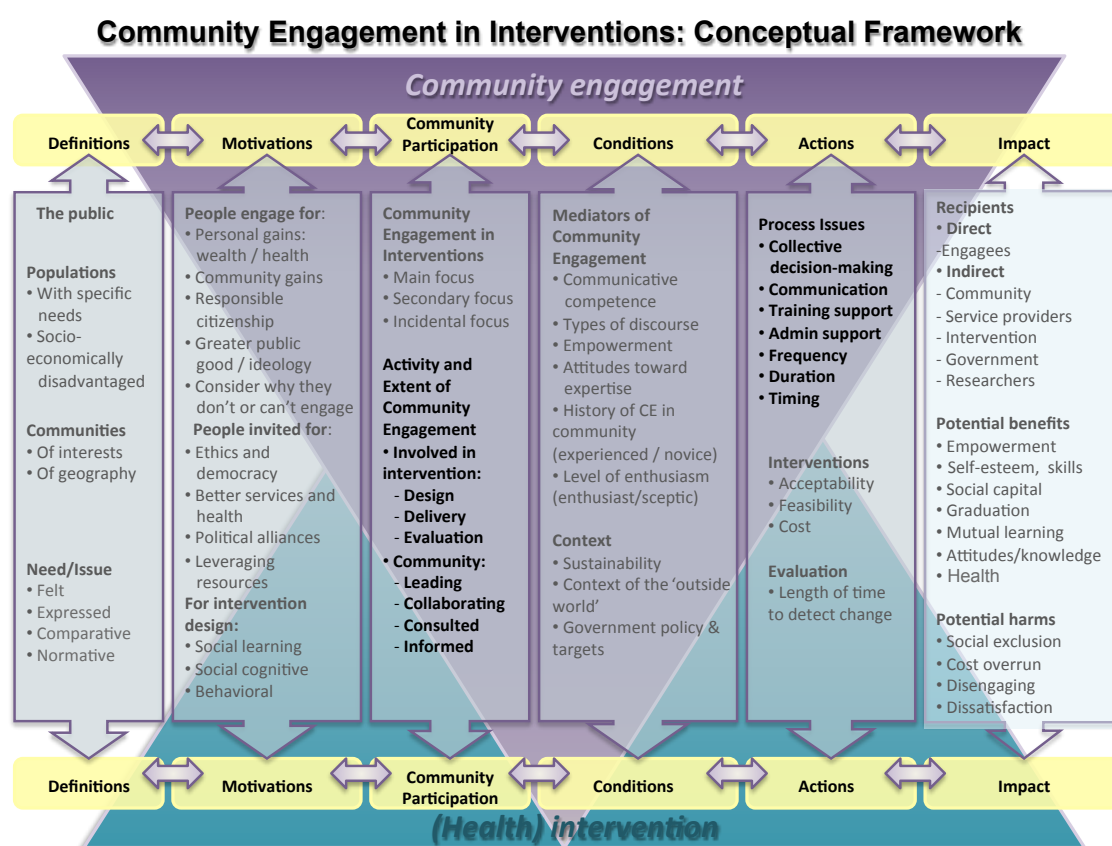


Framework selection

This framework synthesis began by framing the characteristics of community engagement: in particular, the extent of engagement (leading or collaborating versus consulted or

informed across design, delivery and evaluation of an intervention) and modifiable processes of community engagement (bi-directional communication, collective decision making, training support, admin support, time for relationship development, frequency duration and timing of meetings). These originated from the conceptual framework developed in the original community engagement review (5). New processes were added to the framework as they were identified. The initial conceptual framework is shown in Figure 5.3 below, with characteristics of interest for this review highlighted.

Figure 5.3 Initial conceptual framework: specific aspects to be tested



(From O'Mara-Eves et al. 2013)

Indexing

Information on processes of community engagement were extracted from any outcome evaluations included in the review which contained process data. This included any process evaluations linked to included outcome evaluations. Process and extent of community engagement data and were coded according to the initial conceptual framework, with any new processes added.

Charting

To interpret the data, each process was examined for differences in age groups, gender or socio-economic disadvantage. Next, we looked for relationships between processes and effect sizes. Thematic development proved difficult, as study authors often did not go beyond stating a process. In considering the relationship between the extent of engagement (i.e. design, delivery and evaluation) and effects, we hypothesised two potential mechanisms of engagement: (i) the *number* of processes involved; and (ii) the extent of engagement *throughout* an intervention that influenced health outcomes.

Mapping and interpretation

Putting these into a meta-regression showed that there were modest relationships both with processes and extent of engagement. In addition, the extent of engagement and health behaviour outcomes (in longitudinal studies) were found to be associated. Results from the meta-analysis suggested that while a higher number of community engagement processes were modestly associated with statistically significantly larger effect sizes, no one individual process could be attributed with this effect in the meta-analyses. The extent of engagement was interrogated further in later qualitative comparative analyses, which suggested that a low extent of engagement across an intervention's design, delivery and evaluation was most often related to studies showing smaller effects. QCA also suggested that community engagement that included lay delivery as one strategy tended to be aligned with higher effect sizes. Further, those that provided lay delivery across all age groups, rather than focusing on just one group (e.g. young people) were found in studies with higher effect sizes.

Critical assessment of the included studies and their 'fit' with the conceptual framework was undertaken, using the EPPI-Centre risk of bias tool for trials (5). All results were included in the synthesis and sensitivity analyses conducted for different levels of study quality.

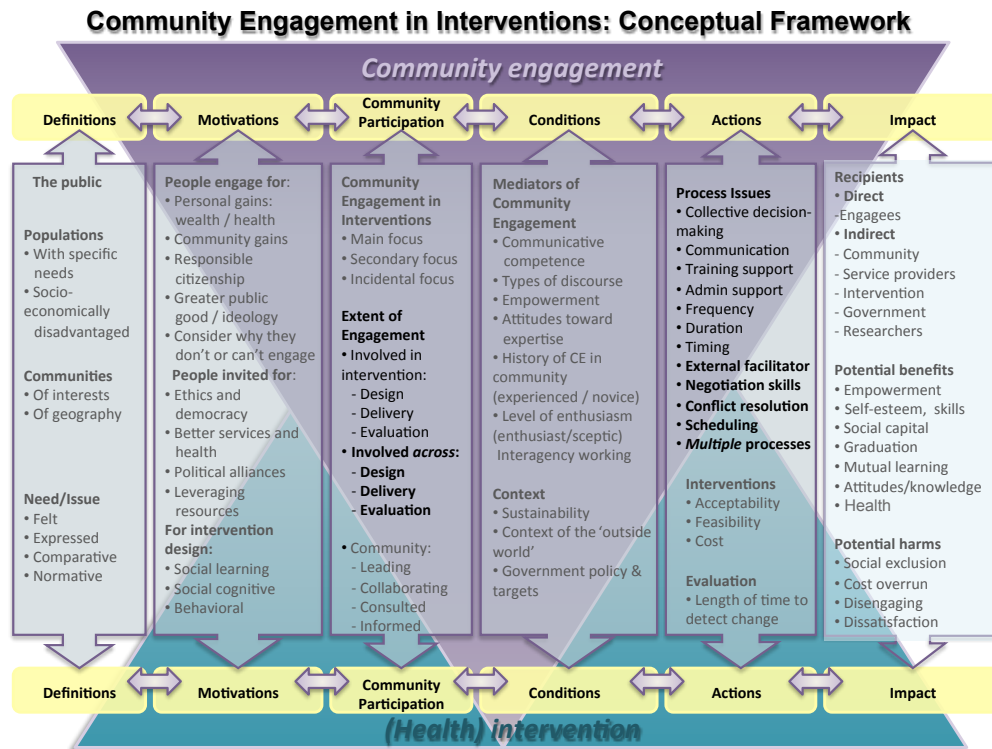
Dissonance assessment was not consciously undertaken at the time. In preparing this chapter, however, one characteristic ('inter-agency working') was identified as a mediator of community engagement rather than a process and was subsequently moved within the framework.

Gap analysis (i.e. examining differences between the *a priori* framework and the new conceptual framework) was undertaken in that we were able to identify some characteristics in the literature that weren't in the existing framework. Further, gaps were identified in complete reporting of the characteristics as well as in the lack of reported evaluation on those characteristics. For example, authors might note that a characteristic such as training was present, but provided no data evaluating the impact of its implementation on health outcomes.

Qualitative sensitivity analysis was undertaken: studies were examined during synthesis to see whether studies of lower quality reported more, fewer, or particular characteristics; however, they did not differ from higher quality studies in this aspect. Reflections were made on the contribution that new characteristics made to the framework in terms of frequency. This reflection did not extend to the contributions made to the 'thickness' of the model (i.e. the range of emergent concepts) (48). Differences in the extent of engagement and the processes used were examined with respect to populations, topic, age and sex.

