**Submitted in partial fulfilment of the Lancaster University Doctorate in Clinical Psychology**

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**Doctoral Thesis:**

# A Qualitative Exploration of the Impact of Persistent Pain

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|---|---|---|---|
| Thesis Abstract | 256 | — | 256 |
| Literature Review | 8,000 | 10,771 | 18,771 |
| Research Paper | 7,961 | 6,447 | 14,408 |
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## Thesis Abstract

This thesis offers a qualitative exploration of the experience of living with persistent pain. Section One presents a systematic review and thematic synthesis considering the experience of parenting children with persistent pain. A search identified 17 relevant papers which were appraised for quality and analysed to reveal six themes. These related to seeking control in an uncontrollable situation; being let down by experts and becoming their own expert; fearing judgment whilst judging themselves; seeking normality even whilst adapting to a 'new normal'; focusing on the child versus an awareness of the impact on the wider family; and the conflicting interests in raising a child with persistent pain. The place of these findings within the wider literature is considered, and clinical implications are discussed.

In Section Two, an interpretative phenomenological analysis approach was utilised to explore the impact of complex regional pain syndrome (CRPS) on participants' identity. Six semi-structured interviews were completed and transcribed verbatim. Analysis yielded four themes: the time taken to re-establish an identity; a sense of alienation from others; shame caused by CPRS; and the importance of control to a sense of self. Findings are discussed in relation to the extant literature, and implications for clinical practice are considered, including therapeutic models which may hold potential for this group.

Finally, in Section Three a critical appraisal considered the findings of the two papers, as well as strengths and limitations of the research paper. Elements of reflexivity are explored, as is the impact of COVID-19 upon the thesis. Suggestions for future research are also outlined.

## Declaration

This thesis reports research undertaken between November 2018 and February 2021, as part of the requirements of the Lancaster University Doctorate in Clinical Psychology. The work presented here is my own, except where due reference has been made in the text. This work has not been submitted for an award of a higher degree elsewhere.

Name: Jess Smith

Signature:

Date:

## Acknowledgments

Firstly, and most importantly, I would like to thank the participants who generously gave their time to be involved in my research. It was a privilege to hear their stories, and I hope that my writing will do them justice. Thanks are also due to my research supervisors, Dr Fiona Eccles and Dr Craig Murray, for their patience and support through the many trials and tribulations of this thesis. Finally, thank you to everyone who sent photos of dogs and babies to sustain me through the completion of this project.

## Contents

**Section One: Literature Review**

# Parents' Experiences of Caring for Children with Persistent Pain: A Systematic Review and Thematic Synthesis of Qualitative Research

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**Abstract**

This systematic review and thematic synthesis examined experiences of parents of children with persistent pain. A systematic search was completed in March 2020. Analysis of 17 papers produced six themes: seeking control in an uncontrollable situation; being let down by experts and becoming their own expert; fearing judgment whilst judging themselves; seeking normality even whilst adapting to a 'new normal'; focusing on the child vs. awareness of impact on the wider family; and dichotomy: the push and pull of raising a child with persistent pain. This experience is associated with a series of competing demands. Clinical implications are considered.

*Keywords*: children and young people, chronic pain, thematic synthesis, parents, systematic review

## Introduction

Persistent pain, or chronic pain, is a common experience for children and adolescents, with an estimated overall prevalence between 11-38% (King et al., 2011). Persistent pain can be defined as pain continuing beyond the normal healing phase (Merskey and Bogduk, 2012). As Merskey and Bogduk discuss, however, this period of healing can be difficult to accurately define, and is variable both within and between conditions; this definition also excludes conditions and syndromes where "normal healing" does not occur, yet pain is a main symptom – such as rheumatoid arthritis (Steingrímsdóttir et al., 2017). For clinical purposes, therefore, pain lasting longer than three months tends to be considered "persistent" (Merskey and Bogduk, 2012).

Pain can be understood as having a protective function – serving as a warning signal to avoid potential injury risk, or encouraging rest to allow an injury to heal (Butler and Moseley, 2003). In persistent pain, however, this is less relevant; persistent pain is not necessarily indicative of tissue damage, and by definition is not constrained to a healing period (Chambliss et al., 2002). In this way, rather than serving to protect, persistent pain can cause further detriment; for example, the fear-avoidance model posits fear of movement can cause individuals with persistent pain to avoid movement and exercise (Asmundson et al., 2012). In the short-term (with acute injury) this can be beneficial, but in the long-term it leads to deconditioning, can exacerbate pain, and is associated with increased disability (Booth et al., 2017).

