

Understanding how children and young people with chronic non-cancer pain and their families experience living with pain, pain management and services: a meta-ethnography

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Scientific summary

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Background

Chronic pain in childhood is widespread, affecting at least 8% of children and young people (CYP) in the UK. For a sizeable number of children, chronic non-cancer 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 has an annual cost of around 4 billion pounds in the UK. Provision of, and access to, services for managing children's chronic pain is limited. We also do not know which treatment outcomes children and families value. To design and deliver services and treatments, which meet the needs of patients and their families, it is vital that we understand how children with chronic non-cancer pain and their families experience chronic pain, pain treatments and services, and which outcomes are important to them. Qualitative research addressing these urgent, important issues exists but must be synthesised to interpret the body of evidence. Existing qualitative evidence syntheses have focused on either a narrow chronic pain population or topic and none produced a theory. Therefore, we conducted a qualitative evidence synthesis using meta-ethnography, which is ideal for synthesising the diverse contexts of children's chronic pain research and developing theory. We refer to the meta-ethnography by the acronym 'CHAMPION' (Children And young people's Meta-ethnography on Pain).

The meta-ethnography was conceived as a Cochrane review to inform and supplement existing Cochrane reviews of intervention effectiveness for children's chronic pain. Cochrane requires qualitative evidence syntheses to be integrated with intervention effects reviews to further understanding of patient experiences, acceptability and implementation of interventions, which outcomes are important to children and their families, and inform further intervention development.

Aim

To conduct a meta-ethnography on the experiences and perceptions of CYP 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.

Review questions

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?

Objectives

1. Conduct comprehensive searches to identify qualitative research literature on the experiences and perceptions of children with chronic pain and their families to address review questions 1-4.
2. Select and synthesise relevant studies using meta-ethnography.
3. Ensure salience of findings via involvement of children with chronic pain and their families in study design, analysis and interpretation.

4. Assess how much confidence can be placed in our synthesised findings using GRADE-CERQual (Grading of Recommendations Assessment, Development and Evaluation-confidence in the evidence from reviews of qualitative research) in order to facilitate use of our findings for NHS decision-making.
5. Identify research gaps regarding review questions 1–4 in order to inform future research directions.
6. Integrate our findings with existing relevant Cochrane treatment effectiveness reviews in order to determine if programme theories and outcomes of interventions match children and their families' views.
7. 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. Disseminate findings to academic, clinical, lay and policy audiences to influence childhood chronic pain policy and practice.

Methods

Design

We conducted a meta-ethnography, a seven-phase interpretive methodology, which takes into account the contexts and meanings of the original studies.

Patient and public involvement

Patient and public involvement (PPI) of children with chronic non-cancer pain and their families was fundamental to all aspects of our meta-ethnography conduct from study inception to dissemination. We also worked with a Project Advisory Group of wider stakeholders. The groups approved the aim and design and search strategy. They participated in workshops to decide study inclusion and sampling criteria decisions, how to group studies for analysis, data interpretation, identification of research gaps and to develop outputs. They also informed and assisted with dissemination of findings.

Review strategy

We carried out comprehensive searches of 12 bibliographic databases including MEDLINE (medical literature analysis and retrieval system online), CINAHL (Cumulative Index to Nursing and Allied Health Literature), PsycInfo and 9 others, with iterative supplementary searches, including citation tracking. We searched for studies that had been published up to September 2022.

Selection criteria

To identify qualitative research studies with children aged 3 months to 18 years with chronic non-cancer pain, and their families, which focused on their perceptions, experiences and views of chronic pain, services and treatments. The final inclusion criteria were agreed with our PPI group. We included qualitative research studies of any design using recognisable qualitative methods of data collection, in any language, with no date restrictions. We did not include children with chronic pain related to cancer or end of life because they have distinct care pathways; Cochrane reviews of effects also synthesise non-cancer pain separately.