In summary, the meta-analysis and QCA syntheses informed the framework in that they indicated that a higher number of processes were seen more often in studies with a higher extent of engagement across design, delivery and evaluation; and this was associated with studies measuring more effective behavioural outcomes, particularly where targeted lay delivery was employed. These findings altered the initial conceptual framework, resulting in the final conceptual framework illustrated in Figure 5.4.

Figure 5.4 Final conceptual framework: Community engagement review update



5.3 Discussion

Similar to our first community engagement review, this systematic review was innovative in that it utilised the existing conceptual framework of community engagement in order to *derive and test* hypotheses about the mechanisms of community engagement, specifically about (1) the modifiable processes of community engagement associated with larger effects; and (2) the relationship between sustained community engagement across the design, delivery and evaluation of an intervention.

Specific hypothesised processes did not show a relationship with outcome effect sizes. This may be because processes were poorly and inconsistently described in the studies, and none of the included studies actually evaluated the processes. However, it is interesting to note that effect sizes were higher where more processes were employed and where there was more engagement across design, delivery and evaluation. These may have been examining the same concept, but only improved reporting in process evaluations will allow rigorous testing of this hypothesis. The utility of this framework lies not only in facilitating the analysis of community engagement and public health, but also in providing an initial

conceptual framework for investigating other overlapping worlds (75). It has also served as a useful adjunct in further developing community engagement theory (76).

CHAPTER 6. Stakeholder consultation to frame and prioritise theory

6.1 Context of the review

The Department of Health's Blood Safety and Supply policy team requested a systematic review to inform understanding about the relationships between extrahepatic conditions and hepatitis C. The review was requested in response to UK Parliamentary debate about iatrogenically infected hepatitis C sufferers who had subsequently been diagnosed with chronic health problems occurring outside the liver (i.e. extrahepatic conditions) (77).

This review was undertaken to assess the evidence of a causal relationship between hepatitis C virus and extrahepatic conditions and the impact of hepatitis C virus (HCV) upon health-related quality of life of people in the UK. Specifically, the review aimed to understand how chronic hepatitis C virus (HCV) infection is associated with health-related quality of life; which extrahepatic conditions are associated with chronic HCV; and any moderating or mediating factors that might explain differences in the strength of the relationship between extrahepatic conditions and health-related quality of life.

This project presented two methodological opportunities for our research team. The first was to develop methods to critically assess and synthesise evidence of causation arising from non-experimental studies. The second was to develop methods of integrating this evidence with that which evaluates health-related quality of life. My own epistemological stance was decidedly realist at the beginning of this review. I saw this as a logical exercise in creating a list of extrahepatic conditions using a previously existing framework. However, the need to prioritise which conditions were most important in relation to quality of life required a more interpretive stance. As the review progressed, we were challenged to rethink what each of us – the researchers, the policy team, and the advocacy groups – meant by 'important' in relation to our research question.

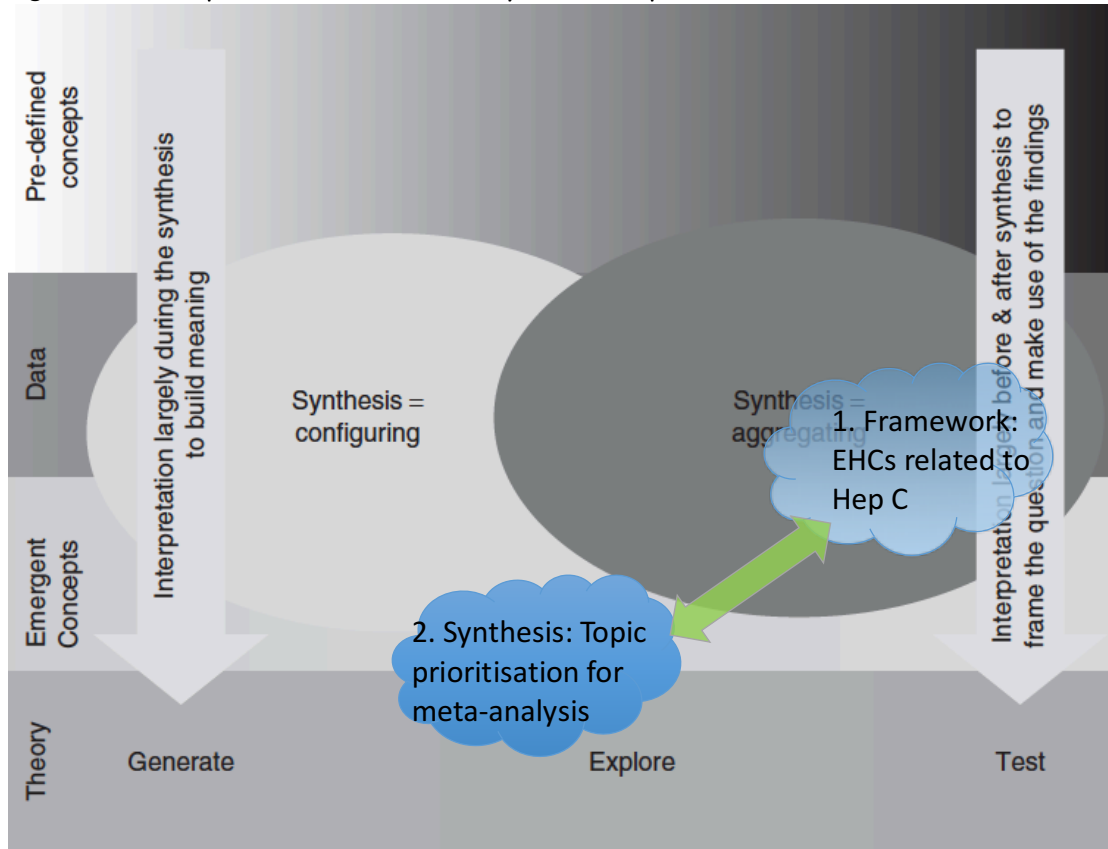
Data in this review were constituted from two sources: (i) research studies of mostly survey, observational or case-control design that examined associations between HCV and various extrahepatic conditions; and (ii) perspectives of scientific advisory and public advocacy groups about what constituted an important extrahepatic condition, and why. Based on discussions with advocacy group members, quality of life in extrahepatic

conditions could be considered a complex topic because (i) there are interactions/moderating effects between extrahepatic conditions and their diagnosis, treatment, social circumstances; (ii) multiple health and non-health outcomes such as quality of life, employment, marital/family circumstances; and (iii) extrahepatic conditions and quality of life experience change over time as the disease progresses or recedes.

The data informing this review contained a large set of clearly pre-defined concepts: the extrahepatic conditions. The data were discrete and, as diagnosed conditions, required no interpretation. The emergent concepts (other extrahepatic conditions) were combined. We were exploring theory around the 'importance' of extrahepatic conditions in relation to people's quality of life by examining the ways that different sources of evidence (research literature, stakeholders) understood and valued what was 'important' in extrahepatic conditions, HCV and quality of life.

The framework began with an *a priori* approach, in which an initial conceptual framework of extrahepatic conditions was created from existing frameworks identified in two related reviews. Conditions identified in research literature located through systematic searching were coded against the initial conceptual framework, with new conditions were added as they were identified. The framework was ordered according to the number of publications on each condition. To determine which conditions were of highest priority, we then tested this framework against stakeholders' perspectives of important extrahepatic conditions, adding new conditions as stakeholder groups identified them. The framework was then subject to emergent interpretation, in that we iteratively reorganised our understanding of what was 'important' by taking into consideration the perspectives offered by each source of evidence: each time another source (i.e. stakeholder) was consulted, the ordering of 'important' extrahepatic conditions changed to reflect the most balanced representation of sources possible. These aspects of synthesis are illustrated below in Figure 6.1.

Figure 6.1 Conceptual model of research synthesis: Hepatitis C review



(From Thomas et al. 2012:181)

6.2 Approach

The concept being framed was the key extrahepatic conditions influencing HCV patients' quality of life. This framing required multiple iterations between the research literature, stakeholder consultations, and researcher interpretation.

To help direct this complex review, we undertook consultation with various stakeholder groups. A Scientific Advisory Group of key experts was created, consisting of DH policy advisors, epidemiologists, virologists, hepatologists and advocacy group representatives. Their role was to help to identify relevant studies, provide contextual understanding of extrahepatic conditions related to HCV, help focus the review in order to determine the studies to include in further analysis, advise on the research questions driving meta-analysis, and comment on initial findings and the final report.