In their systematic review of 41 papers relating to epidemiology of persistent pain, King et al. (2011) found that girls are more likely to experience persistent pain than boys, and prevalence increased with age. Prevalence was found to be affected by psychosocial variables including anxiety and low self-esteem. A recent systematic review of 14 papers by Alsaggaf and Coyne (2020) reported conflicting evidence, but concluded that persistent pain appears to have an overall negative impact on adolescents' school functioning.

Beyond its impact on individuals, paediatric persistent pain has wider social implications; in the UK, cost per adolescent has been estimated at around £8000 per year (Sleed et al., 2005). This may be an over-estimate as it was based on adolescents recruited from tertiary care services; many young people who experience persistent pain do not access these types of services and are therefore likely to incur lower costs (Groenewald and Palermo, 2015). Nonetheless, the financial burden of paediatric persistent pain is likely to be significant, including direct care costs such as appointments and medications, as well as indirect costs such as loss of productivity for parents who take time off work to care for their child (Sleed et al., 2005).

Paediatric persistent pain affects those who are invested in young people's wellbeing (Vetter, 2011). It can affect the family in various ways, from the distress of seeing the child in pain to the financial burden of medical care and other costs associated with a long-term health condition (Schechter, 2014). Lewandowski et al. (2010) reviewed 16 quantitative papers relating to family functioning in families of children with persistent pain, concluding that these families generally had poorer functioning (including factors such as conflict, cohesion, and communication) than healthy controls.

To date, no review (qualitative or quantitative) has been completed considering the impact of paediatric pain on parents separately from other family members; however, several papers have found evidence of negative effects on parents of children with persistent pain. For example, Cohen et al. (2010) found that lower functioning in children with persistent pain was associated with higher parental stress, anxiety, and depression; that is, as the child's functional disability increased, so did parents' distress. Looking specifically at mothers of children with functional abdominal pain, Campo et al. (2007) found that they were more likely than mothers of healthy children to suffer from health conditions including irritable bowel syndrome, migraine, anxiety, and depression. Hunfeld et al. (2001) also surveyed mothers of children with persistent pain, reporting that pain caused stress for parents and

impacted negatively on their social lives. From the findings of such papers, it is apparent that paediatric persistent pain has a significant impact for parents.

The relationship between paediatric persistent pain and parents' responses to that pain is bi-directional; whilst the child's pain has implications for the parents, parental responses have also been shown to impact the child's experience of pain. For example, in their meta-analysis of 36 papers, Donnelly et al. (2020) identified that parents' cognitions, behaviours, and affective responses to their child's persistent pain were associated with the child's level of disability, pain intensity, depression, anxiety, and functioning at school. Donnelly et al. suggest that parental responses may be an appropriate target for intervention in paediatric persistent pain, noting that additional research is required to understand these parental responses more fully.

Chow et al. (2016) carried out a longitudinal study of 195 persistent pain patients aged 8-17 years; they concluded that parental distress and behaviours impact upon the child's distress and levels of functioning over time. Though their follow-up period was only four months, Chow et al.'s work shows the importance of parental responses to persistent pain. In particular, parental avoidance and protective behaviour at baseline was a significant predictor of child functioning at 4-month follow-up. Like Donnelly et al., Chow et al. recommended that interventions for persistent pain in children should include addressing parent factors.

Palermo and Eccleston (2009) describe parents of children with persistent pain as a "neglected but critical aspect of paediatric chronic pain management" (p. 15). There is quantitative evidence of the significant widespread impact of paediatric persistent pain on parents, which in turn affects the child's levels of pain, distress and disability. However, it is not necessarily clear which aspects of parents' experiences contribute to this distress. In order to develop appropriate and effective support for these parents, it is important to understand their experiences and views (Chow et al., 2016; Donnelly et al., 2020). Qualitative research is ideally-placed to offer this perspective, as it focuses on individual experiences; to date, no

review of qualitative research with parents of children with persistent pain has been completed. The present review, therefore, aimed to synthesise research relating to parents' experiences of their child's persistent pain, with a view to better understanding what it is like to have caring responsibilities for a young person with persistent pain. The review question was: What is it like to parent a child with persistent pain?

