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[Qualitative Protocol]

A meta-ethnography of how children and young people with chronic non-cancer pain and their families experience and understand their condition, pain services, and treatments

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ABSTRACT

Objectives

This is a protocol for a Cochrane Review (qualitative). The objectives are as follows:

1. To synthesise qualitative studies that examine the experiences and perceptions of children with chronic pain and their families regarding chronic pain, treatments, and services to inform the design and delivery of health and social care services, interventions, and future research.
2. To explore whether our review findings help to explain the results of Cochrane Reviews of intervention effects of treatments for children's chronic pain.
3. To determine if programme theories and outcomes of interventions match children and their families' views of desired treatments and outcomes.
4. To use our findings to help inform the selection and design of patient-reported outcome measures for use in chronic pain studies and interventions and care provision to children and their families.

Review questions

1. How do children with chronic non-cancer pain and their families conceptualise chronic pain?
2. How do children with chronic non-cancer pain and their families live with chronic pain?
3. What do children with chronic non-cancer pain and their families think of how health and social care services respond to and manage their own/their child's chronic pain?
4. What do children with chronic non-cancer pain and their families conceptualise as 'good' chronic pain management, and what do they want to achieve from chronic pain management interventions and services?

BACKGROUND

Description of the topic

Chronic pain in childhood is widespread: around 20% to 35% of children and young people worldwide are estimated to have chronic pain (King 2011). Frequent severe chronic pain of all types affects 8% of children (Perquin 2000); for approximately 5% of children, chronic pain results in moderate or severe disability (Huguet 2008). The 11th revision of the International Classification of Diseases (ICD-11) defines chronic pain as "pain that persists or recurs for more than 3 months" (Treede 2019). Chronic pain is recognised as a condition in its own right, but it is also a key feature of health conditions, such as inflammatory bowel disease and juvenile idiopathic arthritis. Most importantly, chronic pain is a significant clinical issue in children and adolescents with important negative consequences beyond the child's physical health, also affecting social, psychological and cognitive functioning, and quality of life (Merlijn 2006; Roth-Isigkeit 2005). Families are often highly involved in managing a child's chronic pain, playing both mediating and moderating roles (Donnelly 2020; Liossi 2016; Palermo 2014), as well as often experiencing stress and distress in their caring role (Jordan 2007; Jordan 2017; Law 2019; Palermo 2014). This review seeks to: understand how children and young people with chronic non-cancer pain and their families conceptualise and live with chronic pain; explore their views and experiences of health and social care services and treatments in relation to pain management; and investigate what they consider as optimal pain management and what they want to achieve from interventions and services, with a focus on high-income countries. This review will be crucial to inform health and social care, and therefore improve pain management and hence the lives of children and young people.

How the health condition might affect people

Chronic pain has considerable negative impacts on children's health and quality of life; for instance, surveys have shown that the majority of adolescent children with chronic pain experience poorer physical, mental, and social health (Gauntlett-Gilbert 2007), and perceive themselves to be behind their peers in many aspects of their development (Eccleston 2008). Chronic pain adversely affects social and family relationships (Jordan 2017); results in poorer school attendance (Logan 2008); and is associated with increased use of healthcare services and medication (Scottish Govt 2018). Healthcare costs of chronic pain in adolescents alone have an annual cost of about GBP 4000 million in the UK (Sleed 2005), and USD 19,500 million in the USA (Groenewald 2014). It also costs families to travel to healthcare appointments and to take time off work to care for their child, with some parents giving up work entirely to care for their child (Sleed 2005). Moreover, longitudinal research indicates a high risk of childhood chronic pain continuing into adulthood with further individual, healthcare, and societal costs (Walker 2010).

To our knowledge, there is no single comprehensive theory of children's chronic pain that covers all the aspects of interest in this review (how children and families conceptualise pain, experiences of living with pain and of pain management services, and views of 'good' pain management and services) and that reflects the theoretical stance of the review team. Most of the existing theories have been developed within a specific field, which might narrow our understanding and perspective of how children experience

chronic pain. For instance, psychological theories of children's chronic pain tend to focus only on specific aspects of the pain, such as what causes pain, or they adopt a child development approach to explaining children's understanding of their chronic pain (Carter 2014). More comprehensive theories which are not specific to children's chronic pain but which better reflect our theoretical approach to pain are biopsychosocial theories of chronic illness, which specify the interrelatedness of biological, psychological, and social aspects of illness (Haslam 2021). To our knowledge, there do not appear to be any comprehensive biopsychosocial theories specifically about children's chronic pain; there is at least one which has focused only on clinical assessment and management of children's chronic pain, but not other aspects, such as how children and their families conceptualise and live with chronic pain (Bursch 1998).

There are also programme theories which specify how a complex intervention is thought to work (Noyes 2016a). Existing Cochrane effectiveness reviews have focused on pharmacological treatments of children's chronic pain: antidepressants (Cooper 2017c), non-steroidal anti-inflammatory drugs (Eccleston 2017), antiepileptic drugs (Cooper 2017d), opioids (Cooper 2017b), and paracetamol (Cooper 2017a). These reviews have focused on how the medications work biologically, rather than a broader view of medication use in the 'real world' in terms of how people actually engage with medications. Cochrane Reviews of psychological interventions for children with chronic pain, Fisher 2018, and psychological interventions for their parents have described how a range of psychological interventions, such as behavioural strategies, cognitive strategies, and cognitive-behavioural therapy, are thought to work (Law 2019). However, many interventions in clinical practice are multidisciplinary, combining different kinds of treatments: biological, psychological, and physical (Palermo 2012). It will therefore be important to further develop programme theories. Consequently, there is an opportunity for our review to contribute to theoretical development in this field.

Why is it important to do this review?

Despite the high prevalence and serious impacts of children's chronic pain, current services for managing children's chronic pain are inadequate (CMO 2009; Pain Summit 2012; Palermo 2019). The *Lancet* Commission 'Delivering transformative action on paediatric pain' stated that pain in children is frequently undertreated (Eccleston 2021). There is a lack of evidence from high-quality trials to inform clinical guidelines and thus guide chronic pain management (NICE 2018; Scottish Govt 2018; WHO 2020a), and insufficient knowledge of which outcomes are important to patients and their families to guide design of services and treatments and to inform future research (Cooper 2017b; Cooper 2017c; Cooper 2017d; Eccleston 2017; Fisher 2018). Five Cochrane Reviews on the effectiveness of pharmacological treatments for children's chronic non-cancer pain (antidepressants (Cooper 2017c), non-steroidal anti-inflammatory drugs (Eccleston 2017), antiepileptic drugs (Cooper 2017d), opioids (Cooper 2017b), and paracetamol (Cooper 2017a)) identified a dearth of research to inform pain management, and highlighted the lack of patient-defined outcomes related to pain relief or improvement of function. This indicates an urgent need to identify outcomes of importance to children with chronic pain and their families to inform future trials and effectiveness reviews to guide pain management (Cooper 2017b; Cooper 2017c; Cooper 2017d; Eccleston 2017). Further,

whilst psychological interventions that engage children or parents, or both, improve child outcomes (Fisher 2018; Law 2019), a family-system approach to chronic pain research appears to be lacking despite a call for this over a decade ago (Lewandowski 2007). A review of pharmacological, physical, and psychological therapy intervention effectiveness for the World Health Organization (WHO) found a lack of evidence from high-quality trials (Fisher 2022). With high-quality evidence lacking, children are not receiving evidence-based pain management, which could result in poor short- and long-term outcomes in terms of pain and pain-related disability. Indeed, unaddressed pain in children is a risk factor for continued pain into adulthood (Walker 2010).

