# Study 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


## Background & Rationale

Chronic pain in childhood is widespread, affecting at least 8% of children and young people[1] in the United Kingdom (UK). Chronic pain has considerable negative impacts on children's lives and quality of life and leads to increased use of healthcare services and medication. Treating adolescent pain alone costs the NHS around £4billion a year. In the UK, services for children rely largely on primary care services with limited provision of and access to specialist chronic pain services and multidisciplinary pain management. We also do not know which treatment outcomes children and families value. To develop, design and deliver appropriate services and interventions which meet the needs of patients and their families, it is vital that we understand how children with chronic pain and their families experience chronic pain, their views of health and social care services and treatments for chronic pain, and which outcomes are important to them.

## Aim

To conduct a meta-ethnography on the experiences and perceptions of children with chronic pain and their families of chronic pain, treatments and services. We will use these data to inform the design and delivery of health and social care services, interventions and future research. The review questions are:

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

## Design & Methods

Design: We will conduct a qualitative evidence synthesis using meta-ethnography, a seven-phase, systematic, interpretive methodology well-suited to informing the complex healthcare issues involved in childhood chronic pain. This inductive methodology takes into account the contexts and meanings of the original studies making it ideal for synthesising the diverse contexts of children's chronic pain research.

Review strategy: we will carry out comprehensive searches of 13 bibliographic databases and iterative supplementary searches, e.g. citation tracking, to identify qualitative studies with children aged 3 months to 18 years with chronic non-cancer pain and their families. We will independently screen retrieved references against inclusion criteria agreed with our patient and public involvement group. We will quality appraise studies using the Critical Appraisal Skills Programme tool and purposively sample them to ensure they address the review questions. We will 'extract' data on study aims, focus, characteristics and conceptual findings from wherever they appear in the study reports using NVivo software. We will compare these study data to determine how the studies relate to one another to decide how to order and group them for synthesis. We will synthesise each group of studies separately before synthesising them all together. Analysis and interpretation of studies will involve children with chronic pain and their families and result in theory to inform service design and delivery. We will assess the confidence that decision-makers can place in our findings using the GRADE CERQual tool to facilitate their use for NHS decision making and use a matrix approach to integrate our findings with existing Cochrane reviews on treatment effectiveness for children's chronic pain.

## Patient and Public Involvement (PPI)

Ten patients and lay people, including children with chronic pain and their parents, were involved in proposal development. They gave feedback on the project aims, patient involvement and dissemination plans, and lay summary. A diverse project PPI group of 6-8 children with chronic pain and 6-12 parents will be recruited and receive tailored training. They will participate in 2


[1] From now, we will use the term 'children' to refer to 'children and young people' throughout the document.

workshops with additional communication by teleconference, email and social media to collaborate on finalising the protocol, sampling and organising studies for synthesis, interpreting findings, and dissemination.

## Timelines for delivery

The study will last 24 months from 1 August 2020 to 31 July 2022.

## Dissemination, outputs & anticipated impact

We will disseminate widely to academic, lay, clinical and policy audiences. Outputs will include a three-minute animated film on YouTube for children, three academic journal articles, three conference presentations, a funders' report, a webinar, a lay radio podcast, an infographic, a policy briefing and a public Twitter conversation. The findings will contribute to the development and updating of existing National Institute of Health and Care Excellence and Scottish Intercollegiate Guidelines Network clinical guidelines on chronic pain in children; inform training of health and social care professionals regarding children's chronic pain management; and inform service, treatment and intervention design and delivery. Ultimately, this will improve the care provided and thus the health and quality of life of children with chronic pain and their families, and reduce burden on the NHS.

# 1. Background and rationale

Chronic pain in childhood is widespread: United Kingdom (UK) figures indicate that 8% of primary care consultations by 3 to 17 year olds are for musculoskeletal chronic pain alone (1); at least 4% to 14% of children worldwide are estimated to have chronic pain, but the prevalence could be as high as 24% to 88%, dependent on the type of pain (2). Frequent severe chronic pain of all types affects 8% of children, according to a Dutch survey (3). The International Association for the Study of Pain's widely accepted definition of chronic pain is pain which persists past normal tissue healing time or lasts/recurs for more than 3 to 6 months (4). Chronic pain is recognised as a condition in its own right but it is also a key feature of conditions, such as complex regional pain syndrome, inflammatory bowel disease and juvenile idiopathic arthritis. Chronic pain has considerable negative impacts on children's health and quality of life, for instance, UK surveys have shown that the majority of adolescent children with chronic pain experience disability in physical, mental and social health (5) and feel developmentally behind their peers (6). Chronic pain is associated with increased use of healthcare services and medication (7), adversely affects social and family relationships (8) and results in poorer school attendance (9). For treating adolescent pain alone, the cost to the NHS is estimated at four billion pounds a year (10). UK parents pay £900 out-of-pocket expenses a year supporting their adolescent's chronic pain and have work absences of 7 to 37 days costing on average £750 per family each year, with some parents giving up work entirely to care for their adolescent child (11). Moreover, longitudinal research indicates a high risk of childhood chronic pain continuing into adulthood (12) with further individual, NHS costs and societal costs, for example, adult back pain alone costs the UK economy £11 billion with a direct healthcare cost of £1 billion (13).

Despite the high prevalence and serious impacts of childhood chronic pain, UK provision of and access to specialist children's chronic pain services and multidisciplinary chronic pain management is very limited and of inconsistent quality (7). Furthermore, the 2018 Scottish Intercollegiate Guidelines Network (SIGN) guideline on the management of children's chronic pain indicated that UK-based healthcare professionals require more training in managing chronic pain in children (7). For instance, children in Scotland are referred to national specialist services in Bath or Oxford for intensive pain management programmes because of a lack of local expertise (14). There is also a severe lack of high quality trials research to inform management of childhood chronic pain (7, 15-19). This has been identified in the SIGN guideline (7), and five recent Cochrane reviews of treatment effectiveness trials for children's chronic non-cancer pain (15-19). The National Institute of Health and Care Excellence (NICE) chronic pain guideline, due out in 2020, will not include children under 16 years of age due to the lack of robust trials evidence (20). The Cochrane reviews concluded that there is a clear knowledge gap that must be addressed before further trials are run (15-19), that is: we do not know which outcomes are important to children with chronic pain and their families (15-18). Further, while psychological interventions which engage children and/or parents improve child outcomes (21), a family-system approach to chronic pain research is lacking despite a call for this over a decade ago (22). The reviews also emphasised that future research must recognise the heterogeneity of children's chronic pain (16).