It is hoped that findings will inform development of appropriate support for parents of children with persistent pain. This in turn may improve outcomes for children, given the findings discussed above whereby parents' responses can affect children's experiences of their symptoms and their levels of disability (Palermo et al., 2014). A review may also be beneficial in identifying directions for future qualitative research in this area.

In terms of the scope of this review, it was decided to include all pain diagnoses including specific conditions where pain is a main symptom, such as juvenile arthritis and complex regional pain syndrome (CRPS), as well as more generic persistent pain. There were multiple reasons for this decision; firstly, as Swain et al. (2014) identified from an international survey of pain in adolescents, there is significant overlap between pain conditions, insofar as children with one type of pain are likely to have multiple types. Any attempted separation would thereby be rendered somewhat arbitrary. Furthermore, according to Walker and Greene (1989), there is no apparent difference in psychological distress between paediatric patients with and without an established organic cause for their recurrent abdominal pain, with both groups showing similarly high levels of depression and anxiety compared to healthy controls. Thus, it would be difficult to rationalise discriminating on the basis of pain condition diagnosis. Finally, given the contention surrounding the definition of persistent pain, it would be difficult to operationalise any discrimination between conditions in a meaningful way.

Thematic synthesis has been described as the bringing together and integration of findings from multiple qualitative research papers on a common topic (Thomas and Harden,

2008). The authors describe this approach as going beyond description and critique of the papers included, and seeking to interpret findings from the included papers, synthesising new theory or conceptual understanding of the topic. Thomas and Harden describe this process as allowing the reviewer to stay close to the original data whilst synthesising in a transparent manner, allowing development of new concepts and hypotheses. Thematic synthesis was therefore selected as an appropriate methodology for this review, given the aim of developing a deeper understanding of parents' experiences of caring for a child with persistent pain.

## Method

The steps outlined by Thomas and Harden (2008) for completing a thematic synthesis were followed: (1) Searching; (2) Quality assessment; (3) Extracting data from the studies (including Results or Findings sections of each paper); (4) Coding text; (5) Developing descriptive themes; and finally (6) Generating analytic themes.

### Search Strategy

The 'SPiDER' tool (Cooke et al., 2012) is designed to identify components of a qualitative research question, facilitating development of a comprehensive search strategy. The acronym stands for Sample; Phenomenon of interest; Design; Evaluation; Research type. The results of this tool for the present review are presented in Table 1.

<Table 1 about here>

Results from the SPiDER tool were used, alongside guidance from a specialist subject librarian, to identify appropriate search terms. Search terms were chosen relating to three key areas: parenting; children; and persistent pain. These search terms are presented in Table 2.

<Table 2 about here>

Following discussion and agreement with a specialist subject librarian, six databases were selected for the search: CINAHL, PsycINFO, MEDLINE, PubMed, Scopus, and Web of Science. Results from PubMed were limited to those published in the past year, to identify

papers not yet indexed on MEDLINE, as recommended by the librarian. The search terms were combined using Boolean operators, with subject headings included as appropriate for each database.

The literature search, carried out in March 2020, returned a total of 20,252 references, which were exported to Endnote (Clarivate Analytics, n.d.). 12,301 duplicates were removed, leaving 7,951 references for screening. An initial screen of titles and abstracts of remaining papers was carried out, according to inclusion and exclusion criteria:

Inclusion criteria:

1. Must be qualitative research which includes "bottom-up" analysis, that is, themes must be generated from data, rather than data being ascribed to pre-determined categories.
2. Must relate to parenting a child with persistent pain (may include non-parents who take a parenting role, e.g. where a grandparent takes on parental responsibility).
3. Must be available in English.

Exclusion criteria:

1. Parental views cannot be separated from others', e.g. healthcare professionals, siblings.
2. Research is an evaluation of a specific resource or intervention, rather than relating to parents' experiences generally.
3. Persistent pain is secondary to a potentially terminal diagnosis (e.g. cancer,), or is comorbid with severe cognitive impairment, which may further complicate parents' experiences of their child's pain.

This initial screen excluded 7, 951 papers, leaving 90 papers for full-text screening. The final round of screening identified a total of 17 papers suitable for inclusion (see Table 3 for papers excluded at this stage, along with reasons for exclusion).