Data collection and analysis

Eligible studies were purposively sampled to ensure we could conduct an in-depth, synthesis of the data most relevant to answering our review questions. Sampling, organising studies for synthesis and analysis and interpretation involved our PPI group via two workshops and additional communication by teleconference, e-mail and social media. 'Conceptually rich' and 'thick' studies, that is, those with in-depth explanatory/interpretive findings and contextual details, were included. Sampling was guided by Cochrane Qualitative and Implementation Methods Group Guidance, and our advisory and involvement groups who agreed we should include non-UK studies and studies with moderately rich data to ensure a wider range of pain conditions were represented. We assessed methodological limitations of studies using the Critical Appraisal Skills Programme tool. We extracted data on study

aims, focus and characteristics into Microsoft Excel® and findings using NVivo software. We compared these study data to determine how studies related to one another and decided how to order and group them for synthesis. Stakeholders and our PPI group agreed we should group studies by pain condition. We used meta-ethnography to synthesise each group of studies separately before synthesising them all together. Analysis and interpretation of studies involved children with chronic non-cancer pain and their families and has resulted in theory to inform service design and delivery. Our PPI group clarified the meaning of ambiguous study findings and identified and addressed gaps in the data. We used the GRADE-CERQual approach to assess our confidence in each key review finding and used a matrix approach to integrate our findings with 14 existing Cochrane reviews on treatment effectiveness for children's chronic non-cancer pain.

Results

One hundred and seventy studies met inclusion criteria. We sampled and synthesised 43 most relevant conceptually rich and thick studies, which had minor ($n = 24$) or moderate ($n = 19$) methodological limitations overall. Using GRADE-CERQual, we rated 36 descriptive-level review findings as high ($n = 22$) or moderate ($n = 13$) confidence with only one very low confidence.

Twenty-three included studies were conducted in the UK, 17 in other high-income countries and 4 in low- to middle-income countries (there is a three-country study, so the total does not add to 43 studies). Included studies involved 325 CYP with chronic pain, most aged 10 years or older, and 308 family members, mostly mothers. The studies covered chronic pain conditions including complex regional pain syndrome, juvenile idiopathic arthritis, sickle cell disease, headache, migraine, abdominal pain, musculoskeletal pain, epidermolysis bullosa, dysmenorrhoea, neurological conditions such as cerebral palsy, unspecified chronic pain or a mix of conditions.

Moderate and severe children's chronic pain had profound adverse impacts on the whole family, family life and their wider social connections. Pain affected all aspects of an individual's and a family's functioning: family dynamics and relationships; family members' emotions, well-being, autonomy and a sense of self-identity; parenting strategies; friendships and socialising; children's education and future employment prospects; and parental employment. Families experienced difficulties in seeking and getting help and support from health services to manage their child's pain and its impacts. Most pain management occurred outside of health services supervised by parents, especially mothers who were usually the main caregivers.

Children and young people and their families were aware of the biopsychosocial (physical/biological, psychological and social) impacts of chronic pain on their lives. However, most CYP and parents understood the cause of chronic pain as having solely an underlying biological cause. This influenced their expectations of, and interactions with, health services. However, families' conceptualisations of pain were dynamic and could change over time influenced by experiences of treatments, services and interactions with healthcare professionals.

Interactions with healthcare professionals were frequently negative and stressful. CYP and parents, particularly mothers, felt they were not listened to and believed, and thus felt unsupported. For instance, CYP's experiences of living with pain were ignored or their pain was disbelieved, and mothers felt blamed for their child's pain. Parents of younger children and of children with communication difficulties, in particular, perceived that healthcare professionals did not always recognise their expertise in assessing their child's pain.

A key challenge for children and their families was that they found health services fragmented and very difficult to navigate. Consequently, it was difficult to achieve satisfactory pain management. Families often entered a lengthy cycle, which could last many years, of attending then reattending health services,

and awaiting referral. In addition to these navigation challenges, CYP and their families, particularly CYP with sickle cell disease and those with chronic pain and learning disabilities, such as Down syndrome, experienced prejudice and discrimination in health services.