To design and deliver services and interventions which meet the needs of children and their families, it is crucial that we 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 addressing these urgent and important questions. Our preparatory work and scoping searches indicate there is existing relevant qualitative research to inform these issues (e.g. (23-27)), but there are no existing or planned qualitative evidence syntheses of this research. These conclusions are based on our searches of PROSPERO, the International Prospective Register of Systematic Reviews (in May 2019); bibliographic databases MEDLINE, PsychINFO, and Pubmed; and Google Scholar using keywords (pain/chronic pain, children/paediatric); the Cochrane library; our team's reference databases; and reference lists in policy documents and

Cochrane reviews. We also consulted experts in the field and checked Zetoc (journal article monitoring and search service) alerts of all newly-published qualitative evidence syntheses. We identified only two existing qualitative evidence syntheses which are limited in focus. They look at specific childhood chronic pain populations and topics - living with juvenile idiopathic arthritis (28) and adolescent social relationships (8) - and did not develop a theory to inform pain management. Therefore we will conduct a qualitative evidence synthesis using meta-ethnography (29) - a rigorous, systematic, qualitative evidence synthesis 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 (children with cancer-related chronic pain have different care pathways) and generate theory to inform health and social care. The available body of qualitative evidence is suitable to synthesise using meta-ethnography (see section 3.2.1).

Our study is important to patients' health and the NHS because it will produce robust, novel evidence to inform and support management of childhood chronic pain. It will lead to new conceptual insights and theories (29, 30) that will change healthcare delivery and policy, inform treatments, and indicate gaps in knowledge and hence new directions for chronic pain research (31). Evidence syntheses are important (32) and economical in drawing together existing research to develop generalisable/ transferable insights and understandings. Because chronic pain is an aspect of many health conditions our findings will have wide reach and transferability across patient groups, while recognising the heterogeneity of children's chronic pain (16). Our meta-ethnography will: 1. inform identification of child- and family-centred outcomes, 2. help us better understand how children and families conceptualise and live with chronic non-cancer pain, and 3. inform a more family-orientated approach to chronic pain management in order to support the NIHR HS&DR programme's aim to produce rigorous, relevant evidence on the quality, access and organisation of NHS services for children's chronic pain. We will extend the findings of existing Cochrane intervention effectiveness reviews (15-19) 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 will also direct future effectiveness reviews to address outcomes of importance to patients. These are two of the important 'added-value' roles of qualitative evidence synthesis recognised by Cochrane (32).

## 1a. Why this research is needed now

There is an urgent need for research to enhance our understanding of the experiences, perceptions and needs of children with chronic pain and their families in order to improve services, treatments, and hence children's health and quality of life. The 2018 SIGN guideline (7) on the management of pain in children and five recent Cochrane reviews on treatment effectiveness for children's chronic pain (15-19) identified a dearth of research to inform chronic pain management. The upcoming 2020 NICE chronic pain guideline (20) will not include children under 16 years of age due to the lack of quality trials evidence. The Cochrane Pain, Palliative and Supportive Care (PaPaS) group has recently prioritised research into children's chronic pain (33) and the International Association for the Study of Pain has 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 (34). The NIHR has also recognised the urgent need for research on chronic pain management with its themed call for research in this field.

Despite the high prevalence and serious impacts of children's chronic pain, current services for managing children's chronic pain are inadequate (35, 36); there is a lack of high quality trials evidence to inform NICE (20) and SIGN (7) clinical guidelines, and thus guide chronic pain management; and insufficient knowledge of which outcomes are important to patients and their families (15-18) to guide design of services and treatments and to inform future research. The most recent National Pain Audit in England and Wales in 2011-12 (36) found that services are 'of inconsistent standard and quality and not always available for those who need them. This is particularly true for centres specialising in treating children with chronic pain' (36) (p. 4). Hence, there is an urgent need for the NHS to improve how it supports children with chronic pain and their families, using high-quality evidence to inform design and delivery of services and treatments. The

2018 SIGN guideline (7) mostly relies on expert opinion in the absence of robust research evidence concluding that 'high quality evidence is needed in all areas of paediatric chronic pain' (7) (p. 40). The recent Cochrane treatment effectiveness reviews (15-18) highlighted the lack of patient-defined outcomes related to pain relief or improvement of function, indicating 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. Furthermore, current NHS patient health information websites (37, 38) have inadequate coverage of children's chronic pain and would benefit from including evidence-based information; for example, the information page on cerebral palsy does not mention chronic pain or its management even though it is reported as a common issue for children. Without high-quality evidence, children are not receiving evidence-based good-quality pain management, resulting in poor short-term 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 (12).

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 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 recent Cochrane reviews on managing children's chronic pain (15-19), the SIGN guideline (7), and clinical position statements (39), yet the Core Standards for Pain Management Services in the UK (40) specify that a bio-psycho-social approach, which takes into account the whole range of biological, psychological and social influences on pain, is required. Qualitative research typically adopts a bio-psycho-social perspective (41). It is also well-suited to identifying outcomes valued by children and families to enable future meaningful quantitative synthesis within Cochrane reviews. Meta-ethnography is ideally matched to synthesising qualitative evidence on the complex issues related to children's chronic pain. We will use meta-ethnography (29) to synthesise the relevant qualitative studies on children's chronic pain. The recently published eMERGe meta-ethnography reporting guidance (30, 42-44) and associated methodological publications (45, 46) will facilitate the production of a high quality meta-ethnography, in line with the most up-to-date methodological and reporting guidance which is only now available.

## 2. Aims and Objectives

Aim: To conduct a meta-ethnography on the experiences and perceptions of children with chronic pain and their families of chronic pain, treatments and services to inform the design and delivery of health and social care services, interventions and future research. We define a 'child,' according to the UN Convention of the Right of a Child (UNCRC), as a person under 18 years of age.

Review questions (RQs):

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

Objectives:

1. To conduct comprehensive searches to identify qualitative research literature on the experiences and perceptions of children with chronic pain and their families to address RQs 1 to 4
2. To select relevant studies and synthesise them using meta-ethnography
3. To ensure salience of findings via involvement of children with chronic pain and their families in study design, analysis and interpretation

4. To assess how much confidence can be placed in our synthesised findings using GRADE CERQual (47) in order to facilitate use of our findings for NHS decision making
5. To identify research gaps regarding RQs1-4 in order to inform future research directions
6. To integrate our findings with existing relevant Cochrane treatment effectiveness reviews (15-19) in order to determine if programme theories and outcomes of interventions match children and their families' views
7. To 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.
8. To disseminate findings to academic, clinical, lay and policy audiences to influence childhood chronic pain policy and practice.