<Table 3 about here>

Reference lists and citations of papers identified were screened for any additional potentially relevant papers. This process of 'snowballing' (checking reference lists) and 'reverse snowballing' (tracking citations) (Sayers, 2007) did not identify any further relevant papers; a total of 17 papers were retained for inclusion in the review. This process of screening is summarised in a PRISMA diagram (Moher et al., 2009) (Figure 1).

<Figure 1 about here>

**Details of Selected Studies**

A total of 17 papers were identified for inclusion in the thematic synthesis, including parents and caregivers from 233 families. Publication dates for the included papers ranged from 2002 to 2019; participants were from countries including the UK (n = 8), the USA (n = 3), Canada (n = 2), Norway (n = 1), Sweden (n= 1), and Brazil (n = 1). One paper (Navarro et al., 2018) reported research conducted anonymously online and was therefore unable to state where participants lived. Of these 17 papers, 12 employed an interview-based design; three used mixed-methods designs including an interview component; one paper used focus groups; and the final paper collected data from messages posted on online forums. In terms of analysis, six papers used thematic analysis, four used grounded theory, two used "qualitative content analysis", two used "standard ethnographic procedures", two used interpretative phenomenological analysis, and one used "inductive content analysis". Details of included papers are summarised in Table 4.

<Include Table 4 about here>

Participants were mainly parents of affected children, with three papers focusing exclusively on experiences of fathers (Jordan et al., 2016; McNeill, 2004; Waite-Jones and Madill, 2008). In addition, in one case (Jordan et al., 2007) a grandparent who identified as main caregiver was included. Some papers focused upon specific conditions where pain is a major symptom, such as juvenile idiopathic arthritis (JIA; Britton & Moore 2002a, 2002b; McNeill, 2004; Rossato, Angelo, & Silva, 2007), Complex Regional Pain Syndrome (CRPS;

Navarro et al., 2018), or neuropathic pain (Gaughan et al., 2014); others focused on persistent pain more generally (Brodwall et al., 2018; Carter, 2002; Jordan et al., 2016; Le et al., 2019; MacIver et al., 2010). Two papers (Britton & Moore 2002a, 2002b) reported upon the same group of participants; these papers reported on differing aspects of the parents' experiences, so both were included.

**Critical Appraisal of Papers**

Assessment of quality of papers for inclusion is the second step in thematic synthesis (Thomas and Harden, 2008). Indeed, there is general consensus that any systematic review of qualitative research ought to include an element of critical appraisal, in order to distinguish well-conducted and -reported papers from those of lower quality (Garside, 2014). Each paper was therefore appraised to assess quality, prior to commencing analysis. As recommended by Sandelowski and Barroso (2007), papers were not excluded on the basis of appraisal; however, this process allows some confidence that lower-quality papers were not given disproportionate weighting in the analysis, and allows conclusions to be drawn about the validity of findings.

The Critical Appraisal Skills Programme (CASP) published a checklist for appraisal of qualitative research papers (CASP, 2018); this checklist consists of ten questions to assist researchers in ascertaining quality and validity of qualitative research papers. The questions are divided into three sections, namely, 'Are the results of the study valid?', 'What are the results?', and, 'Will the results help locally?'. Each section consists of one or more questions to guide researchers' appraisals of papers. Questions 1-9 are to be answered, 'Yes', 'No', or 'Can't tell', whilst question 10 is open-ended ('How valuable is the research?'). Other appraisal tools are available, however the CASP is the most widely used (Hannes and Macaitis, 2012).

CASP do not recommend using a scoring system with the checklist, stating that it is intended purely as an educational tool; however, there is precedent for the CASP checklist

being used to generate a score reflecting a paper's overall quality (Duggleby et al., 2012; Feder et al., 2006). Given this precedent, and the fact that papers would not be excluded based upon the quality appraisal, it was deemed appropriate to use the CASP to generate a score for each paper, allowing a means of operationalising the critical appraisal. Each question was given a score out of three, with 'No' answers corresponding to one, 'Can't tell' two, and 'Yes' three; thus, a score between 10-30 was generated for each paper, with higher scores reflecting higher quality research. Question 10 was adapted slightly to fit with this scoring system; the new question 'Is consideration given to the value of the research?' was answered and scored in the same way as questions 1-9.

Scores ranged from 23-29 (mean = 25.9); all papers were therefore deemed to be of reasonable quality. Full results of the quality appraisal are shown in Table 5.