To design and deliver services and interventions that meet the needs of children and their families, it is crucial to understand how they experience and understand chronic pain of different kinds, which treatment outcomes are meaningful to them, and their views and experiences of health and social care services in relation to their pain management. Qualitative research is ideally suited to address these urgent and important questions. There is existing relevant qualitative research to inform these issues (e.g. Carter 2012; Jordan 2007; Jordan 2016; Maciver 2010; Neville 2019), but there are no existing or planned qualitative evidence syntheses of this research. We identified only two existing qualitative evidence syntheses which are limited in focus. The two evidence syntheses looked at specific childhood chronic pain populations and topics: living with juvenile idiopathic arthritis, Tong 2012, and adolescent social relationships, Jordan 2017, and did not develop a theory to inform pain management; theory is important to guide the development of complex interventions, for example (MRC 2019). We will therefore conduct a qualitative evidence synthesis using meta-ethnography (Noblit 1988), a methodology suited to developing theory, to investigate the diverse experiences and perceptions of children up to age 18 with chronic non-cancer pain and their families and to generate theory to inform health and social care. This research will enhance our understanding of the experiences, perceptions, and needs of children with chronic pain and their families in order to improve services and treatments, and hence children's health and quality of life. This meta-ethnography aims to: help us better understand how children and families conceptualise and live with chronic non-cancer pain; inform whether a more family-orientated approach to chronic pain management is needed in order to help improve the quality, access, and organisation of health and social care services; and identify child- and family-centred outcomes to help inform the selection and design of patient-reported outcome measures.

The Cochrane Pain, Palliative and Supportive Care (PaPaS) Group (papas.cochrane.org/) has prioritised research into children's chronic pain (Cochrane 2018), and the International Association for the Study of Pain set its global theme for 2019 as "the year against pain in the most vulnerable" - a group which includes children - in order to raise awareness and improve pain assessment and management (IASP 2018). Furthermore, we developed this review with input from children with chronic pain and their families, pain and children's health charities, healthcare professionals, and academic experts who confirmed the importance of our review aims and objectives.

Our review aims to produce robust, novel evidence to inform and support the management of childhood chronic pain, which is important to health and social care services. This review may also

lead to new conceptual insights and theories (which can change healthcare delivery and policy and inform treatments) (France 2019c; Noblit 1988), and indicate gaps in knowledge and hence new directions for chronic pain research (Campbell 2011).

How the review might inform or supplement what is already known in this area

Two review authors (EF, JN) conducted a qualitative evidence synthesis for the WHO (WHO 2020c), in order to inform the revised guidelines for children's chronic pain management (WHO 2020b). The WHO 2020c synthesis took a global perspective on the management of children's chronic pain, with a particular focus on including research conducted in low- and middle-income countries (LMICs), and which incorporated the views and experiences of healthcare professionals, as well as those of children with chronic pain and their families. It focused solely on the views, perceptions, and experiences of the risks, benefits, and acceptability of three types of intervention: pharmacological, psychological, and physical therapies. Our current qualitative evidence synthesis will take a broader perspective on chronic pain management, including how children and their families conceptualise and live with pain, and consider any kind of intervention or service; it will not explore the views of healthcare professionals (which were explored in the WHO synthesis) and will not focus extensively on LMICs. We will compare the findings of our synthesis to those of the WHO 2020c synthesis to help ensure that there is a global focus to meet global decision-makers' needs.

We will extend the findings of existing relevant Cochrane Reviews on the effectiveness of pharmacological interventions (e.g. Cooper 2017a; Cooper 2017b; Cooper 2017c; Cooper 2017d; Eccleston 2017) and psychological interventions (e.g. Fisher 2018; Law 2019) for children's chronic pain by undertaking a stand-alone qualitative evidence synthesis that provides further clarity concerning phenomena of interest that supplement and add to the Cochrane intervention effectiveness reviews. Our meta-ethnography may also direct future effectiveness reviews to address outcomes of importance to children and their families. These are two of the important 'added-value' roles of qualitative evidence synthesis recognised by Cochrane (Noyes 2018a). There has been inadequate use of qualitative research evidence about children and their families' experiences of chronic pain in the form of qualitative evidence syntheses to inform the design of trials and the outcomes they measure, services, and treatments. A more biomedical approach from the clinician's perspective is typically adopted in the Cochrane Reviews on managing children's chronic pain (Cooper 2017a; Cooper 2017b; Cooper 2017c; Cooper 2017d; Eccleston 2017); yet a bio-psycho-social approach is required (Faculty 2015). Qualitative research typically adopts a bio-psycho-social perspective (Pope 2006), and is also well-suited to developing an understanding of the outcomes valued by children and families which could inform future trials and Cochrane Reviews of intervention effectiveness. Meta-ethnography is ideally matched to synthesising qualitative evidence on the complex issues related to children's chronic pain.

Note: we will use the term 'children' to refer to 'children and young people' throughout the protocol.

OBJECTIVES

1. To synthesise qualitative studies that examine the experiences and perceptions of children with chronic pain and their families regarding chronic pain, treatments, and services to inform the design and delivery of health and social care services, interventions, and future research.
2. To explore whether our review findings help to explain the results of Cochrane Reviews of intervention effects of treatments for children's chronic pain.
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Review questions

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4. What do children with chronic non-cancer pain and their families conceptualise as 'good' chronic pain management, and what do they want to achieve from chronic pain management interventions and services?

METHODS

Criteria for considering studies for this review

We developed the aim and review questions using the SPIDER acronym, as follows.

Sample: children, teenagers, or infants with chronic pain, their siblings, brother, sister, parents, mothers, fathers, grandparents, or other family members.

Phenomenon of interest: experience of any type of chronic non-cancer pain: musculoskeletal, migraine, headache, recurrent abdominal pain, juvenile idiopathic arthritis, complex regional pain syndrome (CRPS), fibromyalgia, endometriosis, inflammatory bowel disease, and so on.