The Scientific Advisory Group included professionals who were well networked with their peers and advocacy groups, such as The British Liver Trust and the Hepatitis C Trust, who

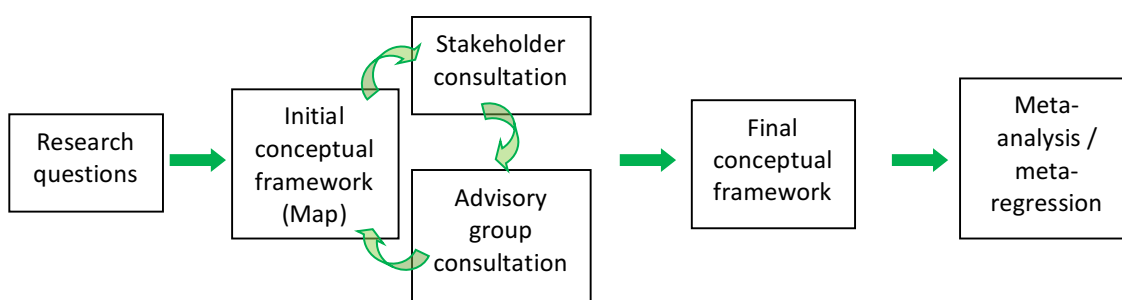
were well connected to people affected by HCV. However, we recognised that other, smaller advocacy groups whose interest in HCV was not their main focus, might also usefully inform the review. To ensure that a wide range of stakeholders had an opportunity to provide their views on extrahepatic conditions and quality of life, organisations such as the Haemophilia Society, Tainted Blood, The Contaminated Blood Campaign and the Manor House Group were invited to provide their views in individual meetings. Their views informed decisions about which extrahepatic conditions to focus on during synthesis.

At the charting stage of the review, the results of this framework were presented to the Scientific Advisory Group and discussed with Advocacy Groups. In each consultation, the views of stakeholders were sought about the most 'important' extrahepatic conditions that impact on HCV patients' quality of life. The feedback from these consultations (particularly the Advocacy Groups) indicated a different set of conditions than those that were most frequently researched. The resulting 'framework' of most frequently researched extrahepatic conditions was presented to Scientific Advisory Committee and Advocacy Groups, and their discussions about what constituted the most 'important' extrahepatic conditions framed the synthesis.

Review design

This was a two-stage systematic review, in which a map of research was undertaken first, in order to understand the breadth and scope of research in the area. An agreed set of studies was subsequently synthesised in-depth. The systematic review process followed the EPPI-Centre's standard stages of searching/scoping, inclusion/exclusion screening, coding/data extraction of study characteristics, risk of bias assessment, outcome extraction and meta-analysis. An initial conceptual framework of extrahepatic conditions, identified in previously existing systematic reviews, was used. Framework synthesis was not described as the method of analysis; however, consideration of the review's framework selection, indexing of studies, charting of conditions and mapping and interpretation of stakeholder views against published research literature indicate its use. This is illustrated in Figure 6.2 below.

Figure 6.2 Flow diagram of Hepatitis C review



Framework selection

This systematic review sought to understand the relationship between extrahepatic conditions and HCV, and to understand the relationship between those same extrahepatic conditions and quality of life in people living with HCV. The initial framework selected was a listing of commonly occurring extrahepatic conditions that was amalgamated from two related reviews on HCV infection that incorporated information on extrahepatic conditions (78, 79). This list identified 43 initial extrahepatic conditions that had been shown to demonstrate an association with HCV.

Indexing

To populate the framework, included studies were coded according to the extrahepatic condition under study. Each extrahepatic condition studied was compared to the framework and new conditions added. The resulting framework of most frequently researched extrahepatic conditions was presented to Scientific Advisory Group and Advocacy Groups. Their discussions about what constituted the most 'important' extrahepatic conditions added to the framework.

Charting

In order to chart the data, information from each source of evidence (research literature, Scientific Advisory Group, Advocacy Groups) were compared and reordered. In total, 94 new extrahepatic conditions were identified in the research and added to the 43 conditions in the initial conceptual framework, which were then ordered by the number of publications on each condition. This initial framework of aggregated conditions associated with HCV provided one way of considering 'important' conditions, i.e. by the amount of research activity generated. For ease of illustration, Table 6.1 below shows the 'top ten' most frequently researched extrahepatic conditions.

Table 6.1 Sources of evidence: Top ten most frequently researched conditions

Extrahepatic Condition	Located Research Publications (N)
Diabetes	244
Non-Hodgkin's lymphoma	162
Depression	148
Mixed cryoglobulinaemia	138
Impaired cognition	113
Insulin resistance	106
Lichen planus	98
Fatigue	66
Hypertension	63
Membranoproliferative glomerulonephritis	62

Consultations with the Scientific Advisory Committee identified somewhat concordantly those most frequently researched extrahepatic conditions, as illustrated in Table 6.2 below.

Table 6.2 Sources of evidence: Most frequently researched and noted by Scientific Advisory Committee

Extrahepatic Condition	Highlighted by Advisory Group
Diabetes	1
Non-Hodgkin's lymphoma	1
Mixed cryoglobulinaemia	1
Insulin resistance	1
Depression	0
Impaired cognition	0
Lichen planus	0
Fatigue	0
Hypertension	0
Membranoproliferative glomerulonephritis	0

Diabetes, non-Hodgkin's lymphoma, mixed cryoglobulinaemia and insulin resistance were conditions noted by Advisory Group members as 'important' in that they were most prominently researched. However, individual consultations with four Advocacy Groups provided different results. Advocacy group members tended to describe extrahepatic conditions more in terms of symptoms or problems rather than diagnoses, e.g. 'heart problems', 'impaired cognition'. Further, the conditions they identified were less often those most frequently researched, as shown below in Table 6.3.

Table 6.3 Sources of evidence: Conditions identified by Advocacy Groups

Extrahepatic Condition	Prioritised by Advocacy Groups (N)
Depression	4
Anxiety	4
Pain	4
Fatigue	4
Impaired cognition	3
Thyroid problems	2
Headaches	2
Brittle teeth	2
Diabetes	1
Circulation problems	1
Renal conditions	1
Cerebral problems	1
Osteopenia	1
Lung disease	1
Gallstone problems	1
Vitamin D deficiency	1
Irritable bowel syndrome	1
Prostate problems	1
Breast cancer	1
Bladder problems	1

Mapping and interpretation

The different sources of evidence needed to be considered in light of the research objectives at this stage. The research evidence showed over one hundred extrahepatic conditions potentially related to HCV: too many to be synthesised in the time available. To further consider which conditions were ‘important’, we consulted key stakeholders to determine their views on which conditions should be prioritised and configured these with those examined in the research literature, thus constituting the interpretive stage of the framework synthesis. We determined priority topics in light of the research questions, the research literature and the perspectives of stakeholders.

Studies included in meta-analysis were considered critically and the quality of the studies assessed in relation to the framework. All included studies looking at the relationship between an extrahepatic condition and HCV were assessed for risk of bias using previously developed tools. Risk of bias ratings were summed into an overall risk rating. All included studies were included in meta-analysis, regardless of risk of bias rating. Sensitivity analyses were undertaken exploring the impact of study quality on effect sizes.

Qualitative sensitivity analysis was undertaken in that the views of the Scientific Advisory Group, the Advocacy Groups and the research literature were examined separately, and then in comparison, to understand their contribution to important extrahepatic conditions. The framework identified gaps suggesting further research, by highlighting a mismatch between stakeholders' views about what constituted important extrahepatic conditions (EHCs) and why, and what had been most commonly researched. Stakeholders did not identify many extrahepatic conditions as important on the initial framework, and also identified new conditions. Configuration of stakeholder views, accumulated research studies, and assessments of quality and gaps in theory resulted in the final conceptual framework shown in Table 6.4 below. The first five topics in bold below were those selected for further synthesis; however due to time constraints only the first four were synthesised.

Table 6.4 Final conceptual framework HCV-related extrahepatic conditions and quality of life

Extrahepatic Condition	Prioritised by Advocacy Groups (N)	Located Research Publications (N)	Discussed by Scientific Advisory Group
Depression	4	148*	0
Anxiety	4	42	0
Pain	4	31	0
Fatigue	4	66*	0
Impaired cognition	3	113*	0
Thyroid problems	2	55	0
Headaches	2	3	0
Brittle teeth	2	0	0
Diabetes	1	244*	1
Circulation problems	1	163	0
Renal conditions	1	145	0
Cerebral problems	1	27	0
Osteopaenia	1	23	0
Lung disease	1	22	0
Gallstone problems	1	12	0
Vitamin D deficiency	1	6	0
Irritable bowel syndrome	1	5	0
Prostate problems	1	1	0
Breast cancer	1	1	0
Bladder problems	1	1	0
Non-Hodgkin's lymphoma	0	162*	1
Mixed cryoglobulinaemia	0	138*	1
Insulin resistance	0	106*	1
Lichen planus	0	98*	0

** – most frequently appearing in the research literature*

6.3 Discussion

This review was innovative in its use of framework synthesis to show transparently how stakeholder consultations provided additional sources of evidence to frame important extrahepatic conditions. The iterative knowledge exchange and researcher reflections undertaken throughout this review allowed us to interpret which conditions were important to quality of life by balancing the weight of research evidence, stakeholder views, and policy needs.