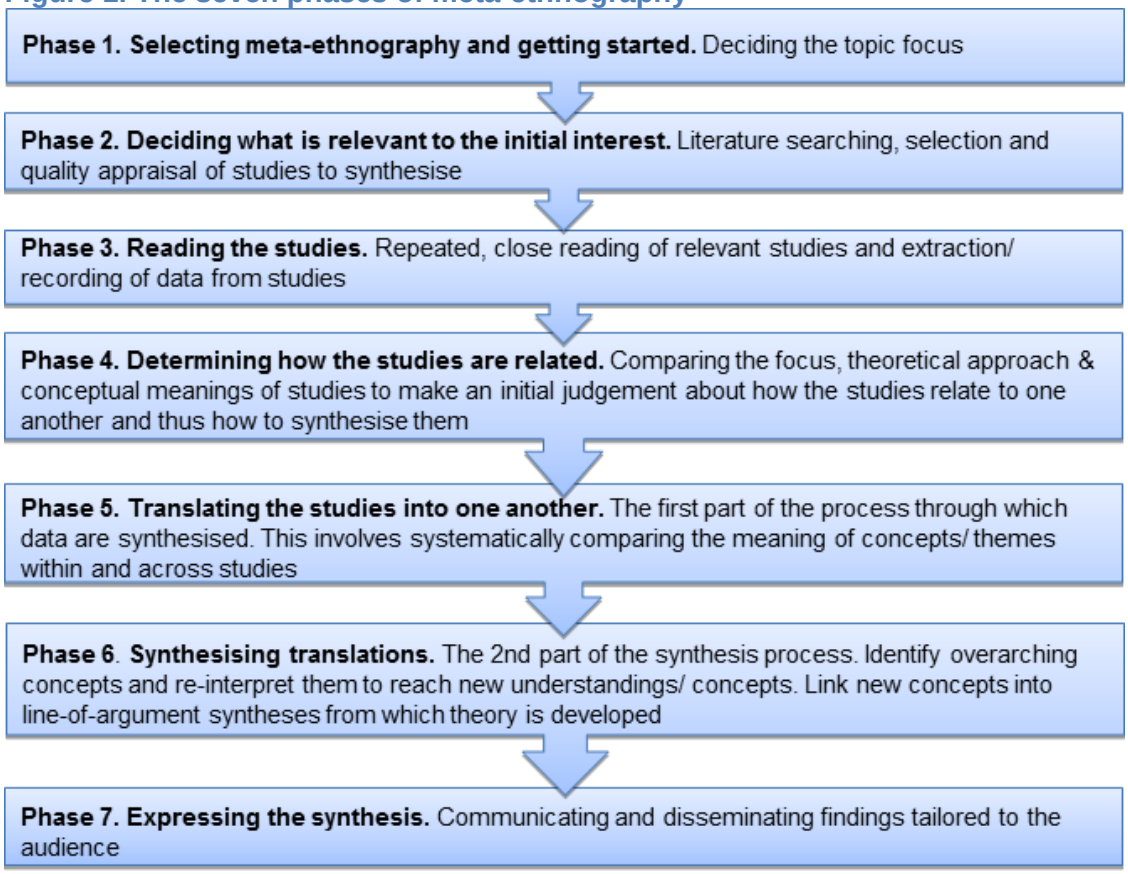
# 3. Research plan/methods

## 3.1 Design

We will conduct a meta-ethnography (29) following the eMERGe meta-ethnography reporting guidance (30, 42-44) and Cochrane Qualitative Implementation Methods Group (QIMG) guidance (32). We have registered our review protocol on PROSPERO, the International Prospective Register of Systematic Reviews (reference: CRD42019161455), we will publish it in an academic journal (see section 4.1) and we have registered it with Cochrane PaPaS (review number 623). We have finalised the literature search strategy in collaboration with our PPI group (see PPI section). Meta-ethnography (29) is ideally suited to informing complex healthcare issues like childhood chronic pain in order to improve services (31). It can be used to develop important new conceptual understandings and theory about how patients experience, understand, and perceive health and illness, interventions, health services, policies and strategies, and inform identification of outcomes of value to children and families. It is the only qualitative evidence synthesis methodology to interpret the conceptual data, e.g. concepts, from existing accounts of primary qualitative studies (e.g. those using in-depth interviews) in order to develop novel conceptual insights which were not apparent in any single study (29, 30). It does not involve simply aggregating findings (29).

Meta-ethnography (29) is a systematic, interpretive qualitative evidence synthesis methodology that can produce robust, novel synthesised evidence for the design and delivery of interventions and services for managing childhood chronic pain. Meta-ethnographies have greater transferability of findings than individual qualitative studies, hence more weight in the evidence hierarchy (48) meaning they have potential to inform - and indeed have informed - clinical guidelines (e.g. 49, 50), service design, and patient-centred care. Meta-ethnographies can also indicate gaps in knowledge, e.g. a lack of conceptual development in a field, and identify new directions for research (31). Meta-ethnography has a unique analytic synthesis method involving systematically comparing the meaning of concepts from primary studies, identifying new overarching concepts, and linking these into one or more 'line of argument' syntheses leading to theory development (31, 51). It is a rigorous, inductive methodology which takes into account the contexts and meanings of the original studies (29) making it ideal for synthesising the diverse contexts of children's chronic pain research. The seven phases of meta-ethnography (29, 30, 42, 45, 46) are described in Figure 1 and, although presented linearly, some phases run in parallel and the process is iterative.

**Figure 1. The seven phases of meta-ethnography**




## 3.2 Search strategy

### 3.2.1. Phase 1 'Selecting meta-ethnography and getting started'

Scoping searches of bibliographic databases were conducted using search terms similar to those given in Box 1 to indicate the volume and suitability of the literature to address our RQs. They indicated a reasonably-sized body of literature from which to sample items for synthesis and that conceptually rich studies, e.g. (26, 27, 52), suitable for meta-ethnographic synthesis exist to address RQs 1 to 4.

### 3.2.2. Phase 2. 'Deciding what is relevant to the initial interest'

A rigorous search for published and unpublished ('grey' literature) studies will be conducted via bibliographic databases and forensic searches, as outlined below. Grey literature will be included 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 meta-ethnography, the 'worth' of study is determined by its contribution to the synthesis (29)(p.17) - lower quality studies will contribute less than higher quality studies to the outputs, regardless of their peer-review status (see quality appraisal of relevant studies in section 3.3.1.2). RT will lead the design and conduct of literature searches assisted by the research fellow (RF).

#### 3.2.2.1. Bibliographic database searches

We will search 14 bibliographic databases selected for their good coverage of qualitative research and spectrum of relevant disciplines – see Table 1. We will also hand-search for articles published in the last 24 months in key journals chosen for their relevance to our research questions and which publish qualitative health research, such as Sociology of Health and Illness, European Journal of Pain, Clinical Journal of Pain, Journal of Pediatric Psychology, BMC Pediatrics,

Qualitative Health Research, and Social Science and Medicine, to identify any articles not indexed yet in databases.