Design: interviews, focus groups, case studies, surveys, observation, ethnography.

Evaluation: views, experiences, attitudes, perceptions, beliefs, conceptualisations, feelings, understandings of living with chronic pain and of chronic pain services and treatments.

Research type: qualitative, mixed methods.

Types of studies

We will include qualitative primary research studies of any design (e.g. ethnography, phenomenology, case studies, grounded theory studies) that used qualitative methods for data collection (e.g. focus group discussions, individual interviews, observation,

diaries, document analysis, open-ended survey questions) and data analysis (e.g. thematic analysis, framework analysis, grounded theory). We will exclude studies that have not used qualitative methods for data collection or analysis, or both (e.g. studies that analysed qualitative data quantitatively) and qualitative literature reviews. We will include published and unpublished studies, as well as studies published in any language. We will include mixed-methods studies where it is possible to extract the data that were collected and analysed using qualitative methods.

Topic of interest

We will include studies focusing on the experiences and views of children with chronic pain and their families towards chronic pain, health services, and treatments. 'Child' is defined according to the UN Convention of the Right of a Child (UNCRC) as a person under 18 years of age.

Draft inclusion and exclusion criteria are below. We will decide the final criteria with our Patient and Public Involvement (PPI) group (approximately 8 children and young people with chronic pain and 8 to 12 parents/family members purposefully recruited to be diverse) at the project outset and in line with qualitative evidence synthesis methods; inclusion criteria may also be revised during analysis phases. Revising inclusion criteria is appropriate for meta-ethnography, the purpose of which is to build understanding and theory, rather than produce a definitive conclusion about the effectiveness of an intervention (Ames 2019; Benoot 2016). We will identify and read relevant articles and make decisions in collaboration with our PPI group, to refine inclusion and exclusion criteria in order to ensure that a focused, manageable, and meaningful synthesis can be conducted to answer our research questions. In this way, we will use an iterative process to select texts for synthesis.

Draft inclusion criteria are as follows.

- Published or grey literature, i.e. peer-reviewed journal articles, published reports, book chapters, books, PhD theses.
- Contains qualitative research data on chronic pain, e.g. pain lasting for 12 weeks or more, relevant to the research questions.
- Reports the views of children with chronic pain from 3 months up to age 18 years or their family members (e.g. parents/guardians, grandparents, siblings).
- Uses recognisable qualitative methods of data collection and analysis.
- In any language.

Draft exclusion criteria are as follows.

- Acute pain, i.e. pain lasting for less than 12 weeks, such as that caused by medical procedures.
- Cancer pain.
- Pain in neonates and babies < 3 months old.
- Focuses on end-of-life pain management.
- Non-empirical article, e.g. editorial, commentary, study protocol.
- Findings do not differentiate between participants with acute or chronic pain.
- Findings do not differentiate between adult and child participants.

Search methods for identification of studies

We conducted a scoping search of MEDLINE bibliographic database using search terms similar to those given in [Appendix 1](#) to indicate the volume and suitability of the literature to address our review questions. We retrieved 861 references, 117 (14%) of which met the inclusion criteria; 42% of these eligible publications were conceptually rich. This indicated a reasonable-sized body of literature from which to sample items for synthesis, and that conceptually rich publications (e.g. [Carter 2012](#); [Gaughan 2014](#); [Maciver 2010](#)) suitable for meta-ethnographic synthesis exist to address the review questions (see [Objectives](#)). Based on the scoping search results combined with our experience of running systematic review searches, we anticipate that there will be 8000 to 9000 unique records to screen from across all databases searched.

We will conduct a rigorous search for published and unpublished ('grey' literature) studies via bibliographic databases and forensic searches, as outlined below. We will include grey literature as an

important potential data source for all research questions. While peer review can be a marker of quality, unpublished studies, such as doctoral theses, can offer rich, high-quality data. In a meta-ethnography, lower-quality studies will contribute less in terms of data and conceptual insights and understanding than higher-quality studies to the synthesis findings ([Noblit 1988](#)), regardless of their peer-review status (see the 'Assessing the methodological limitations of included studies' section below). RT is leading the design and conduct of literature searches, assisted by the research fellow. We finalised the literature search strategy in collaboration with our PPI group.

Electronic searches

We plan to search 13 bibliographic databases selected for their good coverage of qualitative research and spectrum of relevant disciplines. See Table 1 below.

Table 1. Bibliographic databases to be searched

Discipline/type of literature	Databases
Health and social care	CINAHL (Cumulative Index to Nursing and Allied Health Literature) Embase Child Development & Adolescent Studies MEDLINE (including MEDLINE in Process and ePub ahead of print) Social Care Online
Psychological	PsycINFO
Sociological	ASSIA (Applied Social Sciences Index & Abstracts) Social Sciences Citation Index
Education	British Education Index
Multidisciplinary	Scopus
Grey literature	HMIC (Health Management Information Consortium database) OpenGrey ETHOS

The database search strategy for MEDLINE is presented in [Appendix 1](#). It combines three key search concepts:

- qualitative study designs;
- population: children and their families;
- phenomenon of interest: chronic pain.

The strategy is informed by existing reviews that represent good practice for identifying the study design, population and/or phenomenon ([Fisher 2018](#); [Scottish Govt 2018](#)), and testing against a set of key papers. There will be no language or date restrictions. We will adapt the MEDLINE strategy to the remaining bibliographic databases listed in Table 1.

Grey literature

We will identify grey literature by searching the following.

- Three bibliographic databases listed in Table 1 (HMIC, OpenGrey, and ETHOS).
- Websites of key organisations representing chronic pain health conditions, as informed by our PPI group. These include: The British Pain Society (www.britishpainsociety.org/), Department of Health (www.gov.uk/government/organisations/department-of-health-and-social-care/), NIHR Journals Library (www.journalslibrary.nihr.ac.uk/#/), the Sickle Cell Society (www.sicklecellsociety.org/), Versus Arthritis

(www.versusarthritis.org/), the International Association for the Study of Pain (www.iasp-pain.org/), CRPS (Complex Regional Pain Syndrome) UK (crps-uk.org/), Fibromyalgia Action UK (www.fmauk.org/), Crohn's & Colitis UK (www.crohnsandcolitis.org.uk/), Reflex Sympathetic Dystrophy Syndrome Association (RSDSA) (rsds.org/), The European League Against Rheumatism (EULAR) network (www.eular.org/index.cfm), European Pain Federation (europeanpainfederation.eu/), Pain Relief Foundation (painreliefoundation.org.uk/), Children's Health Scotland (www.childrenshealthscotland.org/), children's hospitals.