CHAPTER 7. Discussion

7.1 Introduction

In order to address the research questions posed in this thesis, my systematic reviews submitted for consideration (hereafter referred to as ‘thesis reviews’) were compared to those identified in the systematic review of framework synthesis described in Chapter 2, with respect to the contexts in which they were undertaken; their epistemological approach; their methods of framing; and the gaps assessed between and within study quality and their conceptual frameworks. Constant comparative synthesis methods revealed that these concepts interacted throughout the review process, shaping its progress. Findings from other reviews and reports (Chapter 2) demonstrated the use of framework synthesis in what tended to be reviews focused on patients’ perspectives of medical illness, undertaken within a particular discipline and social context; for example examining treatment burden in stroke (40), or factors affecting patient participation in chronic obstructive pulmonary disease programmes (40, 45). Here, initial theories were translated into a framework and coding tool for easy, consistent, efficient collection and analysis of data by teams who may have little or no experience of the method: in effect, the theories interpret the data. Thesis reviews, in contrast, are problem-focused reviews, which inevitably cross boundaries between contexts, methods and disciplines. Topics focused on broader issues (e.g. community engagement in health care) experienced across social contexts. Here, the framework guides the collection and analysis of multiple types of data in a manner that is both coherent and systematic, building on (i) methods used in framework analysis and synthesis elsewhere, and (ii) stakeholder involvement. The theoretical frameworks interpret the data arising from located studies, but data also develops and tests theory. Thesis reviews are compared to the wider literature referenced in Chapter 2 in more detail below, with respect to the context, epistemology, methods of framing, and gaps analysed.

7.2 Context

Systematic review research questions and their methodology are influenced by the specific contexts in which the review is undertaken (4, 80, 81). Reviews were first examined for

their context: their aims, the level at which issues were conceptualised and disciplinary focus; and the extent of stakeholder engagement.

Aims, disciplinary focus and level of issue

The aims, issues and disciplinary focus were more complex in thesis reviews than in other literature. Other 'applied' reviews generally aimed to understand research on stakeholder views of health care services, which were focused on a single dimension about medical illness within a single discipline; for example, barriers and facilitators to disabled children's physical therapy or cancer pain management (41, 42). Thesis reviews, in contrast, appear to focus on multiple dimensions of a *problem or outcome*, examining barriers and facilitators to walking and cycling and their fit with evaluated interventions; conceptualisations of community engagement and factors influencing its effectiveness; and 'important' extrahepatic conditions influencing quality of life. The breadth of each of these issues went beyond a single disciplinary focus.

Broad public health issues such as these are thought to operate at multiple levels: those of the individual, family, community and society (82). In contrast to other reviews which examined individual and family level influences (e.g. patients' and caregivers' perspectives of cancer pain management), thesis reviews looked beyond these to more broadly examine issues at the community and society levels; for example, by applying perspectives of walking and cycling from individuals and families to community-level interventions. Examination of thesis reviews suggests that they extend beyond previously conducted framework synthesis reviews by examining problem-focused issues that span disciplines at multiple levels of influence.

Stakeholder engagement

Systematic reviews are often commissioned because of specific gaps in policy knowledge and it has been suggested that they should rely on interactions between policy makers, practitioners and researchers who can best make use of the knowledge gained (83-85). In addition to these stakeholders, thesis reviews also involved members of the public from key interest groups. Engaging stakeholders in the systematic review process is important. It has been argued that involving members of the public, professionals, policymakers and researchers can reduce waste in research effort, bring a more

democratic and broader range of perspectives to bear on the complex issues under study, and ultimately produce a more relevant and useful piece of research (50, 86). Research commissioners in the UK require stakeholder involvement throughout the research process (87); and increasingly, examples of stakeholder engagement in systematic reviews are emerging (88, 89). The findings from this thesis suggest that previous reviews utilising framework synthesis have rarely reported involving public, professional or academic stakeholders in the review process. In contrast, each of the thesis reviews has reported engaging a wide range of stakeholders. Their role ranged from informing the development and interpretation of the developing conceptual framework to interpreting the results of testing those frameworks. Because thesis reviews sought to develop, explore and test theory, stakeholder involvement was considered crucial in order to ensure that the theory was grounded in the context of those who had most experience of the issues – practitioners, researchers, and most importantly, affected members of the public. While similar roles have been described in other types of systematic review (88, 89), this appears to be an innovation in systematic reviews using framework synthesis. This type of broad engagement points to models of knowledge transfer in which systematic reviews seek to ‘enlighten’ as well as ‘inform’ end users of the review (90). With one exception (22), most other reviews provided little evidence of stakeholder involvement in the research process. In comparison, each of the thesis reviews engaged stakeholders from multiple disciplines and members of the public throughout the review to help set priorities, cross disciplinary boundaries, and provide context.

Thesis reviews thus spanned disciplinary boundaries in order to examine broad, problem-focused issues that operated at multiple levels. Policy-relevant systematic reviews such as these are influenced by a variety of interconnected factors, including the diversity of the policy environment, the motivations of funders and academics, the engagement between them, and the structures and procedures that support knowledge synthesis and exchange (91). Such broad engagement has been described as a type of ‘boundary work’ in which lay, professional and policy stakeholders with diverse expertise and disparate agendas can work together in a transdisciplinary way to shape the research process in a way that meets each stakeholder’s needs (92-94). Such transdisciplinary approaches encourage researchers to look beyond discipline-bound ways of working in order to find methods that will solve wider problems (95, 96).

7.3 Epistemological approach

The epistemological approach of thesis reviews and other reports of framework synthesis were compared by examining: the researcher's stance; their stated approach; whether approaches were *a priori* or emergent; how theory was developed; and the extent of iteration.

Researcher stance, stated approach and *a priori* or emergent methods

Researcher stance can influence the approach taken in reviews, as researchers may have a preference for working with data that arises from a positivist or interpretive view of knowledge (49). *A priori* (deductive) research approaches aim to understand the world from a logical positivist, realist, or post-positivist point of view (97). Here, a theory aiming to explain (usually causal) relationships between phenomena is already in existence, and new data are applied to test the theory. These most often utilise quantitative data, designs and analysis methods to aggregate data (18). Emergent (inductive) research approaches arise from an interpretivist or constructivist point of view. These aim to explore the meaning of phenomena by using data to contextualise findings and develop theory (18, 27, 97). While these two approaches have been frequently polarized as separate paradigms (98), it has also been suggested that in practice researchers may view these approaches along a continuum in which a combination of *a priori* and emergent approaches are used to both aggregate and configure findings (4, 18).

Where the other literature included in this thesis described an epistemological approach, it was called a realist method. These other reviews and reports most often described framework synthesis as an *a priori* method of applying an existing theory in order to interpret qualitative data. In contrast, thesis reviews applied an evolving approach where higher order synthesis of concepts was undertaken, such as the development of themes of walking and cycling (Chapter 3), the identification of 'important' extrahepatic conditions affecting quality of life in hepatitis C (Chapter 6) and where mechanisms of community engagement interventions and processes were derived from a conceptual framework (Chapter 4, 5). My stance as a researcher was realist throughout, although my understanding of critical realism and its embodiment in research synthesis has evolved. Initially, I recognized that within certain structures (e.g. walking and cycling behaviours), some mechanisms (such as the weather, or fears of safety) might influence whether an

event (e.g. walking or cycling to school) was observed or not. However, even then I recognized that this behaviour was more complex than simply a few mechanisms and that the actualization of these would be influenced by a range of conditions, for example, the current UK policies on active transport. With each subsequent review, my stance evolved to appreciate the complexity of the issues under study, particularly in relation to understanding the impact of issues on people's lived experiences and the utility of critical realism as a foundation to co-create knowledge with those who are most directly affected by the policies under study: the members of the public. In this way, I came to recognise and appreciate the emancipatory potential of critical realism (53) with each successive review project.

This developing critical realist stance is evidenced in the final framework synthesis, in that a range of mechanisms were identified that, to differing degrees, can be considered to affect the methods of framework synthesis potentially used in a systematic review. This understanding of the nature of framework synthesis may change over time, as new publications are identified which use and reflect on it, demonstrating my understanding and appreciation of the fallibility of this knowledge.

Theory development and extent of iteration

Research synthesis offers an opportunity to generate, explore or test theory by combining findings from multiple primary studies (4, 50). Which of these is chosen depends on reasons for undertaking a systematic review, its research question and ultimate purpose (18). Where consensus exists on pre-defined theoretical concepts and the review will provide answers which support or refute a hypothesis, a review is *testing* theory. Systematic reviews can also *explore* theory when they seek to provide more detailed findings within a set of answers, for example in determining whether an intervention is more effective for one group versus another. Here, the pre-defined concepts may not be clearly defined at the start of the review. Systematic reviews can also *generate* theory where not all concepts are defined and their inter-relationships are still to be understood (12). Some methods are more obviously at one end of this continuum: for example, meta-ethnography and meta-narrative approaches lie at the theory-generating pole, while meta-analysis is more aligned with theory testing. Critical interpretive synthesis and realist synthesis could be placed as approaches that explore theory, in that the former seeks to build theory inductively but then interpret it in the light of research evidence (99); whilst

the latter aims to make intervention programme theory explicit, then looks for empirical evidence to populate this framework (100):p.v).