**Table 1. Bibliographic databases to be searched**

| Discipline/type of literature | Databases |
|---|---|
| Health and social care | CINAHL<br>Child and Adolescent studies<br>EMBASE<br>MEDLINE<br>MEDLINE in Process<br>Social Care Online (scie) |
| Psychological | PsycINFO |
| Sociological | Social Sciences Citation Index |
| Education | British Education Index |
| Multidisciplinary | Scopus |
| Grey literature and theses | HMIC<br>OpenGrey<br>EtHOS |

The database search strategy for MEDLINE is presented in Box 1. The search strategy combines three key search concepts: (A) qualitative study designs; (B) population – children and their families children; and (C) phenomenon of interest - chronic pain. The strategy has been informed by existing reviews that represent good practice for identifying the study design, population and/or phenomenon (7, 21, 53). The strategy will be finalised by further testing and refinement against a set of key articles known to fit the inclusion criteria, and then adapted to the functionality of each database listed.

### 3.2.2.2. Forensic searches

We will also conduct iterative supplementary or 'forensic' searches (54): citation tracking, contacting experts, and searching websites of key organisations, e.g. The British Pain Society, Department of Health, NIHR Library, the Sickle Cell Society, Versus Arthritis, CRPS (Complex Regional Pain Syndrome) UK, Fibromyalgia Action UK, Crohn's & Colitis UK, Reflex Sympathetic Dystrophy Syndrome Association (RSDSA), The European League Against Rheumatism (EULAR) network, European Pain Federation, Pain Relief Foundation, Children's Health Scotland, Royal Hospital for Sick Children (NHS Lothian) and others. 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 (55).

**Box 1. Draft MEDLINE Search Strategy**

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. (adolescen* or preadolescen* or baby or babies or infan*2 or 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 cramping)).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.

## 3.3 Review strategy and strategy for reviewing literature

### 3.3.1 Literature screening and selection

One reviewer will conduct initial screening of retrieved references by title to exclude off-topic texts, i.e. those clearly not about childhood chronic pain. Following initial piloting and standardisation

between reviewers, each reference will be independently screened by title, abstract and then full text by two reviewers (the research fellow and RT) to assess their relevance using Covidence systematic review management software (www.covidence.org). Disagreements will be resolved through discussion and then referred to a third reviewer, if necessary (EF,IU, JN).

Draft inclusion and exclusion criteria are below - the final criteria will be decided with our PPI group and may also be revised during analysis phases (see PPI section 7). As relevant articles are identified and read, decisions will be made, in collaboration with our PPI group, to refine inclusion and exclusion criteria to ensure a focused, manageable and meaningful synthesis can be conducted to answer our research questions. In this way, selecting texts for synthesis will be an iterative process; this is appropriate for meta-ethnography the purpose of which is to build understanding and theory, rather than produce a definitive conclusion about effectiveness of an intervention (56, 57).

Draft inclusion criteria:

- Published or grey literature, i.e. peer-reviewed journal articles, published reports, book chapters, books, PhD theses
- Contains qualitative research data on chronic pain, i.e. 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 (e.g., focus group discussions, individual interviews, observation, diaries, document analysis, open-ended survey questions) and analysis (e.g. thematic analysis, framework analysis, grounded theory)
- In any language

Draft exclusion criteria:

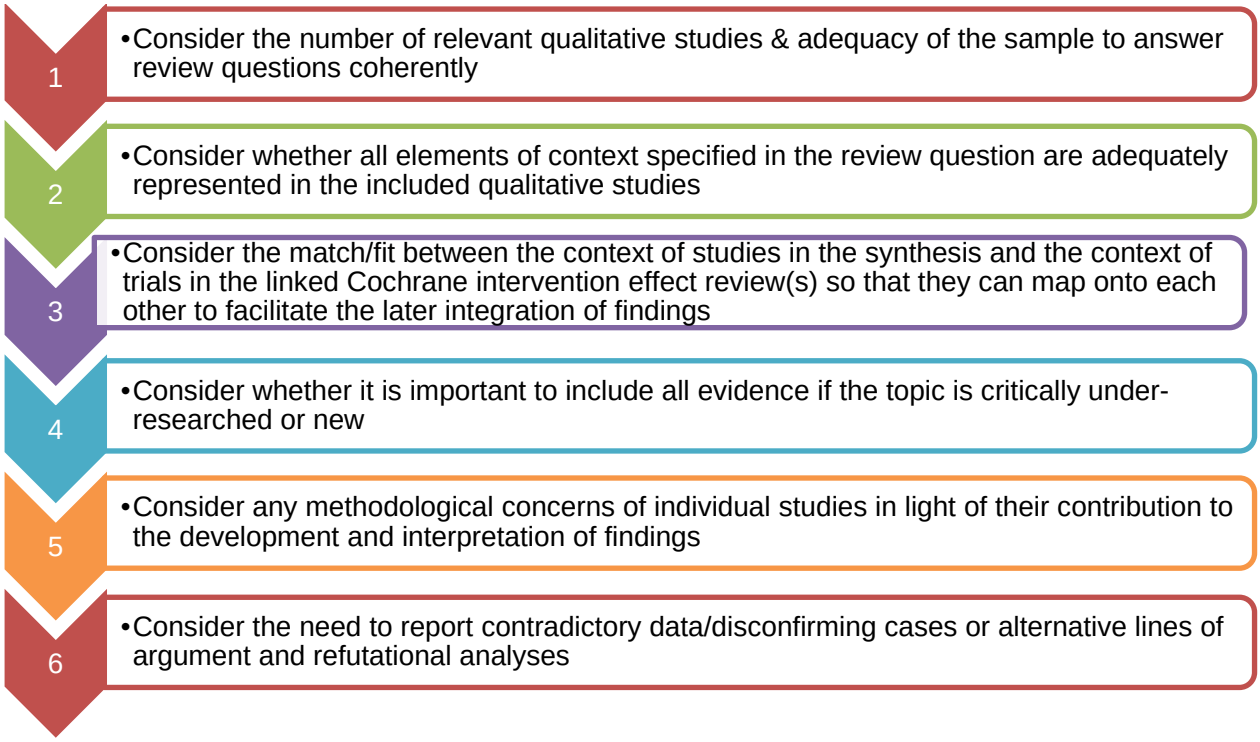
- Not qualitative design or not a mixed methods design including qualitative data
- Not reporting views of children with chronic pain or their family members
- Acute pain i.e. pain lasting for less than 12 weeks, such as that caused by medical procedures
- Cancer pain
- Pain in neonates and babies <3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

To answer research question 3 on how children and families view services we will select studies conducted in the UK. To answer research questions 1, 2 and 4 on how children and families conceptualise and live with chronic pain and conceptualise good pain management, we will focus mainly on UK studies. However, international studies, e.g. from economically developed countries, might indicate new service models (research question 4) we could use in the UK. Findings regarding how children and their families conceptualise and live with pain (research 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 2 in section 3.4.2). Decisions will be made in light of the characteristics and content of the whole body of relevant studies and with our PPI group.