<Table 5 about here>

**Synthesis**

Papers identified for inclusion were imported to NVivo (QSR International, n.d.). Each paper was read to develop a level of familiarity with the data before stage four of Thomas and Harden's (2008) procedure – coding – was commenced. The process of coding involved allocating short sections of each paper a 'code' which was descriptive of its contents – for example, the statement, "…geographical remoteness of the clinic caused them continuing practical and communication problems" (Britton & Moore, 2002a, p. 377) was coded as 'practical difficulties in accessing treatment/support'. Some sections were assigned multiple codes, where statements appeared to relate to more than one relevant topic – for example, the statement, "It was apparent that professionals had a major impact on the way in which the families viewed themselves and how they felt others viewed them" (Carter, 2002, p. 32) was coded as both 'communication between family and services' and 'being judged/feeling judged/fear of judgement', as it was deemed to relate to both topics. Coding was an iterative process, with subsequent papers sometimes highlighting further codes within

previous papers. Papers were therefore reviewed more than once, to ensure that all potential codes had been drawn out. Coding was limited to Results and Discussion sections of each paper; this ensured that only data from participants, and authors' interpretations of this data, were included in analysis.

The next phase of analysis included reviewing the codes; these were combined where they were seen to be broadly similar; for example, the codes 'silver linings/benefits for family' and 'positive impact for siblings – compassion/consideration' related to similar topics of the perceived positives of having a child with persistent pain in the family, and were therefore combined into one code, 'silver linings'. Codes were then grouped into five descriptive themes: (1) Pain takes over; (2) Parents as a unit; (3) The impact of other people; (4) Stress and difficult emotional responses; and (5) Moving forwards in positive ways. Again, this process was iterative, with themes being reviewed, adjusted, separated, and combined to best represent the data in the papers reviewed.

The final stage of analysis, according to Thomas and Harden, is development of analytical themes; in this stage, the review is aiming to "go beyond the content of the original studies" (Thomas & Harden, 2008, p. 7). Thomas and Harden acknowledge that this stage is dependent upon the "judgment and insight" of reviewers (p. 7), as third-order interpretations – that is, interpretations of interpretations (of authors of included papers) – are generated. This was an iterative process whereby descriptive themes and initial codes were compared, contrasted, and re-structured to develop analytic themes. The descriptive themes did not necessarily map directly onto the final analytic themes, but rather contributed to the researcher's immersion in the data and to the iterative process of analysis. The development of the final themes can be seen in in Appendix A.

Following this process of analysis and synthesis, six final themes were identified: (1) Seeking control in an uncontrollable situation; (2) Being let down by experts and becoming their own expert; (3) Fearing judgment whilst judging themselves; (4) Seeking normality

even whilst adapting to a new normal; (5) Focus on the child whilst recognising the impact on the wider family; and (6) Dichotomy: The push and pull of raising a child with persistent pain. Each of these themes is considered in detail below.

## Results

### Theme 1: Seeking control in an uncontrollable situation

Parents experienced their situation as being beyond their control, and made efforts to retain and regain (that is, keep hold of control where possible, and take back where it was perceived to have been lost) control. A common topic of discussion by parents within the papers reviewed was the uncertainty, unpredictability, and lack of control they felt in relation to their child's persistent pain, as described by a participant in Jordan et al. (2016): "The most difficult thing about it is not knowing how he's going to be when I get home" (p. 2469). The unpredictable nature of their children's conditions made it difficult for parents to make plans, in both the short- and long-term (Jordan et al., 2016; Waite-Jones and Madill, 2008). Parents were fearful of this lack of control, and that things would be this way forever (Gaughan et al., 2014; Sallfors and Hallberg, 2003).

This uncertainty and lack of control led to parents feeling they had to be constantly alert in case of a pain flare-up or a change in their child's condition (Britton and Moore, 2002b; Jordan et al., 2007; Le et al., 2019; MacIver et al., 2010; McNeill, 2004; Rossato et al., 2007; Sallfors and Hallberg, 2003; Smart and Cottrell, 2005; Yuwen et al., 2017). Parents described having to stay strong in the face of the uncertainty of their child's pain, in an effort to protect both the child and the rest of the family (MacIver et al., 2010; McNeill, 2004; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008). This was described by one mother in MacIver et al.'s study: "You really have to toughen up to the pain side of things, because otherwise you just- I would have just gone, by now" (p. 1277).