Other reports of framework synthesis most often described framework synthesis generally as a method to 'develop' theory; however, reports varied in describing whether reviews aimed to specifically generate, explore or test theory. Some reported framework synthesis was useful to explore theory, i.e. where a relevant conceptual framework already existed (43). Others suggested that framework synthesis was useful for undertaking thematic analysis without generating new theory (50). In contrast, thesis reviews employed framework synthesis to generate, explore and test theory. For example, a conceptual framework of factors influencing walking and cycling was generated and then tested by comparing the fit between qualitative and quantitative synthesis findings (Chapter 3). A conceptual framework of community engagement and potential mechanisms was generated and explored through a descriptive map of trials (Chapter 4); then the hypothesised mechanisms were generated and tested with findings from a meta-analysis of trials. In a subsequent review, different mechanisms (i.e. processes and levels) of community engagement were explored and their influence tested on outcome effect sizes (Chapter 5). Theories were generated about 'important' extrahepatic conditions by comparing stakeholder consultation with research evidence, in order to subsequently test the causal relationship between important extrahepatic conditions and hepatitis C (Chapter 6).

It has been suggested that the degree of iteration differs between types of research synthesis, with *a priori* approaches using less iteration than more emergent ones (18, 24, 49). Iteration can occur: between data and the framework, as new concepts identified by the data are added into a conceptual framework (24); between data/concepts and the research process itself, where researchers revise their decisions and search iteratively in order to locate and test theory (47, 49, 101); between study quality and review findings, to check the impact of studies' methodology or relevance on the findings of a review (60); and between the conceptual framework and the researchers, as discussions arise to understand patterns in the data which inform the conceptual framework (100). Other reports of framework synthesis and thesis reviews consistently identified the use of iteration between data and conceptual frameworks, and qualitative sensitivity analyses were routinely undertaken. Thesis reviews reported additional iteration between

researchers, stakeholders and the developing conceptual framework, suggesting another type of iteration not previously described.

Comparison of the other reports and reviews of framework synthesis and thesis reviews suggest that the epistemological approach is realist and *a priori* in nature, generally testing theory. However, thesis reviews also incorporated emergent approaches and more iteration to generate and explore theory.

7.4 Methods of framing

Next, thesis reviews were compared to other reviews and reports with respect to the nature of the data, the review design, the rationale for selecting framework synthesis, and the ways in which framework was applied.

Nature of the data and review design

Other reviews synthesised one type of research data arising from qualitative research of participants' experiences or perspectives or of policy setting, most often utilising one type of data: qualitative. In contrast, thesis reviews sought to understand multiple issues within active travel, community engagement and quality of life in extrahepatic conditions associated with hepatitis C infection, asking multiple questions and integrating evidence from a mixture of quantitative and qualitative data arising from research of participants' experiences, effectiveness studies, process evaluations, theoretical discussion papers and from stakeholder consultations. These constituted 'mixed methods' or 'mixed sources' reviews (4, 18). Different mixed methods synthesis typologies have been developed, following methods described in mixed methods primary synthesis (97, 102, 103). The walking and cycling review (Chapter 3) is most similar to reviews employing thematic synthesis methods because it developed theory to explain intervention effects, using comparative methods (12, 97). In contrast, the qualitative studies results concerning community engagement (Chapter 4, 5) were transformed into a conceptual framework which was utilised to generate and then test mechanisms of community engagement. This 'convergent qualitative synthesis' is most similar to reviews undertaking critical interpretive synthesis, meta-narrative synthesis (where concepts are being established), and realist synthesis (97). The review in Chapter 6 converted analysis of research on extrahepatic conditions to variables conceptualizing the most 'important' conditions (i.e.

by the frequency of each condition appearing in the literature); and these were compared to the conditions identified by key stakeholders as having a significant influence on their quality of life. This 'convergent quantitative synthesis' is most similar to Bayesian approaches, in which data is transformed to inform further statistical analysis (97, 101, 104).

It is important to note that, while this mixed studies typology is helpful in clarifying the distinction between whether findings from qualitative studies informed subsequent quantitative syntheses or vice versa, it does not clearly communicate the iterative interplay of synthesis, interpretation and new hypothesis generation using findings from qualitative and quantitative syntheses described earlier (see Chapter 4). For example, the convergent qualitative synthesis design used in the community engagement reviews to develop theory resulted in the development of mechanisms, which were further tested empirically using meta-regression (Chapter 4, 5) and qualitative comparative analysis (Chapter 5). These in turn generated new hypotheses for future exploration and testing.

Framework selection and application

Other reviews and reports of framework synthesis suggested it was chosen for pragmatic reasons, to facilitate rapid examination of evidence within small and inexperienced research teams, using a realist perspective. While some suggested it was useful for creating structures that facilitated stakeholder discussion and for aggregating and configuring data in mixed methods designs, these uses were rarely borne out. Thesis reviews chose framework synthesis for all of these reasons; however, it was also selected for its ability to include wider stakeholder perspectives, organise broader and more complicated issues and because it was flexible enough to support types of synthesis beyond thematic synthesis.

Thesis reviews consistently constructed an initial conceptual framework, rather than selecting an established conceptual framework or theory. Instead, a series of characteristics known to influence a phenomenon were utilised as a series of logic models to generate theory. This methodology was similar to, and indeed influenced by, the previous systematic review undertaken by colleagues in which the conceptual framework was built on historical researcher knowledge and logic models gleaned from scoping searches at the background stage of the review (22). In each of the other 'applied' reviews,

frameworks were selected from the same discipline from within which the research question was set. Most initial conceptual frameworks were lone theories, although two used more than one, a method later suggested to be appropriate (47). Theory development is important in providing and evaluating health care, as it is thought to shape that way researchers and health care professionals collect and interpret evidence, thus influencing how care is understood (30, 105). The use of logic models, as a type of 'low-level theory' (30) can be useful to help scope a review, define and conduct a review, and make it relevant to policy and practice by acting as a communication tool (33, 106). Its use in clarifying the conceptual thinking that occurs during the systematic review process has been noted by others (32); and the findings from this thesis help to make more explicit the interactions between framework development, stakeholder feedback and research team conceptualisations.

Across my thesis reviews, framework synthesis was used at the indexing stage to order, categorise and represent a totality of research by its existing and emergent characteristics. Findings from thesis reviews suggest that the framework was ordered and re-ordered throughout the review process, as the emerging conceptual framework was tested against the research evidence and in light of stakeholder consultations. The conceptual framework acted here as an 'iterative logic model' to help structure and communicate ideas with stakeholders (107):p.31).

In addition, while other reports of framework synthesis indicate thematic analysis of indexed data, thesis reviews used this plus a variety of synthesis methods to build higher order knowledge. For example, while the walking and cycling review (Chapter 3) utilised thematic synthesis to derive overarching themes in the first framework synthesis, in the second framework synthesis a constant comparative method was used to examine the fit between implications derived from views studies and interventions (108). This same approach was used in the hepatitis C review (Chapter 6), in which the fit between theory of 'important' conditions affecting quality of life and research evidence were grounded in stakeholder views about what constituted important conditions. The fit between theory and interventions was assessed in other thesis reviews using meta-regression (Chapter 4, 5) and qualitative comparative analysis (Chapter 5) to synthesise higher order knowledge derived from data. The variation in these methods to synthesise data from different

sources of evidence, and the different purposes to which the findings need to speak, suggests that framework synthesis can employ more methods beyond thematic synthesis.

In summary, thesis reviews extended beyond other uses and descriptions of framework synthesis identified in the literature: (1) by utilising it in mixed methods synthesis; (2) by using it to build as well as explore and test theory, necessarily using a variety of synthesis methods; and (3) in consultation with stakeholders to iteratively develop the conceptual framework throughout the research process.

7.5 Assessment of matches and mismatches

It is important to assess matches and mismatches as part of the review process, in order to ensure the findings are robust but also to build new knowledge by understanding where information is lacking (4, 97). Thesis reviews were compared to other literature in terms of how they assessed gaps between theory, data and findings.

Study quality, dissonance and gap analysis

Other reports of framework synthesis suggested that assessing study quality is important, in that all but one of the applied reviews conducted quality assessment of included studies (but used different tools). Authors also consistently advocated consideration of the impact of gaps in study quality on review findings. Similarly, each thesis review conducted quality assessment of included studies using previously developed tools, and considered the impact of study quality on findings. Dissonance assessment was undertaken to consider 'missing or uncomfortable' data (20, 48); however this was not undertaken or described consistently across other reviews and reports. Researchers generally agree the importance of critically assessing the quality of studies suited to the research questions under study, however little consensus exists on the methods by which this should be undertaken. It has been suggested: that using a prescriptive tool to analyse all qualitative studies in the same manner can be too restrictive; that the relevance of the study to the review question is as important as the rigour with which it has been conducted; and that the 'quality' of included studies will emerge naturally as part of critical synthesis (60, 109). Others assert that every qualitative study should at least report its methods of sampling, data collection and analysis in order to allow readers to understand the study's context (110, 111). Further, the use of

a tool may encourage reviewers to be more explicit about their judgments of quality (66). These are challenges experienced across qualitative synthesis methods (112).