#### 3.3.1.1. Sampling

Sampling from the body of relevant literature is an iterative process. In Phase 2 there will be an initial selection of relevant studies which meet the inclusion criteria, as described in Section 3.3.1 above. This will be followed by further purposive sampling of relevant studies to ensure we conduct a focused, meaningful synthesis which answers our research questions (56, 57). 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 RQs (58) - see Figure 2 for QIMG key assessment criteria. For example, we might revise our inclusion criteria, e.g., to include or exclude studies in which adults give retrospective accounts of their own childhood chronic pain. We will document the reasons for 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. The number of studies it is possible to synthesise is dependent on the volume of relevant data studies contain relative to team resources (45). Prior high quality meta-ethnographies following the key principles of the original methodology have been conducted successfully on 40 (31) to 50 studies (59).

**Figure 2. Key criteria to consider when selecting studies to synthesise, adapted from Noyes et al (58)**




#### 3.3.1.2. Quality appraisal of relevant studies

Two independent reviewers will quality appraise relevant studies using the Critical Appraisal Skills Programme (CASP) qualitative tool (60), which meets Cochrane criteria. As part of the appraisal process we will judge the conceptual richness of the primary studies, i.e. whether the findings are explanatory rather than just descriptive. We will select rich studies for inclusion (45). We will not exclude studies which are limited by poor methodological reporting because there is a distinction between quality of methodological reporting and quality of output/findings, but we will exclude studies which are judged to be fatally flawed, e.g. methodologically unsound. If a team member is the author of a relevant study they will not be involved in quality appraising it to ensure an unbiased appraisal. We will transparently record all decision making and reasons for study exclusion. Results of quality appraisal will inform CERQual judgements of how much confidence can be placed in our synthesised findings (see section 3.4.5).

## 3.4. Data extraction, analysis & synthesis – meta-ethnography phases 3 to 6

### 3.4.1. Phase 3 Reading the studies

Studies will be read in full and repeatedly by at least two team members (all members will read some studies). 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 (61). 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. As analytic phases overlap, reading is not a one-off activity.

### 3.4.2. Phase 4 Determining how studies are related

We will determine how the studies relate to one another by comparing their aims, focus, characteristics and findings. Then we will logically order 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. infants, ≤5 years old, 6-12 years, 13-16 years, 16-18 years), and synthesise each group of studies separately before synthesising them all together (31). This approach has been used successfully in quality meta-ethnographies (31) and enables synthesis of diverse studies. The precise method for grouping studies can 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 (45).

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) (45). Refutational data are important for developing comprehensive understandings and theory building (62). Including some studies from outside the UK could be an important element of identifying similarities and differences in the conceptualisation of chronic pain and what 'good' chronic pain management looks like among different ethnic, national and cultural groups residing in the UK. Where studies report gender/gender differences we will explore gender differences in the views and experiences of children and their parents.

### 3.4.3. Phase 5 Translating the studies into one another

Similar to the constant comparison method of grounded theory (63), concepts from studies are compared systematically to identify the range of concepts and whether their meanings are similar or contradictory. Concepts alike in meaning are matched and merged. We will use a method similar to that described by Campbell et al (31) which compares concepts one by one, study by study in a logical order, e.g. chronologically, for each grouping of studies. This method has an advantage over other methods (45): 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 and Hare's (29) original method. The process of translation is key to conceptual interpretation and synthesis in meta-ethnography so it is important to adhere to the principles of translation (45). We will 'translate' each group of studies separately before synthesising across groups (31).

### 3.4.4. Phase 6 Synthesising translations

We will identify and further interpret overarching concepts to reach new interpretations which we will link in one or more 'line-of-argument syntheses' which we will develop into an explanatory theory. An example of a possible theory we will produce 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, Phases 5 and 6 will involve at least three team members (primarily EF, AJ, IU and the research fellow) with input from the wider team. Six to eight young people with chronic pain and parents will participate in a data analysis and interpretation workshop (see PPI section for details). 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.

### 3.4.5. Assessing confidence in synthesised findings

To facilitate the use of our findings for decision making by the NHS and policy-makers, we will apply the GRADE CERQual tool to our meta-ethnography findings (47), guided by JN who co-developed CERQual. Using CERQual, the overall confidence in the synthesised evidence for each finding is evaluated according to its adequacy, coherence, relevance and the methodological limitations in the primary studies (47), drawing on the results of our quality appraisal of studies.

### 3.4.6. Integration with Cochrane reviews

We have registered our meta-ethnography with Cochrane PaPaS (review number 623). It will be important for decision-making to develop an overall understanding of intervention effect, feasibility, acceptability and factors that create the context for barriers and facilitators to successful implementation. We will therefore integrate our qualitative findings with the results of five recent Cochrane intervention effectiveness reviews (15-19) using an appropriate quantitative/qualitative data integration method from Cochrane QIMG (64) to determine if the programme theories and outcomes of interventions match families' views and expectations. 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.

#### 3.4.6.1 Integration mechanisms for quantitative results and qualitative findings

There are various points in overall meta-ethnography production at which integration can occur (64, 65). We have/will integrate during review question formulation and 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 recent Cochrane reviews (see, for example, (66)). 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.

## 4. Dissemination, outputs and anticipated impact (Phase 7)

Our dissemination plan was developed with PPI input. We will disseminate findings to academic, lay, clinical and policy audiences throughout the project. Incorporating a range of stakeholders from the project outset will facilitate dissemination activities and impact. Specific planned outputs for each audience are listed below, along with how these will be communicated/ disseminated.

### 4.1. Outputs

**Academic outputs:** We will publish: the study protocol in an academic journal (e.g. *BMJ Open*) and via the PROSPERO register, a final report in the NIHR HS&DR journal including a Cochrane review report integrating the meta-ethnography findings with existing relevant Cochrane reviews, and two open-access journal articles reporting the meta-ethnography findings, e.g. children and families' views of treatments and health services in the *European Journal of Pain* and practice implications of families' theories of chronic pain in S*ocial Science and Medicine*.

**Lay outputs:** We will produce a lay podcast of study findings and their implications for Pain Concern Radio; host a 'Twitter chat' - a scheduled, organised, public Twitter conversation via a pain charity (audience not limited to lay people). Specifically for children, we will create a three-minute animated film of findings and implications for sharing via social media including YouTube/YouTube Kids. We will inform the updating of pain charities' existing resources for patients and the public.