Searching other resources

We will conduct 'forensic' style supplementary searches for published and unpublished research (Harris 2018), including the following.

- Website searches as detailed in the section on grey literature.
- Handsearching the past 24 months of the following key journals relevant to our research questions or qualitative health research, or both.
 - *BMC Pediatrics* (bmcpediatr.biomedcentral.com/)
 - *Clinical Journal of Pain* (journals.lww.com/clinicalpain/pages/default.aspx)
 - *European Journal of Pain* (europeanpainfederation.eu/european-journal-of-pain/)
 - *Journal of Pediatric Psychology* (academic.oup.com/jpepsy)
 - *Qualitative Health Research* (journals.sagepub.com/home/ghr)
 - *Social Science and Medicine* (www.springer.com/journal/44155/)
 - *Sociology of Health and Illness* (onlinelibrary.wiley.com/journal/14679566)
- Contacting experts in the field for recommended studies, including ongoing research.
- Checking the reference lists of included papers for any further relevant studies.

Furthermore, if a relevant study lacks contextual information, we will perform 'cluster searches', which involve identifying 'clusters' of related study reports to reconstruct the study context (Booth 2013b).

Selection of studies

One review author will conduct initial screening of retrieved references by title to exclude off-topic texts, that is those clearly not about childhood chronic pain. Following initial piloting and standardisation between review authors, two review authors (any two of the research fellow, RT, EF, IU, LF, AJ, and JN) will independently screen each reference by title and abstract, and then by full text to assess their relevance using Covidence systematic review management software (Veritas Health Innovation 2022). Any disagreements will be resolved through discussion or by referring to a third review author, if necessary.

The funder of this research has a key interest in the UK context of the National Health Service. We will include both UK and international studies to answer review questions 1, 2, 3, and 4 on how children and families conceptualise and live with chronic pain and experience and conceptualise good pain management.

Findings regarding how children and their families conceptualise and live with pain (review questions 1 - 2) (e.g. from international studies of sickle cell disease-related pain) could be transferable to a UK context, particularly given the diverse ethnic and cultural make-up of the UK population, or could provide a contrasting perspective to inform theory-building (see paragraph 1 in the 'Topic of interest' section above). International studies (e.g. from economically developed countries) might indicate new service models or interventions (review question 4) for potential use in the UK. We will indicate whether findings relate to the UK or to other countries when reporting the review findings. We will make decisions in light of the characteristics and content of the whole body of relevant studies and with our PPI group. A PRISMA flow diagram will record/show the search results and the results of screening and selecting studies for inclusion.

Language translation

During the screening process, all titles and abstracts that are published in a language in which none of the review team is proficient (i.e. languages other than English, Portuguese, Spanish, and French) will undergo an initial translation through open source software (e.g. Google Translate (Google Translate)). We will consult members of the Cochrane Task Exchange platform (taskexchange.cochrane.org/) or other networks that are proficient in that language to assess the full text of the paper if the initial translation indicates inclusion or if the translation is inadequate to permit a decision. If a suitable translator cannot be found, we will assess the study as awaiting classification to ensure transparency in the review process.

Sampling of studies

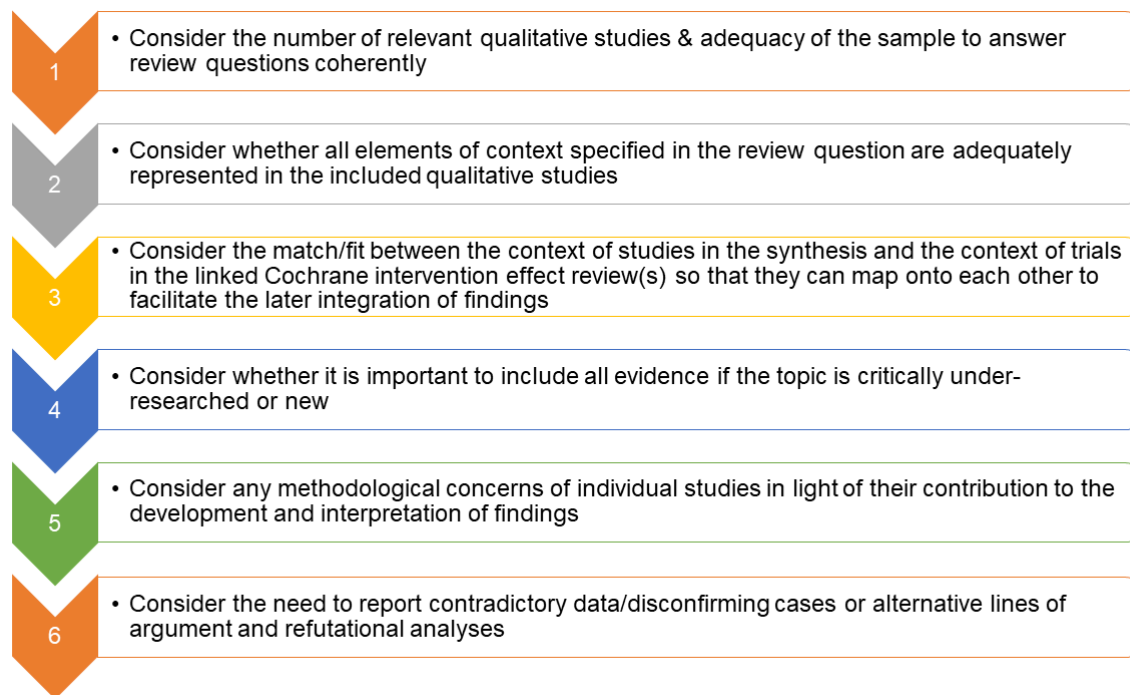
Sampling from the body of relevant literature is an iterative process. We will make an initial selection of relevant studies that meet the inclusion criteria. This will be followed by further purposive sampling of relevant studies to ensure we conduct a focused, meaningful synthesis that answers our review questions (Ames 2019; Benoot 2016). In a qualitative evidence synthesis, it is neither necessary nor desirable to include every relevant study to produce meaningful results because the purpose is to develop understanding of a phenomenon, not to make predictions or to produce a definitive conclusion about the effectiveness of an intervention (Ames 2019; Benoot 2016). Having too many studies, and therefore too much data, to synthesise can interfere with the ability to conduct an in-depth analysis and so result in a superficial analysis (Ames 2019). The volume of relevant data in the studies, relative to team resources, can thus influence the number of studies it is desirable to synthesise (France 2019e). Several authors have conducted prior high-quality meta-ethnographies following the key principles of Noblit 1988's original methodology successfully on 40 to 50 studies (Campbell 2011; Germeni 2018), whilst other adaptations of Noblit 1988's methodology have synthesised a greater number of studies (e.g. 77) (Toye 2013).