Matches and mismatches in context

Thesis reviews, similar to other reviews, examined the gaps between study quality and findings, and between theory and data. However, the mixed methods design used in thesis reviews also allowed the examination of matches and mismatches between different types of study. The review of walking and cycling (Chapter 3) tested the themes derived in the first framework synthesis of participants' views against evaluated interventions in a constant comparative synthesis. The first community engagement review (Chapter 4) iteratively developed the initial conceptual framework using research data and stakeholder consultations, and then derived potential mechanisms that were tested against effect sizes extracted from evaluated interventions using meta-analysis methods. The second community engagement review (Chapter 5) synthesised hypothesised interactions between processes and extent of engagement, tested them in meta-regression and explored these further in qualitative comparative analysis to further develop theory. In addition, stakeholder involvement brought additional perspectives to the research process, allowing us to examine the fit between theories and lived experiences. Here, gaps in the framework of extrahepatic conditions identified in the Hepatitis C review (Chapter 6) were interpreted using the findings from stakeholder consultations in a constant comparative methodology. Thus, it is argued that the method of framework synthesis was utilised to critically assess gaps between theory and multiple types of data.

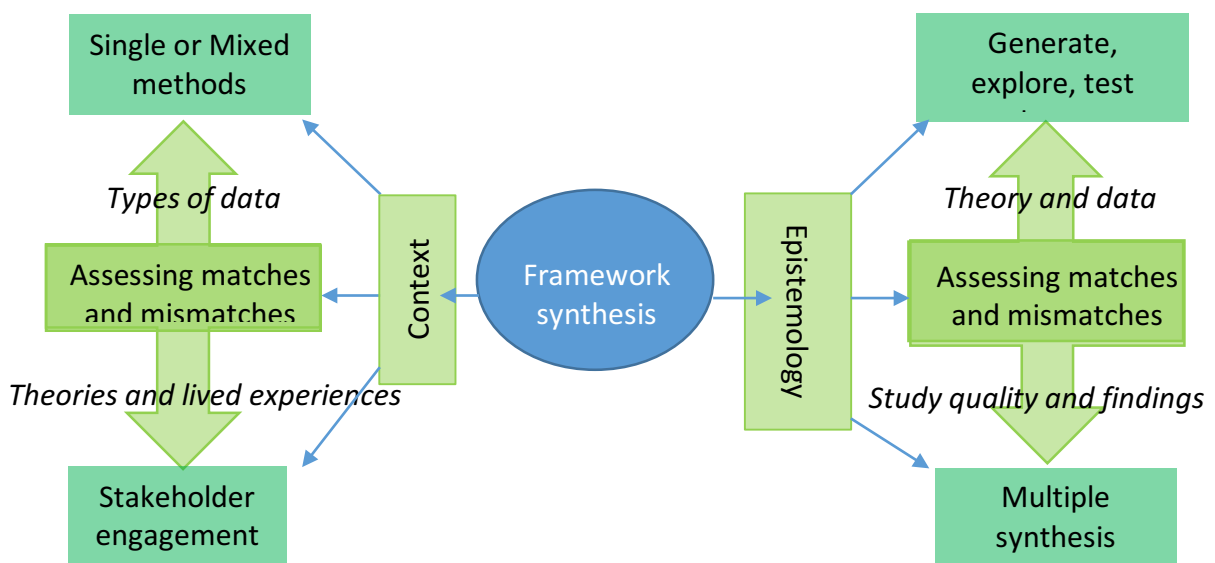
7.6 Review strengths

Comparison of the literature on framework synthesis and my thesis reviews indicate that most other reviews sought to synthesise one type of research evidence (i.e. qualitative research of participants' experiences or case studies) in order to 'frame' those perspectives against a previously existing theory. In contrast, my thesis reviews built conceptual frameworks from the views of lay people (i.e. their expressed barriers and facilitators); discussed the frameworks and the research they frame with review stakeholders; and applied multiple research synthesis methods (e.g. QCA, meta-regression, constant comparative analysis) within them. These innovations made it easier to address stakeholder priorities and to draw on conclusions and recommendations relevant to

stakeholders. As such, these are transdisciplinary systematic reviews designed for decision making because they are framed not by academic disciplines or methodologies but by concepts that transcend them.

To understand the ways in which framework synthesis methods vary across past systematic reviews and compare these with my thesis reviews, the conditions, epistemology, methods of framing and gaps assessments were examined. An interaction between these was identified, which is illustrated in Figure 7.1 below.

Figure 7.1 Final thesis conceptual framework



Framework synthesis methods are derived from context, which influences (i) whether ‘single’ or ‘mixed methods’ designs are used, and matches and mismatches between types of data are assessed; and (ii) how matches and mismatches between theories and lived experiences are considered through stakeholder engagement throughout the review process. Framework synthesis methods are also derived from epistemology, which influences (iii) whether theory is generated, explored or tested and gaps between theory and data assessed; and (iv) the synthesis methods used where gaps between study quality and findings are examined.

7.7 Limitations

Four potential limitations were identified to the methodological approach of this thesis. First, each stage of the review process was undertaken by only one researcher. This has

been identified as a potential source of bias in that findings may be influenced by not having a second reviewer 'double check' each stage of the review (4). However, searches were informed by an information scientist, a second researcher was utilised in screening, and synthesis and interpretation was planned in discussion with supervisors and research colleagues. These steps will help to strengthen the rigour of the review and confidence in its findings.

Second, it is possible that some previous examples of framework synthesis were not located because to be included, authors had to refer to it specifically as a method. Indeed, one of my thesis reviews (Chapter 4) was not conceptualised as a framework synthesis in the report. However, on subsequent discussion with co-authors it was agreed that the overarching method to pull together all of the separate syntheses was framework synthesis (113-115).

Third, using framework synthesis to undertake a systematic review about framework synthesis appears on the surface to be somewhat overcomplicated. However, synthesis should be guided by a review's purpose and context (12). This review aimed to use an identified (although nascent) conceptual framework in order to build a more comprehensive understanding of the use and utility of framework synthesis. It was undertaken to further develop my knowledge of the method and to provide a structure in which my publications can be placed within the body of knowledge on framework synthesis methods. These aims and objectives suggested a need for a realist approach that allowed for some interpretation of data in order to configure a new understanding of the method – something that framework synthesis is well-suited to address.

Finally, no Advisory Group informed this work. However, as this is a methodological review and not for policy decision making, it could be argued that the thesis will benefit from the perspectives generously provided by my two PhD supervisors and by external examiners, all of whom bring considerable expertise. Framework synthesis methodology will further develop and benefit as a result of this consideration.

CHAPTER 8. Conclusions

Several research questions were posed in this thesis, to understand my use of framework synthesis in relation to how others have used or discussed framework synthesis. These are addressed below.

8.1 Addressing the thesis research questions

1. What have my reviews added to current knowledge of the method?

Framework synthesis has been described by others as a method which uses a predominantly *a priori* approach to aggregate and configure data, most often by applying data to a previously identified conceptual framework. Other systematic reviews and reports of framework synthesis revealed that the method also varies around how and when a previously existing conceptual framework is identified and applied, the extent of iteration and testing that occurs, and the context in which a review is undertaken. Comparison of thesis reviews to these other described uses of framework synthesis revealed that thesis reviews have extended the use of framework synthesis in three ways: (1) to demonstrate its use in mixed studies reviews incorporating data from research on participant's experiences with that of empirical data arising from trials or studies of association; (2) to highlight its utility in structuring information from multiple sources of evidence that also include stakeholder engagement, which in turn shapes the resulting framework synthesis product; and (3) in illustrating its use of a range of methods beyond thematic synthesis (such as meta-regression and QCA) to build knowledge. These innovations allow for more transparent, relevant and appropriate integration of theory into the systematic review process, thus filling a need to address more comprehensively the complex issues often identified by policy makers while also taking the perspectives and opinions of a variety of stakeholders into account (Oliver, 2015). Ultimately, the main contribution of this thesis has been to distinguish two key approaches to framework synthesis: the approach that constructs a framework (often in discussion with stakeholders) to accommodate research from across academic disciplines and/or policy sectors; and the 'best-fit' approach that borrows a framework from a related area to initiate synthesis within a narrower disciplinary or policy scope. This constitutes a new understanding of framework synthesis methods.