**Clinical outputs:** We will present to the Scottish Paediatric Pain Action Group (SPPAG), a group of multi-specialty clinicians from all the main centres around Scotland that deal with children's chronic pain and equivalent networks in England, e.g., the Paediatric Chronic Pain Network organised by Great Ormond Street Hospital. We will create an infographic of key findings for health and social care professionals to be distributed to chronic pain services and networks including specialist hospitals such as Great Ormond Street, Royal United Hospitals Bath; Chronic Pain Managed Clinical Networks e.g. Managed Clinical Network - Chronic Pain West of Scotland; the Faculty of Pain Medicine - Royal College of Anaesthetists; child and adolescent mental health services; and institutions delivering training courses that specialise in working with families and children where there is chronic illness/chronic pain, e.g. The Tavistock, London, and postgraduate programmes in pain management e.g. UCL, University of Cardiff, University of Edinburgh. The infographic will also be made available online.

**Policy outputs:** We will develop a policy briefing of key findings to share with relevant government all-party/cross-party groups and governmental organisations, e.g. the Department of Health and Social Care, the Scottish Parliament Information Centre (SPICe), The House of Commons Library, the National Advisory Committee for Chronic Pain.

**Multi-audience outputs:** a webinar for clinicians, families, policy-makers and other professionals; a Twitter chat (described above).

### 4.2. Dissemination routes

We will disseminate outputs via a wide variety of routes, many with multiple audiences including: a project webpage; websites of our institutions and partner PPI organisations; university press releases; social media e.g. Twitter, YouTube; our professional networks; the Scottish Parliament cross-party group on chronic pain (LC is a member); the NIHR Dissemination Centre; the National Centre for Population Health and Wellbeing Research, Wales, which has a focus on children; the PROSPERO register; the Cochrane Qualitative and Implementation Methods Group (JN and EF are members); the NMAHP Research Unit quarterly newsletter, sent to all NHS Boards in Scotland; CHAIN (Contact, Help, Advice and Information Network) online network of 13,000+ practicing health care professionals, researchers and educators; academic mailing lists e.g. our list www.jiscmail.ac.uk/META-ETHNOGRAPHY, paediatric pain listservs; the Scottish School of Primary Care network; research bulletins of the Royal Colleges of Nursing, of General Practitioners, and of Paediatrics and Child Health; the networks of our advisory group members who have links to, for example, guideline development groups, and government and patient groups. We will present at 3 conferences with academic, clinical and policy audiences: the British Pain Society Annual Scientific Meeting (co-presenting with PPI group members), The International Symposium for Pediatric Pain, and The International Institute for Qualitative Methodology's Qualitative Health Research annual conference. We will meet with NICE and SIGN clinical guideline groups to present our findings to them.

## 5. Project Management

The study will be sponsored by the University of Stirling. EF, the chief investigator, will maintain oversight of the whole study. The project group, including all study co-applicants and the research fellow and chaired by EF, will oversee the study. The project group will have monthly meetings,

half via teleconference and half face-to-face. Interim Skype/conference call meetings will be held as necessary during crucial phases of the study, e.g. when producing outputs from the study. In addition, to monitor study progress EF will communicate weekly with the co-applicants involved in the current phase of the study and the research fellow. An experienced research fellow will be employed half-time for 4 months and full-time for 20 months to undertake the day-to-day tasks involved in conducting the study. They will be based at the University of Stirling and will be line managed by EF. The research fellow will also lead on PPI co-ordination.

We will form a Project Advisory Group (PAG), comprised of children with chronic pain and their family members, health professionals and other stakeholders, which will advise the project team on four key areas: 1. methodological issues, 2. clinical and lived experience of chronic pain, 3. study conduct, 4. project dissemination. The PAG will have two independently chaired meetings with the research team in January or February 2021 and February 2022. We will recruit up to 6 patients/and their family members (2 parents already recruited) as well as patient representatives from the third sector including from Pain Concern, Children's Health Scotland and from NHS and government e.g. Healthcare Improvement Scotland. We have already recruited 4 clinicians/academic clinicians with expertise in chronic pain (Dr Ewan Wallace, Dr Jeremy Gauntlett-Gilbert, Professor Blair Smith, and Professor Lesley Colvin) - see section 9 for further membership details. We will continue to recruit and ensure we have wide representation. Incorporating a range of stakeholders from the project outset will maximise the likelihood that the research will be acceptable and relevant to children and families and health professionals. The PAG chair will be independent from the co-applicants and their institutions with the necessary skills to act as chairperson.

We will comply with NIHR requirements for 6-monthly progress reports. In the interim the research team will monitor budgets and progress supported by our institutions. The proposed monitoring will ensure that a high quality study is delivered on time and within budget.

## 6. Ethics / Regulatory Approvals

This is an evidence synthesis, no primary research will be conducted, therefore ethical and regulatory approvals are not required. The research findings to be synthesised are in the public domain; no permission is required from authors to synthesise their work. There may, however, be ethical issues connected to the impact of the research findings of the study, for example, if they lead to new care pathways or approaches to pain management. We anticipate that such changes to healthcare will improve care rather than cause distress or harm to patients and their families.

Patient and public involvement in research does not require ethical approval as stated by INVOLVE (www.invo.org.uk). Nonetheless, we will conduct our PPI ethically to be sensitive and responsive to patient and families' needs and wishes. We will agree with our PPI group at the outset how their intellectual contribution will be recognised in project outputs. Participants will be briefed prior to PPI sessions. PPI group members might become upset if the project material causes them to reflect on any of their experiences related to service use or illness that had been upsetting. At the end of each session the project team will lead a debrief to address any emotional reactions to the proceedings. After meetings the research fellow will offer a follow-up phone call the next day to each family to check on their emotional well-being. We will also provide the PPI group with a list of potential sources of support. The group will be encouraged to ask questions at any timepoint during their involvement in the study. Several of the project team have backgrounds in psychology and health professions and working with children. In addition the CI has experience of conducting PPI with people with severe and enduring mental health needs.

To accommodate the age and health status of child PPI participants and avoid burdening their families, PPI sessions will be as short as possible. We will use accessible venues with comfortable seating in central locations. Overnight accommodation will be provided if required. Online sessions will allow members to participate from the comfort of their own homes. All sessions will include

regular breaks with the option for unscheduled breaks or discontinuing their participation, as required. Further information on our ethical approach to PPI is described in more detail in section 7.

# 7. Patient and Public Involvement (PPI)

Ten individual lay people – 3 children with chronic pain and 1 with a long-term condition, 4 parents, and 2 adult lay members of a university PPI panel have been involved in developing this proposal as well as two patient organisations, Children's Health Scotland and Pain Concern. Our PPI approach is informed by and will follow the new UK National Standards for public involvement in research (67) to include inclusive opportunities, appropriate collaboration, provision of support/learning, clear communication, defining roles and responsibilities, governance and appropriate financial reimbursement. PPI group members will be able to direct their own level and focus of involvement throughout the study. We will strive to create an environment which is inclusive, recognises the contributions of all participants, and in which people work together to achieve a shared understanding. Funding has been requested for all PPI activities, to ensure that their time is reimbursed for project activities, undertaking training and dissemination and for out-of-pocket expenses.