To help guide sampling decisions, we will judge the conceptual 'richness' of included studies, that is whether the findings are explanatory (see 'Assessing the methodological limitations of included studies' section below), and select rich studies for synthesis. If necessary, we will sample iteratively; first, we will assess the richness of all eligible UK studies and select rich studies. We will then identify the non-UK studies the aims of which most closely match our review questions; assess these for richness; and

select rich non-UK studies. Sampling decisions will be made in collaboration with our PPI group, to ensure that the synthesis addresses what is of greatest importance to children and families, and using Cochrane Qualitative Implementation Methods Group (QIMG) guidance on how to select a sample of studies to answer our review questions (see [Figure 1](#) for QIMG key assessment criteria) ([Noyes 2018b](#)). For example, we might revise our inclusion criteria,

for example to include or exclude studies in which adults give retrospective accounts of their own childhood chronic pain. We will document the reasons for any such decisions. We will take into account the potential importance of the distinction between primary (e.g. fibromyalgia) and secondary pain conditions (e.g. sickle cell disease) when sampling studies.

Figure 1. Key criteria to consider when selecting studies to synthesise, adapted from Noyes and colleagues (Noyes 2018b).



Data extraction

At least two team members will read all of the studies in full and will read studies again as needed throughout the analysis process (all members will read some studies). As analytic phases overlap, reading is not a one-off activity. We will record study characteristics (e.g. aim; methods of data collection and analysis; country; number and type of participants (e.g. patients, parents or other family members, gender, age, diagnosis, ethnicity, etc.)). We will refer to the PROGRESS-Plus criteria (place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, and social capital) when extracting data on participant characteristics ([O'Neill 2014](#)). We will also record or 'extract' studies' conceptual findings wherever they appear in the article, not just from the findings sections, using NVivo 12 qualitative analysis software ([QSR International 2018](#)).

Assessing the methodological limitations of included studies

Two review authors will independently assess the methodological limitations of relevant studies using the Critical Appraisal Skills Programme (CASP) qualitative tool ([CASP 2018](#)). As part of the appraisal process, we will judge the conceptual richness of the primary studies, that is whether the findings are explanatory rather

than just descriptive ([Popay 1998](#)). We will select rich studies for inclusion ([France 2019e](#)). We will grade references against preset criteria describing conceptual richness by adapting Ames and colleagues' scale for assessing data richness ([Ames 2017](#); [Ames 2019](#)), informed by Popay and colleagues' approach to judging richness and Cochrane QIMG guidance ([Noyes 2022](#); [Popay 1998](#)). [Popay 1998](#) differentiated between descriptions that state facts in isolation from the context, intentions, or circumstances (which we refer to as conceptually 'poor') and those which provide the context, intentions, and meanings behind qualitative findings (which we refer to as conceptually 'rich').

We will not exclude studies that are limited by poor methodological reporting because there is a distinction between quality of methodological reporting and quality of output/findings; however, we will exclude studies that we judge to be fatally flawed (e.g. methodologically unsound). Ultimately, the quality of studies will be determined by the degree to which they contribute to the synthesis findings. Any disagreements will be resolved by discussion or by consulting a third review author if necessary. If a team member is the author of a relevant study, they will not be involved in quality appraisal of that study to ensure an unbiased appraisal. We will transparently record all decision-making and reasons for study exclusion in Microsoft Excel ([Microsoft Corporation 2018](#)). Results of quality appraisal will inform GRADE-

CERQual (Confidence in the Evidence from Reviews of Qualitative research) judgements of how much confidence can be placed in our synthesised findings.

We will assess methodological limitations according to the following domains.

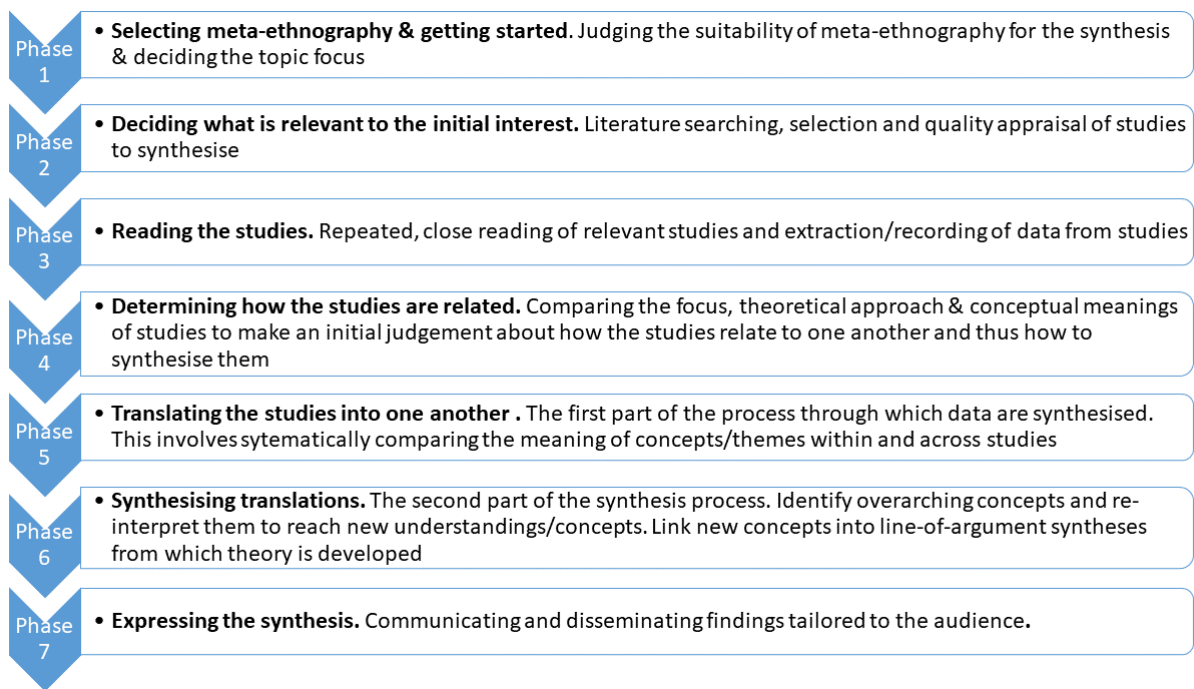
- Was there a clear statement of the aims of the research?
- Is a qualitative methodology appropriate?
- Was the research design appropriate to address the aims of the research?
- Was the recruitment strategy appropriate to the aims of the research?
- Were the data collected in a way that addressed the research issue?
- Has the relationship between researcher and participants been adequately considered?
- Have ethical issues been taken into consideration?
- Was the data analysis sufficiently rigorous?
- Is there a clear statement of findings?