2. How do these methods compare with other methods of framework synthesis used or discussed?

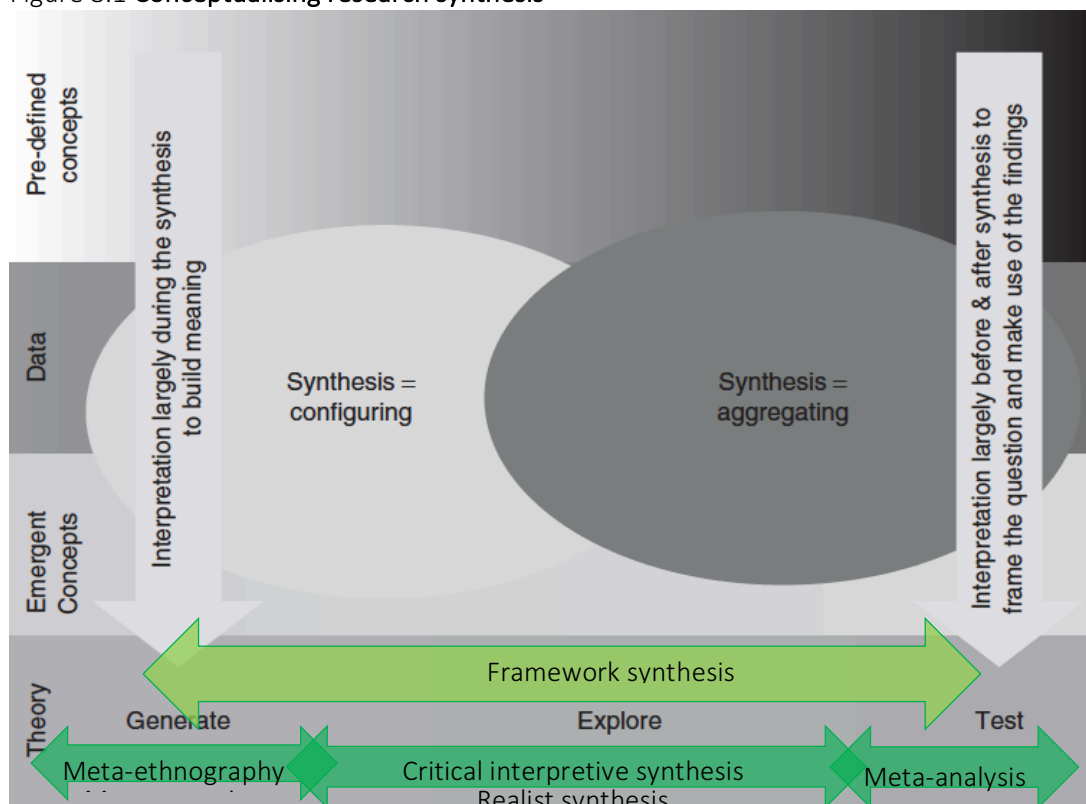
Most other reviews that have utilised framework synthesis methods sought to synthesise one type of research evidence in order to 'frame' those perspectives against a previously existing theory. In contrast, my thesis reviews synthesised multiple types of research or other evidence separately and then brought these findings together into a third synthesis,

using different types of synthesis methods. These ‘mixed studies’ and ‘mixed sources’ reviews seek to address broad and complex policy questions that go beyond questions of ‘is it effective?’ and ‘what are people’s experiences?’ to include broader issues of how something might work and under what circumstances. In addition, framework synthesis provided an environment conducive to stakeholder engagement because of its use as both a structure to organise theory and as a communication tool to facilitate discussion with a range of stakeholders.

3. Where is framework synthesis situated within research synthesis methods?

Within the ever-widening spectrum of research synthesis methods, framework synthesis appears to be a realist method that is positioned ‘in the middle’ in terms of its *a priori* and emergent reasoning and iteration to generate, explore and test theory using heterogeneous data. In general, research synthesis methods can be thought of as lying along a continuum of theory generation, exploration or testing, as illustrated in Figure 8.1 below.

Figure 8.1 Conceptualising research synthesis



(From Thomas et al. 2012:181)

All of these approaches overlap in terms of the extent to which theory is generated, explored or tested. However, framework synthesis spans the entire continuum, which methods such as meta-ethnography, meta-narrative and meta-analysis cannot do. It can be argued that framework synthesis allows enough interpretive creativity for it to move between critical realism (where knowledge of reality is mediated by people's perceptions and beliefs) and scientific realism (in which knowledge of the world ever more closely approaches an external 'truth'); and findings from some included reports suggest that this may be dependent on the extent to which stakeholders' views are employed either as participants in the research process or as the phenomenon under study. This suggests it is a useful alternative to realist synthesis and critical interpretive synthesis (99, 100). However, it is important to clarify which stakeholders will be involved in the review, and in what ways, prior to selecting a method of synthesis. It appears that differences exist between thesis reviews and realist synthesis regarding the involvement of public stakeholders, as it has been suggested that the role of public stakeholders has not been clarified in realist review methods guidance (117).

4. What problems are addressed by framework synthesis and not by other methods?

Framework synthesis may help to address some problems not currently addressed by other methods because it can help make reporting of methods more transparent. Readers can see clearly the original conceptual framework, the themes that are derived from the data populating that framework, and how the themes are translated back into the original framework to further develop that theory. The lack of reporting clear methods of synthesis has been flagged as a challenge in qualitative research synthesis (110).

Framework synthesis also offers a method of combining data that allows both aggregation and configuration of findings fit to purpose and context of the review. This suggests a 'selective eclecticism' approach that could address calls for a paradigm shift in research methods (18, 56, 109).

The results of this review suggest that the philosophical stance of a study should fit the research question, which itself is derived from both the context in which the phenomenon under study is occurring and from the researcher's own preferences (18, 118). Further, clearly communicating these will help readers to understand and interpret the review's methods and findings.

8.2 Implications for future use of framework synthesis

Some recommendations for future research were identified. In order to allow readers to assess the fit between the synthesis method and the end use of the review, future use of framework synthesis should include descriptions of researchers' stance and/or the ontological and epistemological stance underpinning the review. To reduce the potential for selection bias, researchers could provide more detail on how the initial framework was identified and adapted for use (if this occurred), as noted by others (47, 48). The initial and final conceptual frameworks should be included for readers to assess how the data changes the theory. Finally, it will benefit understanding about stakeholder contributions to the review process and to specific stages of framework synthesis if there is more careful documentation of stakeholders' role in shaping the conceptual framework as it develops through the review.

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APPENDIX 1: Candidate publications for consideration

Chapter 3: Brunton G, Oliver S, Oliver K, Lorenc T (2006) *A synthesis of research addressing children's, young people's and parents' views of walking and cycling for transport*. London: EPPI-Centre, Social Science Research Unit.

URL: <http://eppi.ioe.ac.uk/cms/Default.aspx?tabid=942>

Chapter 4: O'Mara-Eves A, Brunton G, McDaid D, Oliver S, Kavanagh J, Jamal F, Matosevic T, Harden A, Thomas J (2013) Community engagement to reduce inequalities in health: a systematic review, meta-analysis and economic analysis. *Public Health Research*. 1(4): 1-548.

URL: http://www.journalslibrary.nihr.ac.uk/_data/assets/pdf_file/0006/94281/FullReport-phr01040.pdf

Chapter 5: Brunton G, Caird J, Stokes G, Stansfield C, Kneale D, Richardson M, Thomas J (2015) *Community engagement for health via coalitions, collaborations and partnerships: a systematic review*. London: EPPI-Centre, Social Science Research Unit.

URL: <http://www.nice.org.uk/guidance/GID-PHG79/documents/community-engagement-update-review-2-community-engagement-2015-brunton2>

Chapter 6: Brunton G, Caird J, Sutcliffe K, Rees R, Stokes G, Stansfield C, Llewellyn A, Simmonds M, Thomas J (2014) *Depression, anxiety, pain and quality of life in people living with chronic hepatitis C: a systematic review and meta-analysis*. London: EPPI-Centre, Social Science Research Unit.

URL: <http://eppi.ioe.ac.uk/cms/Default.aspx?tabid=3468>

APPENDIX 2: Background systematic review (Chapter 2) methods

Search strategy

In order to identify research that would inform the review's research question and scope, key papers were identified from colleagues, those used in other systematic reviews, and a Google Scholar search using the phrase 'framework synthesis'. These scoping papers were used to derive the research questions developed for the PhD proposal. Full searching using free text terms were undertaken in ASSIA, PsycInfo, PubMed, and Web of Science using the search string illustrated in Appendix 3; reference lists of included studies were searched; and key experts contacted. Thesaurus specific terms for 'framework synthesis' were not found and thus not used. Searches were conducted from database inception up to 1 January 2015, with a search update undertaken to include references up to 31 December 2015. No language limits were set on the searches. These methods were undertaken as recommended in current research synthesis guidance (36, 119).

In the *Framework Identification* stage, I sought a suitable conceptual framework. Background scoping of literature often identifies a relevant conceptual framework to which included studies can be applied/assessed. Where no such framework is identified through background scoping, it can be developed from previous research, stakeholder input and researcher knowledge (22).

No explicit conceptual framework of framework synthesis in systematic reviewing was located, although several papers describing the method were located (20, 47, 48). A conceptual framework of key issues was derived from discussion papers identified in the scoping searches undertaken for writing the PhD proposal, from the researcher's knowledge of qualitative research methods, and from standard data extraction tools developed over several years of systematic reviewing at the EPPI-Centre. These sources indicated that framework synthesis was likely to vary according to:

- the aims of the review in which it was used;
- the stages of framework synthesis used;
- where in the systematic review process the framework was applied;
- the reflections authors make on its use, relevance or applicability;

- the strengths and/or limitations of the method;
- what authors infer from using the method; and
- whether authors claimed a *deductive* or inductive use of framework synthesis.