## 7.1. Aims of active involvement in the project

PPI at the outline stage helped to ensure the study aim, review questions and outcomes are important for patients and their families and that the lay summary is appropriate. For this stage 2 proposal, they also gave feedback on our PPI and dissemination plans. We aim to collaborate with and consult our PPI group during 2 workshops and interim communication, e.g. via email, teleconferences, and/or social media such as a private Facebook or Whatsapp group, depending on members' preferences. Table 3 shows the key aspects of the project we envisage the PPI group will be involved in. Individual members may choose to get involved in only certain aspects but will have the option to be involved across all phases. We will record the impact of their contributions using the GRIPP2 reporting guideline for PPI in research (68).

**Table 3. PPI in different stages of the meta-ethnography**

| Phase | Activity | Level of involvement | Method of involvement |
|---|---|---|---|
| **Planning of Proposal & Phase 1** | Feedback on study aims, objectives, review questions, lay summary & dissemination strategy | Consultation | Email |
| **Phases 1 & 2** | Finalise the study protocol e.g. the literature search strategy | Collaboration & consultation | Teleconference, email, online |
| **Phase 2** | Finalise inclusion/exclusion criteria e.g. the types of chronic pain included, the characteristics of the population we will include. Sample studies for synthesis | Collaboration & consultation | Online workshop 1, March 2021 |
| **Phases 3 & 4** | 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 |
| **Phases 5 & 6** | 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 |

| | | | |
|---|---|---|---|
| **Phase 7** | Producing outputs, dissemination.<br>We will invite 2 members to co-present a conference paper and the group to co-develop lay, patient, and policy outputs. The group will help ensure 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 1 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 (69) for involving users in systematic reviews calls this the PPI group 'influencing' the research.
'Collaboration' refers to when the children and families will be involved in doing the research as well as setting priorities and making decisions. The ACTIVE framework (69) calls this the PPI group 'controlling' the research.

## 7.2. Description of patients and the public to be involved

Children with chronic pain aged 8 to 18 and parents and informal carers (i.e. not clinicians) of children aged 3 months to 18 years will take part as experts in the experience of living with chronic pain. We will aim to recruit parents/carers of children under 8 years because the nature of the tasks/activities, e.g. reading, is less suited to younger children's developmental stage. We aim to involve 6 to 8 children and young people with chronic pain aged 8 to 18 years and parents/family members of children aged 3 months to 18 years (~16-20 people) in the PPI group. They can also choose to join a Project Advisory Group (PAG).
We will recruit a diverse sample to represent a wide range of experiences, ages, ethnic groups, socio-economic statuses and types of chronic pain including children with primary (e.g. fibromyalgia) and secondary pain conditions (e.g. arthritis) via adverts distributed through pain and children's health charities, social media, and NHS services across the UK. We will also include patient representatives from Pain Concern and Children's Health Scotland, and other third sector organisations (to be recruited).

## 7.3. Methods of involvement

We will be flexible 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 intend to use a combination of 2 workshops (either online or face-to-face in a central location, e.g. London, depending on the current Government guidance and restrictions on face-to-face meetings ), , and interim online communication (email, teleconference calls, social media e.g. a private Facebook group, depending on their preferences) across the life of the project. We will tailor meetings to the children's needs e.g. short duration, frequent breaks. If attending workshops proves challenging or unappealing to children and their families, we will explore taking the research out to them. The project team will 'lead' i.e. initiate the meta-ethnography and have lead responsibility for its conduct and completion, in the terminology of the ACTIVE framework for involving users in systematic reviews (69). The PPI group may contribute to: finalising the study design, deciding which studies to include and how to organise them for synthesis, sharing their experiences and comparing findings with their experiences, identifying important areas missing from existing research, dissemination of findings including deciding the content of outputs.

We will use creative means of involving children in the research, e.g. visual and interactive methods, such as those used for conducting research with children (70). For our analysis workshop, to get children involved in informing interpretation of the studies, we will present key themes from study findings in a fun, interesting way according to their preferences, e.g., an improvised play or drama, short stories or written 'vignettes', and/or visual images, and invite them to share their relevant experiences.

### 7.4. Training and support for PPI group

There is little guidance on involving children in systematic reviews/ evidence syntheses (71), so we will draw on and adapt existing guidance on their involvement in primary research (70). We will survey the training and support needs of our PPI group members prior to commencing PPI activity. Training for PPI members will be tailored to their needs and will evolve as the project progresses in line with their wishes. Support will be provided and PPI members will have the research fellow as their named contact. To support and enable full involvement, we will develop and regularly update a project 'jargon buster'; collate existing online resources, e.g. Cochrane Training online which explains evidence-based healthcare and systematic review processes and YouTube presentations on qualitative evidence synthesis. Pre-existing resources should be suitable for parents and young people aged 12+. AJ and LC have experience of engaging children and young people with chronic pain in research. AJ and LF have won awards for public engagement and involvement respectively. EF has experience in explaining and developing resources for lay audiences on systematic reviewing and meta-ethnography through the prior eMERGe project she led.

All PPI members will be provided with feedback on their contributions, following published guidance (67, 72). 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 (www.invo.org.uk).

## 8. Project/ research expertise

Our multi-disciplinary research team, which has a history of prior successful collaboration, has extensive expertise and in-depth understanding in children's health, chronic pain, meta-ethnography, systematic reviewing and qualitative research. We have backgrounds in sociology (IU, RT), psychology (EF), health psychology (AJ, LC), family therapy (LF), nursing children with chronic pain (JN) and development of evidence synthesis methodology (JN, RT), children's pain research (AJ, LC) and health services research (all) and a track record of effective collaborations and successful grant management. Our team of competent, credible experts is experienced in conducting meta-ethnographies and other qualitative evidence syntheses and research on children's chronic pain. We are capable of delivering a robust methodology, interpreting qualitative data on children's chronic pain, and complex analysis related to the advanced qualitative methodology of meta-ethnography.

The team includes input from PPI representatives (see section 7) to ensure acceptability to children and families, and clinicians to ensure relevance to clinical practice. Expert advisors: Dr Ewan Wallace, consultant in paediatric anaesthesia and pain medicine, NHS Greater Glasgow and Clyde, leads chronic pain clinic at Yorkhill Children's Hospital; Dr Jeremy Gauntlett-Gilbert, senior clinician and research lead at Bath Centre for Pain Services; Prof Blair Smith, clinical academic at University of Dundee, consultant in Pain Medicine and National Lead Clinician for Chronic Pain, Scottish Government, Vice Chair of the National Advisory Committee on Chronic Pain; Prof Lesley A. Colvin, Chair of Pain Medicine, Honorary Consultant in Anaesthesia and Pain Medicine, University of Dundee; chair of the 2018 SIGN guideline Development Group for children's chronic pain. Further expert advisors will be recruited.