We created a model of how families navigate chronic management with two key routes labelled Path A and B. On Path A, families seek a cure from services but often repeatedly seek services without pain resolution. When their expectations of services are not met, they try to deal with pain alone, often unsuccessfully, which can dominate family life. Psychological treatments are only used as a last option. Path B depicts effective communication and expectation management by healthcare professionals which can help families to shift their focus from a cure to trying to live well despite the pain. Families might also realise independently over time that a cure is unlikely. Families on Path B tend to be more receptive to biopsychosocial approaches for pain management, due to lack of other options. Families can move between the paths depending on input from services and if pain worsens.

We developed new interpretive findings resulting in the family-centred theory of children's chronic pain and its management, which incorporates how children and families conceptualise pain, their experiences of living with pain and of pain management services, and defines 'good' pain management and services. Our mid-range theory describes good pain management and services as holistic, biopsychosocial, family-centred pain management which acknowledges the impact of CYP's chronic pain on all aspects of family life and their social relationships. In the theory, pain management strategies are tailored to the family's needs. Schools, social services, social care and healthcare services function as inter-related aspects in a whole system approach. This approach aims to reduce service fragmentation making services more intuitive for families. Empathetic communication is a core component of this approach, and it is fundamental for building trust between families and services. The theory includes assessment of chronic pain and of families' psychosocial needs to enable effective triage and trigger appropriate actions from/referrals to different services (schools, social care and health services).

Cochrane systematic reviews of intervention effects and trials did not measure some outcomes important to children and families, including effects of chronic pain on the family and resolution of pain. Reviews have mainly neglected a biopsychosocial approach when considering how interventions work.

Limitations

There were few conceptually rich qualitative studies focusing on common pain conditions like migraine, headache, abdominal pain; some rarer conditions; children with learning disabilities and under 5 years old; siblings; boys with chronic pain; fathers; and experiences of treatments/services for chronic pain. Our findings may not well represent the experiences and perceptions of the poorly represented groups and there were gaps in the evidence regarding experiences and perceptions of treatments/services.

Three per cent ($n = 1$) of our findings were assessed as very low confidence, none were low confidence and 36% ($n = 13$) were moderate confidence, using GRADE-CERQual. Additional evidence could increase our confidence in those findings.

We excluded studies focusing on chronic cancer pain, end-of-life pain management and experiences of healthcare professionals.

Conclusions

Currently, children's chronic pain management does not involve holistic, biopsychosocial health and social care tailored for children and integrated with support in the community, which considers the needs of the entire family and the impact on the family life.

The theory and findings developed in this meta-ethnography have the potential to contribute to the development and updating of existing clinical guidelines on children's chronic non-cancer pain; 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 could improve the care provided and thus the health and quality of life of children with chronic non-cancer pain and their families, which in turn could reduce the burden on health and social care services. Findings are being disseminated widely to academic, lay, clinical and policy audiences. Outputs include a short, animated film on YouTube for children (<https://youtu.be/Oz5I7xKYf5o>), academic journal articles, a webinar, a lay podcast, an infographic, a policy briefing and other social media outputs. In addition, we have delivered eight conference presentations.

Future work

Family-centred outcomes should be included in future trials of chronic pain interventions. Conceptually rich, primary qualitative studies are needed to explore families' experiences of services and treatments, including pain management plans, pain assessment, opioid use in children and social care services; experiences of children with autism and learning disabilities, under 5 years old, and with certain common pain conditions, such as headache, migraine and abdominal pain and with some rarer conditions such as Ehlers–Danlos syndrome; experiences of siblings and fathers of children with chronic pain; and healthcare professionals' experiences and perceptions of chronic pain management. A qualitative evidence synthesis on chronic pain management for children with cancer is needed. Development and testing of family-centred children's chronic pain interventions, services and treatments are needed.

Study registration

This study is registered as PROSPERO, the International Prospective Register of Systematic Reviews (reference: CRD42019161455) and Cochrane Pain, Palliative and Supportive Care (review number 623).

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