These dimensions became the initial conceptual framework¹ for my systematic review.

Next, the ***Indexing stage*** was undertaken as part of the inclusion screening and data extraction processes of the review. Studies were sorted to determine their relevance to the review questions and to identify their main characteristics.

Inclusion screening

To be consistent and transparent in assessing all retrieved references, each was screened using *a priori* exclusion criteria based on the research questions. First, in order to be included in synthesis, reports screened on title and abstract had to:

- specifically indicate the use of ‘framework synthesis’; and
- be relevant to health care, health policy or public health.

The full text reports of references meeting both of these criteria were retrieved and screened again. At this stage, reports had to also:

- describe framework synthesis as a synthesis method; and
- provide textual descriptive data from authors that reflected on its use in some way.

Data extraction

Systematically coding each report according to its characteristics allows patterns to emerge from the data, enabling subsequent comparison across reports. In order to consistently examine each report, data were extracted using a coding tool found in Appendix 4, developed from the review’s research questions and the conceptual framework described above. Any characteristics not addressed by these codes were added to the framework and applied to all included reports.

Synthesis

The synthesis stage of a systematic review can build new knowledge by ‘going beyond’ the original studies (12). This corresponds to two stages of framework synthesis: ***Charting*** and

¹ Note that the eventual framework that develops and is described in the Charting and Mapping section is referred to as the *framework synthesis* – the new conceptual framework.

Mapping and Interpretation: the former stage analyses the main characteristics of each research paper, by grouping characteristics into categories and deriving themes directly from those data. During the latter stage, derived themes are considered in light of the original research questions. At this stage, findings from the review can be presented in various formats (e.g. forest plots, tables, figures, or narratives) for ease of reader interpretation.

Part of synthesis also refers to the quality assessment process. Systematic reviews should assess their included studies, assisting researchers to consider the quality and relevance of the approach undertaken (3). Because the current work is an ‘overview of reviews’, and because both substantive systematic reviews and discursive methodological articles were included, the findings relevant to this review were expected to be drawn from discursive pieces of data that reflected and interpreted the use of framework synthesis. Thus, traditional quality assessment of included studies normally undertaken in a systematic review of health interventions was not considered appropriate. Instead, critical consideration of each argument was undertaken instead.

Interpretation and communication

The findings arising from synthesis were next considered in relation to the original research questions, the wider research literature and the context in which the review was originally undertaken. This process of presenting findings corresponds to the **Mapping and Interpretation** stage of framework synthesis.

Quality assurance

Quality assurance measures undertaken in systematic reviews ensure that the review is consistently and transparently conducted, to reduce the likelihood of bias (i.e. drawing incorrect conclusions from studies included in the review that contain systematic selection or reporting errors) (4, 34). As part of quality assurance, an Information Scientist informed the development of literature searches. The PhD candidate and a second researcher undertook screening, with any references identified by either researcher included for full-text retrieval. I conducted coding and synthesis, in consultation with both PhD co-supervisors and EPPI-Centre colleagues. Data integrity and analysis were maintained with the use of EPPI-Reviewer software (12).

APPENDIX 3: Search strategy terms

Search undertaken: Database inception to 31 December 2015

Sources:

PubMed

“framework synthesis” in TI,AB

“framework analysis” in TI,AB AND PT=review

ASSIA

“framework synthesis” in TI,AB

“framework analysis” in TI,AB AND PT=review

Sociological Abstracts

“framework synthesis” in TI,AB

“framework analysis” in TI,AB AND PT=review

Web of Science

“framework synthesis” in TI,TS (topic)

“framework analysis” in TI,TS

PsycInfo

“framework synthesis” in TI,AB

“framework analysis” in TI,AB AND PT=review

APPENDIX 4: Coding tool: Review of other literature on framework synthesis

Aims of report described?

Enter as stated by authors in the information box

Did authors provide a reason why they used framework synthesis?

Did authors explain why framework synthesis was selected over other methods?

Can their use be compared to Ritchie and Spencer's method?

i.e., did they list the steps as described by Ritchie and Spencer or refer to them explicitly?

To which stage(s) of SR was the framework synthesis applied?

How did framework synthesis get used? Did authors apply framework synthesis to one stage of the review (e.g. analysis stage only), or did they describe its use in setting the research question, community engagement, etc.?

Were the strengths of the method discussed?

Did authors reflect on the strengths of using framework synthesis?

Did authors discuss any limitations of the method?

Did the authors reflect on any limitations in using framework synthesis methods?

Were any future research methods work recommended by authors?

Did authors describe any methods work that needs to be addressed as a result of conducting this study?

APPENDIX 5. Excluded studies: Reasons for exclusion

1. Title and Abstract Screening

EXCLUDE 1: Not about Framework Synthesis

GUIDANCE: *Has to be about framework synthesis methods (stated by the authors as such), as used in systematic review health research synthesis. The ref might be about framework analysis methods (primary study synthesis) but would be excluded here.*

N=73 citations

Alajarín Mateo, Bonillo Baltasar, Vidal Angel, and Bautista Delia. (2007). Bis(heterocumulenes) derived from the 1,4-diphenyl-1,3-butadiyne framework. Synthesis of three new classes of axially chiral biheteroaryls.. *The Journal of organic chemistry*, 73(1), pp.291-4.

Baranov A I I, Isaeva A A A, Kloo L, and Popovkin B A A. (2003). New metal-rich sulfides Ni(6)SnS(2) and Ni(9)Sn(2)S(2) with a 2D metal framework: synthesis, crystal structure, and bonding.. *Inorganic chemistry*, 42(21), pp.6667-72.

Beverley Catherine A, Bath Peter A, and Barber Rosemary. (2011). Health and social care information for visually-impaired people. *ASLIB PROCEEDINGS*, 63(2-3), pp.256-274.

Bolton Ruth, and Saxena-Iyer Shruti. (2009). *Interactive services: A framework, synthesis and research directions*.. [online] Netherlands. Available at: <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=psyc6&NEWS=N&AN=2009-21638-010>.

Boote Jonathan, Baird Wendy, and Sutton Anthea. (2012). Involving the public in systematic reviews: a narrative review of organizational approaches and eight case examples. *Journal of comparative effectiveness research*, 1(5), pp.409-420.

Boote Jonathan, Baird Wendy, and Sutton Anthea. (2011). Public involvement in the systematic review process in health and social care: a narrative review of case examples. *Health Policy*, 102(2), pp.105-116.

Brunton G, Oliver S, Oliver K, and Lorenc T. (2006). *A synthesis of research addressing children's, young people's and parents' views of walking and cycling for transport*. London: EPPI-Centre, Social Science Research Unit, pp.1-131. Available at: <http://eppi.ioe.ac.uk/cms/Default.aspx?tabid=942>.

Budinsky RA, Schrenk D, Simon T, Van den Berg M, Reichard JF, Silkworth JB, Aylward LL, Brix A, Gasiewicz T, Kaminski N, Perdew G, Starr TB, Walker NJ, and Rowlands JC. (2014). Mode of action and dose-response framework analysis for receptor-mediated toxicity: The aryl hydrocarbon receptor as a case study.. *Critical reviews in toxicology*, 44(1), pp.83-119.

Bunck David N N, and Dichtel William R R. (2012). Mixed linker strategies for organic framework functionalization.. *Chemistry (Weinheim an der Bergstrasse, and Germany)*, 19(3), pp.818-27.

Carbonell Carlos, Stylianou Kyriakos C C, Hernando Jordi, Evangelio Emi, Barnett Sarah A A, Nettikadan Saju, Imaz Inhar, and Maspoch Daniel. (2013). Femtolitre chemistry assisted by microfluidic pen lithography.. *Nature communications*, 4, pp.2173.

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GUIDANCE: Exclude here if reference is reporting on framework synthesis used in an area other than health care, health policy or public health.

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2. Full Report Screening

EXCLUDE 1: Not about framework synthesis

Guidance: *Has to be about framework synthesis methods (stated by the authors as such), as used in systematic review health research synthesis. The ref might be about framework analysis methods (primary study synthesis) but would be excluded here.*

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GUIDANCE: Exclude here if report is a non-systematic review or is a protocol for a review

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EXCLUDE 3: No data on methods

GUIDANCE: Exclude here if reference only reports on a phenomenon that utilised framework synthesis without discussing/analysing the use of framework synthesis. Main focus is on substantive findings rather than methodological findings.

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EXCLUDE 4: Duplicate

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APPENDIX 6. Master and Linked Reports

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LINKED 2: Lawton Rebecca, McEachan Rosemary R. C, Giles Sally J, Sirriyeh Reema, Watt Ian S, and Wright John. (2011). Development of an evidence-based framework of factors contributing to patient safety incidents in hospital settings: a systematic review. *BMJ Quality & Safety*, , pp.bmjqs-2011-000443.

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