### 8.1 Individual contributions of co-applicants

**Emma France** (30% FTE, 24 months): chief investigator. A leading methodological expert in meta-ethnography, led development of NIHR HS&DR funded eMERGe project to develop meta-ethnography reporting guidance. Expertise in psychosocial aspects of long-term/life-limiting conditions in children. Specialises in qualitative methods. Skills in non-Cochrane systematic review and mixed methods research including intervention development. Academic and commercial

project management skills. Experienced in producing audio-visual research outputs and use of social media. Responsibilities: overall project management and administration; manage research team including a research fellow (RF); key active role in analysis and interpretation, leading analysis phases 3-6 and screening, selecting, appraising, data extracting and co-analysing publications; assist with recruitment and involvement of project advisors and PPI group; present at conferences. Overall responsibility for project outputs and PPI.

**Jane Noyes** (10%, 24 months): Professor in Health and Social Services Research and Child Health. Specialises in qualitative evidence synthesis and methodology. Lead convenor of Cochrane Qualitative and Implementation Methods group, Member of the Cochrane Methods Executive and Scientific Committee, and major publisher of meta-ethnographies as editor of Journal of Advanced Nursing. Will give specialist methodological input; contribute to study screening and selection, data analysis and interpretation, dissemination; and provide expertise in applying CERQual, integrating findings with Cochrane reviews, and children's nursing.

**Isabelle Uny** (10%, 24 months): sociologist with expertise in meta-ethnography (30, 42-46, 73, 74) and other qualitative evidence synthesis methodologies, grounded theory (relevant for meta-ethnography conduct) and qualitative research. Was main researcher on eMERGe project to develop meta-ethnography reporting guidance. Will have active role in analytic synthesis - will contribute to study screening, selection and appraisal; data extraction, analysis, interpretation; and dissemination.

**Ruth Turley** (10% FTE months 1-4; 3% FTE months 5-24): freelance researcher and former research fellow/ systematic reviewer in DECIPHer (Development and Evaluation of Complex Interventions for Public Health Improvement), associate editor and former searching coordinator for Cochrane Public Health Group. Lead/contributor of qualitative evidence synthesis to 3 NICE guidelines including child health. Involved in prior eMERGe project. Will lead the identification of eligible studies, literature searches and study screening with support from the RF, contribute to data analysis, interpretation, and dissemination.

**Abbie Jordan** (7%, 24 months): Health psychologist specialising in children's chronic pain, parenting and qualitative methods including qualitative evidence synthesis (8). Leads the pain in child theme of the Bath Centre for Pain Research, an international centre of excellence for paediatric pain research. Holds current research grants to examine issues around social development, identity and mental health in young people with chronic pain and their parents. Won 'I'm A Scientist' 2018 award for public engagement in the area of childhood. Experienced in hosting Twitter Chat. Will contribute expertise in children's pain; have active role in data extraction, analysis and interpretation; advise on/assist with engagement of children in research; and dissemination.

**Line Caes** (3%, 24 months): health psychologist with expertise in the social context of children's pain experiences, e.g. functional abdominal pain, inflammatory bowel disease. Member of Scottish Parliament chronic pain cross-party group. Reviewer of 2018 SIGN guideline on children's pain. Led bibliometric analysis of children's pain research published from 1980-2010 (75). Current research evaluates experiences of childhood chronic pain and treatment from a family perspective. Quantitative and mixed methods skills. Experience in involving young people with chronic pain in research. Will contribute expertise in children's pain and bring a quantitative perspective; contribute to data analysis and interpretation, and dissemination.

**Liz Forbat** (3%, 24 months): research psychologist and family therapist with expertise in physical health. Provides family systems expertise on the systemic impact of serious ill health on families and wider networks. Qualitative researcher, expertise in grounded theory (relevant for meta-ethnography conduct) and discourse analysis. Will contribute expertise in family systems and ill health; PPI methods; data analysis and interpretation, and dissemination.

All co-applicants will attend project meetings, contribute to all project outputs including an NIHR report and to dissemination including conference presentations and journal articles.

## 8.2 Contributions of other staff

**Research Fellow** (RF) (50% FTE, month 1; 100% FTE, months 2-24): conduct day-to-day project tasks, key role in Phases 2-7. Will conduct literature searches with RT in months 1-4, do the majority of study screening, data extraction, and appraisal with close involvement of EF. The RF will collaborate with the wider team to achieve the project outputs. Their continuous involvement over 24 months will ensure the successful day-to-day running of the project. They will lead on PPI activities with support from the team. We will seek to recruit someone with experience of meta-ethnography and/or interpretive qualitative methodologies compatible with meta-ethnography e.g. grounded theory methodology. Meta-ethnography is a complex methodology so the experience needed is commensurate with post-doctoral level.

**Secretarial support** (10%, 24 months): assist with organisation of 14 project and PAG meetings, conference calls, 2 workshops and PPI group training, routine correspondence, other project administration. It is more cost-effective for these tasks to be done by a secretarial grade with the relevant skills and expertise than by the project team.

The research fellow and the secretary will be based with the chief investigator within the Chief Scientist Office Nursing, Midwifery and Allied Health Professional Research Unit (NMAHP-RU) at the University of Stirling where they will be in day-to-day contact and have more formal weekly project supervisory meetings. University of Stirling has an annual formal appraisal system for all staff that line managers conduct. LC will be located in psychology and IU in the Institute of Social Marketing and supervised through their management structures.

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## Version Control Table

| Version number | Purpose/change | Author | Date |
|---|---|---|---|
| V1 | Minor changes: added reference section, PROSPERO registration number, updated information on Cochrane PaPaS registration, updated reference #70, removed staff salary/grade information, impact and 'risks' section compared to the NIHR-approved 'detailed research plan Version 2'. Added further detail from the approved PPI section of the full final proposal. | Emma France, chief investigator | 16/12/2019 |
| V2 | Details updated, most due to change of start date & change of institution of a co-applicant:<br>-recorded new bibliographic databases/ changes of some databases due to lack of access since RT is now freelance.<br>-Recorded RT's reduced time contribution to the project & the research fellow's increased time.<br>-Clarified some systematic review inclusion criteria and exclusion criteria<br>–Removed non-essential material on scoping search findings from methods section.<br>-Minor changes to the search terms following piloting. | Emma France, chief investigator | 03/11/2020 |

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| | -Added Cochrane PaPaS review registration details.<br>-Listed new online sources to be searched (for 'forensic' searches) added after PPI input.<br>-Updated the timeline & meeting dates | | |
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