We will report our assessments in a Methodological Limitations table.

Data management, analysis, and synthesis

We intend to conduct a meta-ethnography (Noblit 1988), provided that the available data in primary studies are sufficiently rich, following the eMERGe meta-ethnography reporting guidance, France 2019a; France 2019b; France 2019c; France 2019d, and QIMG guidance (Noyes 2018a). Meta-ethnography is suited to developing new understandings and theory (Noblit 1988), and can also indicate gaps in knowledge and thus new directions for research (Campbell 2011). A meta-ethnography involves interpreting the concepts, findings, or themes from existing accounts of primary qualitative studies (e.g. those using in-depth interviews) in order to try to develop novel insights that were not apparent in any single study (France 2019c; Noblit 1988). It does not involve simply aggregating findings (Noblit 1988). The seven phases of meta-ethnography are described in Figure 2; although presented linearly, some phases run in parallel, and the process is iterative (Cunningham 2019; France 2019c; Noblit 1988). Meta-ethnography has a unique synthesis method that involves systematically comparing the meaning of concepts from primary studies; identifying new overarching concepts; and linking these in order to develop theory (Campbell 2011; France 2019c).

Figure 2. The seven phases of meta-ethnography (Noblit 1988).



Once we have identified relevant studies, we will determine how the studies relate to one another by comparing their aims, focus, characteristics, and findings. Next we will organise studies, for example by health conditions (e.g. juvenile idiopathic arthritis, inflammatory bowel disease) and type of pain (e.g. chronic migraine, musculoskeletal pain); by whose views are presented (e.g. the child, parents, or siblings); or the child's age (e.g. 0 to < 2 years old, 2 to ≤ 5 years, 6 to 12 years, 13 to < 16 years, 16 to < 18 years), and synthesise each group of studies separately

before synthesising them all together (Campbell 2011). Quality meta-ethnographies have used this approach successfully, and it enables synthesis of diverse studies (Campbell 2011). The precise method for grouping studies will only be decided once we have identified relevant studies and become familiar with their content in order to determine the best way of grouping and organising them for synthesis (France 2019e).

In our final sample, we will aim to ensure a balance of heterogeneity and homogeneity of studies so that we can conduct ‘reciprocal translation’ (looking for similarities in meaning), but also include contradictory findings through ‘refutational translation’ (looking for differences in meaning) (France 2019e). Refutational or contradictory data are important for developing comprehensive understandings and theory building (Booth 2013a). Including studies conducted in a range of countries could be an important element of identifying similarities and differences in the conceptualisation of chronic pain and what ‘good’ chronic pain management looks like amongst different ethnic, national, and cultural groups. We will compare the findings of our synthesis to those of our prior WHO synthesis to help ensure that there is a global focus to the synthesis (WHO 2020c). Where studies report gender/gender differences, we will explore gender differences in the views and experiences of children and their parents.

We will use a synthesis method similar to that described by Campbell 2011, which compares concepts one by one, study by study (e.g. in chronological order), for each grouping of studies. This method has the following advantages over other methods: it does not impose an analytic framework on the data; it allows the researchers to stay close to the meanings and contexts of the original studies; and is faithful to Noblit 1988’s original method (France 2019e). The process of translation is key to conceptual interpretation and synthesis, so it is important to adhere to the principles of translation (France 2019e). We will ‘translate’ or synthesise each group of studies separately before synthesising across groups (Campbell 2011).

We will aim to reach new interpretations which we will develop into an explanatory theory. An example of a possible theory to be produced is an evidence-based model of the attributes that children with chronic pain and their families want in a pain management service.

For rigour and richer interpretation, the analytic synthesis phases will involve at least three team members with input from the wider team. Six to eight young people with chronic pain and parents from our PPI group will participate in a data analysis and interpretation workshop. We will maintain a reflexive approach during analysis and make clear any potential conflicts of interest, for example when interpreting any studies by our team that are included in the synthesis.

If a meta-ethnography is not possible or appropriate for the data identified in primary studies (e.g. if they are mostly or

all conceptually poor), then an alternative suitable qualitative evidence synthesis approach will be selected following Cochrane QIMG guidance (Noyes 2022).

Assessing our confidence in the review findings

Two review authors (the research fellow and EF) will use the GRADE-CERQual approach to assess our confidence in each finding (Lewin 2018). GRADE-CERQual assesses confidence in the evidence, based on the following four key components.

- Methodological limitations of included studies: the extent to which there are concerns about the design or conduct of the primary studies that contributed evidence to an individual review finding.
- Coherence of the review finding: an assessment of how clear and cogent the fit is between the data from the primary studies and a review finding that synthesises those data. By cogent, we mean well-supported or compelling.
- Adequacy of the data contributing to a review finding: an overall determination of the degree of richness and quantity of data supporting a review finding.
- Relevance of the included studies to the review question: the extent to which the body of evidence from the primary studies supporting a review finding is applicable to the context (perspective or population, phenomenon of interest, setting) specified in the review question.

After assessing each of the four components, we will make a judgement about the overall confidence in the evidence supporting the review finding. We will judge confidence as high, moderate, low, or very low. The final assessment will be based on consensus amongst the review authors. All findings start as high confidence, and will then be downgraded if there are important concerns regarding any of the GRADE-CERQual components.

Summary of qualitative findings tables and evidence profiles

We will use summary of qualitative findings tables to present summaries of the findings and our assessments of confidence in these findings, as shown in Table 2. We will use evidence profiles to present detailed descriptions of our confidence assessments, as shown in Table 3.

Table 2. GRADE-CERQual summary of findings table template

Summary of review finding	Studies contributing to the review finding	GRADE-CERQual assessment of confidence in the evidence	Explanation of GRADE-CERQual assessment

Table 3. GRADE-CERQual evidence profile table template

Summary of review finding	Studies contributing to the review finding	Methodological limitations	Coherence	Adequacy	Relevance	GRADE-CERQual assessment of confidence in the evidence	Explanation of GRADE-CERQual assessment

Integrating the review findings with the Cochrane intervention reviews

We will integrate our qualitative findings with the results of all relevant Cochrane intervention effectiveness reviews (e.g. [Cooper 2017a](#); [Cooper 2017b](#); [Cooper 2017c](#); [Cooper 2017d](#); [Eccleston 2017](#)), using an appropriate quantitative/qualitative data integration method from Cochrane QIMG ([Harden 2018](#)), to determine if the programme theories and outcomes of interventions match families' views and expectations. We will check that the contexts (e.g. the population and setting) of the intervention studies are sufficiently similar to the contexts of the qualitative studies prior to integration ([Noyes 2016b](#)). Our findings will help to explain why and how certain interventions seem to be more effective than others in specific contexts and for specific children. They will inform the design of future treatment effectiveness reviews by suggesting family-centred outcomes and generating hypotheses that can be tested out, for example, in future subgroup analyses. They will also contribute to developing more relevant, acceptable, and effective interventions through greater understanding of the pain experience from the perspective of children, parents, and wider family members.

There are various points in overall meta-ethnography production at which integration can occur ([Harden 2018](#); [Noyes 2019](#)). We have integrated during review question formulation and will integrate the following during synthesis.

- Question formulation: the meta-ethnography review questions have been formulated to address known gaps in the Cochrane intervention effectiveness reviews.
- Synthesis: we plan to use a matrix approach adapted from one used previously in several Cochrane Reviews (for example [Munabi-Babigumira 2017](#)). Our matrix will explore whether potential implementation factors (acceptability, feasibility, patient values, preferences and desired outcomes, etc.) identified in our meta-ethnography have been acknowledged or addressed in the intervention programme theories in the related Cochrane Reviews of intervention effectiveness.

Review author reflexivity

The review team (the authors plus the research fellow) have varied professional backgrounds including sociology (IU, RT), psychology (EF, LF), health psychology (AJ, LC), family therapy (LF), nursing children with chronic pain (JN), physiotherapy with children who have chronic pain (research fellow), development of evidence synthesis methodology (JN, EF, RT, IU), children's pain research (AJ, LC), and health services research (all). Four of us (JN, LF, AJ, research fellow) have clinical backgrounds, and seven of us (EF, IU, RT, LC, AJ, JN, LF) have social science backgrounds. The review team does not have personal experience of chronic pain in childhood, either their own or as a parent; however, three members have experienced chronic pain as an adult. We believe that children have a fundamental right to pain treatment and to be pain-free and that all pain is real and to be believed.

If a team member is the author of a relevant study, they will not be involved in assessing its methodological limitations to ensure an unbiased appraisal; a senior team member will take overall responsibility for the assessments of such studies. We will maintain a reflexive approach during analysis and make clear any

potential conflicts of interest, for example when interpreting any studies by our team that are included in the synthesis. AJ and LC have authored several qualitative study publications on children's chronic pain, but the other team members are independent of that research. EF and JN conducted a qualitative evidence synthesis on interventions for children's chronic pain globally for the WHO.

All review authors will keep a reflexive stance during all the stages of the review process. The chief investigator and research fellow, who will input the greatest time contribution to the review, will keep a reflexive journal during the review process. The review process and progress will be regularly assessed and discussed between the researchers and using PPI input. The PPI input throughout the review will also minimise the risk of influencing our selection of studies, analysis, and the interpretation of the findings based on our preconceptions and backgrounds.

Based on our collective and individual experiences (as clinicians, academics, and researchers), we anticipate the findings of our review might reveal a mismatch between the current pain management treatments and services and the design and outcomes of trials research compared to the needs and wants of children with chronic pain and their families. They might also show that the rhetoric of seeing chronic pain as something that impacts the whole family is not borne out by people's experiences. We will take a reflexive approach throughout the review by interrogating how our professional and personal assumptions could influence our interpretation of the data and our interpretation of our own findings.

Project advisory group and patient and public involvement group

We have established a Project Advisory Group (PAG), comprised of children with chronic pain and their family members, healthcare professionals, representatives from health services and government, and patient representatives from the third sector, which will advise the project team on four key areas:

- methodological issues;
- clinical and lived experience of chronic pain;
- study conduct;
- project dissemination.

The PAG had two independently chaired meetings with the research team in May 2021 and April 2022. Incorporating a range of stakeholders, selected for heterogeneity of chronic pain experience, from the project outset will maximise the likelihood that the research will be acceptable and relevant to children and families, and health professionals.

In addition, we have PPI in the review guided by UK national PPI standards ([UK 2019](#)). We had PPI input to help to ensure the study aim, review questions, and outcomes were important for patients and their families. They also provided feedback on our PPI and dissemination plans. Our UK-based project PPI group will consist of a purposefully diverse group of 6 to 8 children with chronic pain aged 8 to < 18 years and parents and informal carers (i.e. not clinicians) of children aged 3 months to < 18 years. We will seek an international PPI perspective through consulting with global patient support organisations and groups.

We aim to collaborate with and consult our project PPI group during two workshops and interim communication, for example via email, teleconferences, and/or social media, such as a private Facebook or WhatsApp group, depending on members' preferences. Table 4

shows the key aspects of the project in which we envisage the PPI group to be involved.

Table 4. Patient and public involvement in different stages of the meta-ethnography

Phase	Activity	Level of involvement	Method of involvement
Planning of review	Feedback on study aims, objectives, review questions, lay summary & dissemination strategy	Consultation	Email
Protocol	Finalise the study protocol, e.g. the literature search strategy	Collaboration & consultation	Teleconference, email, online
Selection & sampling of studies	Finalise inclusion/exclusion criteria, e.g. the types of chronic pain included, the characteristics of the population to be included. Sample studies for synthesis	Collaboration & consultation	Online workshop 1, March 2021
Analysis & synthesis	Decide how studies will be organised/grouped for analytic synthesis, e.g. grouping them by type of chronic pain, age of participants	Collaboration	Online workshop 1, March 2021
Analysis & synthesis	Analyse & interpret primary study findings, e.g. to check if our interpretation of the study findings is different from or the same as children and families' interpretations, check if their experiences are similar or different to those of the people in the studies, if important areas are missing from research	Consultation	Face-to-face or online workshop 2, September 2021
Dissemination	Producing outputs, dissemination. We will invite two members to co-present a conference paper and the group to co-develop lay, patient, and policy outputs. The group will help ensure that the development of lay dissemination materials for children and families is appropriate and relevant.	Collaboration & consultation	Teleconference, email, online. Co-present at a conference

Table 4 Key: 'Consultation' refers to when the team will prepare information about research and discuss this with the PPI group, who will be asked to comment on and present their views and experiences in response. The ACTIVE framework for involving users in systematic reviews calls this the PPI group 'influencing' the research (Pollock 2019).

'Collaboration' refers to when children and families will be involved in performing the research as well as in setting priorities and making decisions. The ACTIVE framework calls this the PPI group 'controlling' the research (Pollock 2019).

We will be flexible in response to how children and young people with chronic pain and their families want to be involved, and tailor our involvement methods to their needs. We will survey the training and support needs of our PPI group members prior to commencing PPI activity, and training for PPI members will be tailored to their needs and will evolve as the project progresses in line with their wishes. We aim to collaborate with and consult our project PPI group during two workshops and interim communication, for example via email, teleconferences, and/or social media such as a private Facebook or WhatsApp group, depending on members' preferences. Table 4 shows the key aspects of the project we envisage the PPI group will be involved in. All meetings will be tailored to the children's needs, for example short duration, frequent breaks, appropriate language, and interactive formats (e.g. cartoons, videos). PPI input, based on their experiences, will

contribute to finalising the study design, helping to define sampling strategy and subgroups for synthesis. We will compare our findings with their experiences, identifying important areas that are missing from the existing research. We will also consult PPI to help us decide the best approach to disseminating our findings, including deciding the content of outputs. For our meetings, we will use creative and fun ways to get children involved that are tailored according to their preferences. For example, in our second meeting we will present key themes from study findings in a fun, interesting way according to their preferences, using visual and interactive methods, such as videos and cartoons, and invite them to share their relevant experiences.

The contribution and impact of the PPI group will be recorded prospectively throughout the study by the research fellow, and

integrated into the final report and other dissemination outputs where appropriate. PPI members will be reimbursed for their time and out-of-pocket expenses in line with [INVOLVE](#) guidance.

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Editorial and peer-reviewer contribution

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The following people conducted the editorial process for this article.

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- Assistant Managing Editor (conducted editorial checks and supported the editorial team): Kerry Harding, Oxford University Hospitals (OUH) NHS Foundation Trust, Oxford, UK
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- Lisa Winer (copy-editing and production): Cochrane Copy Edit Support
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APPENDICES

Appendix 1. Search strategy for Medline

1. Qualitative Research/ or Interview/ or Nursing Methodology Research/,
2. (ethnonursing or phenomenol* or emic or etic or hermeneutic* or heuristic* or semiotic* or theoretical sampl*).ti,ab.,
3. (qualitative adj3 (study or research or method* or analysis or cod* or them* or interview* or question*1 or data)).ti,ab.,
4. (thematic analysis or ethnological research or ethnograph* or life stor*).ti,ab.,
5. (theme*1 adj2 (qualitative or analysis or coding or codes or grouping or identif*)).ti,ab.,
6. (grounded adj2 (theor* or study or studies or research or analys?s)).mp.,
7. (data adj1 saturat*).ti,ab.,
8. ("social construct*" or postmodern* or post-structural* or post structural* or poststructural* or post modern* or post-modern* or feminis* or action research or cooperative inquir* or co operative inquir* or co-operative inquir* or humanistic or existential or experiential).mp.,
9. (field adj (study or studies or research)).ti,ab.,
10. (human science or biographical method or participant observ*).ti,ab.,
11. ((purpos* adj4 sampl*) or (text* adj1 analysis) or (focus group* or observational method* or "content analysis" or "narrative analysis")).mp.,
12. (unstructured or open-ended or open ended or narratives or life world or life-world or conversation analys?s or personal experience* or theoretical saturation).mp.,
13. ((lived or life or patient or carer* or guardian* or parent* or mother* or father* or family*) adj2 (account or accounts or perspective* or interpretations or experience*)).ti,ab.,
14. ((children* or adolescent*) adj2 (account or accounts or perspective* or interpretations or experiences or experience)).ti,ab.,
15. or/1-14 [**Concept A - study design - qualitative**]
16. (adolescenc* or preadolescenc* or baby or babies or infan*2 or toddler* or preschool* or pre-school* or child or children or childhood or girls or boys or kid or kids or juvenile or teen* or preteen* or youth or youngster*).ti,ab.,
17. (pupil or pupils or school-aged or school pupil* or schoolchild* or paediatric* or pediatric*).ti,ab.,
18. exp child/ or adolescent/ or Parent-Child Relations/,

19. ((carer* or caregiver* or family or families) and (child or children or young*)).ti,ab.,
20. (parent*1 or mother*1 or father*1 or daughter*1 or son or sons).ti,ab.,
- 21 or 16-20 [**Concept B – Population – children and their families**]
22. exp Chronic Pain/ or exp Complex Regional Pain Syndromes/,
23. ((chronic or longterm or long?term or persist* or sustain* or continued or continuous or recurr*) adj5 (pain* or cephalalgi* or ache or aches)).ti,ab.,
24. ((chronic or longterm or long?term or persist* or sustain* or recurr* or frequent) adj5 (headache or migraine or cramps or cramp-ing)).ti,ab.,
25. (pain* adj3 (condition or conditions or disorder or disorders or illness or illnesses or disease or diseases or recurrent or debilitating or complex or long*)).ti,ab.,
26. (((chronic or long-term) adj3 (condition or conditions or disorder or disorders or illness or illnesses or disease or diseases)) and pain*).ti,ab.,
27. (pain* adj3 (neuropathic or syndrome*)).ti,ab.,
28. (pain* and (sickle cell disease or arthritis or chronic pancreatitis or lupus or costochondritis or tietze syndrome or "ehler's" or fibromyalgia or irritable bowel syndrome or ibs or reflex sympathetic dystrophy or non-cardiac chest pain or chronic fatigue syndrome or myalgic encephalomyelitis or "me/cfs" or endometriosis or Dysmenorrhea or Inflammatory bowel disease or IBD)).ti,ab.,
29. exp Pain/ and exp Chronic Disease/,
30. or/22-29 [**Concept C – phenomenon – Chronic pain**]
31. 15 and 21 and 30 [**Concept A AND B AND C**]

Box 1 Key:

ti,ab = keyword search in title and abstract;

* = truncates a keyword;

adjn = number of words away one search term is from the other, in any order;

/ = subject heading;

? = option for any letter e.g. 'analys?s' would pick up analysis or analyses.

CONTRIBUTIONS OF AUTHORS

The protocol authors all contributed to the conception or design of this research, drafting the protocol or revising it critically for important intellectual content, and final approval of the version to be published. EF is the guarantor of the review.

DECLARATIONS OF INTEREST

Emma France declared no financial conflicts of interest. She is a member of the Cochrane Qualitative & Implementation Methods Group, whose publications we will use, and is an author of the meta-ethnography reporting guidance we will use.

Jane Noyes declared no financial conflicts of interest. She was involved in developing and publishing the original GRADE-CERQual publications we are using. She leads the Cochrane Qualitative & Implementation Methods Group whose publications we will use, and is an author of the meta-ethnography reporting guidance we will use.

Liz Forbat declared no financial conflicts of interest.

Isabelle Uny declared no financial conflicts of interest. She is an author of the meta-ethnography reporting guidance we will use.

Abbie Jordan declared no financial conflicts of interest. She has authored several qualitative study publications on children's chronic pain which might meet the review's inclusion criteria.

Line Caes declared no financial conflicts of interest. She has authored several qualitative study publications on children's chronic pain which might meet the review's inclusion criteria.

Ruth Turley declared no financial conflicts of interest. She is an author of the meta-ethnography reporting guidance we will use.

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