Parents described many and varied ways in which they attempted to take back control of the situation. These included advocating for appropriate support for their child

(Brodwall et al., 2018; Gaughan et al., 2014; Jordan et al., 2007; McNeill, 2004; Navarro et al., 2018), and developing their medical knowledge, relating to both the conditions themselves and to possible treatments (Waite-Jones and Madill, 2008; Yuwen et al., 2017). Some parents had to learn new skills, such as how to deliver injections to their child, contributing to a sense of mastery or control (Yuwen et al., 2017). Some parents viewed these skills as a source of pride; one father described how nurses had complimented his bandaging skills (Waite-Jones and Madill, 2008).

Other parents fought to retain control of the situation, by denying there was a problem at all, as one mother explained about her partner: "He doesn't really talk about it… He didn't believe it was happening and tried to ignore it. Tried to pretend it wasn't happening" (Britton & Moore, 2002b, p. 416). Another approach described by parents involved taking a pragmatic stance and focusing upon what could be controlled, which often meant living in the moment and taking each problem as it came (McNeill, 2004; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008).

**Theme 2: Being let down by experts and becoming their own expert**

Parents described feeling let down by medical professionals, feeling they were not taken seriously (Carter, 2002). Parents frequently felt invalidated and disbelieved – "All the families perceived many professionals to be suspicious about their child's pain. Eventually, they were referred to a psychologist, which was seen by the families as evidence of their failure to get professionals to believe the reality of their child's pain." (Carter, 2002, p.34). Parents also felt that professionals did not understand what it was like for them as parents trying to manage their child's pain. Parents described being frustrated by not receiving the support, reassurance, and information they needed from healthcare teams, and feeling unrecognised as a central component of the team around the child (Britton and Moore, 2002a; Britton and Moore, 2002b; Brodwall et al., 2018; Carter, 2002; Gaughan et al., 2014; Jordan et al., 2016; Jordan et al., 2007; Le et al., 2019; MacIver et al., 2010; Navarro et al., 2018;

Sallfors and Hallberg, 2003; Smart and Cottrell, 2005; Waite-Jones and Madill, 2008; Yuwen et al., 2017). Parents experienced professionals as being unaware of best practice standards (Le et al., 2019; MacIver et al., 2010; Navarro et al., 2018; Sallfors and Hallberg, 2003), which contributed to a lack of trust in healthcare teams at times (Britton and Moore, 2002a; Britton and Moore, 2002b; Brodwall et al., 2018; Carter, 2002; Gaughan et al., 2014; Sallfors and Hallberg, 2003; Smart and Cottrell, 2005; Yuwen et al., 2017).

In light of this, parents came to view themselves as experts on their own child (Navarro et al., 2018; Sallfors and Hallberg, 2003; Smart and Cottrell, 2005). In many cases, they felt that they had better understanding of their child's pain, and how to manage it, than professionals involved in their care – for example in Navarro et al. (2018) again: "Sarah portrayed herself as being superior to the 'floundering' consultants. Use of the word 'floundering' suggests Sarah's perception of the consultants as indecisive and to be experiencing difficulty, enabling her to position herself as the expert." (p. 4).

**Theme 3: Fearing judgment whilst judging themselves**

Parents feared judgment from health professionals, which included being disbelieved about their child's pain – for example in Brodwall et al. (2018): "Some parents were afraid to be viewed as 'hysterical mothers'. They… feared not being taken seriously by doctors" (p. 4). Communication received from health professionals also contributed to parents' distress (Britton and Moore, 2002b; Brodwall et al., 2018; Carter, 2002), which makes sense when they feel dismissed, blamed, or disbelieved. Brodwall et al. (2018) considered how, without adequate and sensitive explanation, the biopsychosocial model of pain could lead to parents feeling blamed for their child's pain, and ashamed that they could not make it stop – exacerbating this fear of judgment. These feelings are perhaps intensified further when parents feel that professionals are suspicious of them, and that their coping strategies are dismissed by the healthcare team (Carter, 2002).

Parents also feared judgment from peers and social contacts (Gaughan et al., 2014; Jordan et al., 2007; Smart and Cottrell, 2005; Yuwen et al., 2017). Gaughan et al. described parents' friends being supportive and helpful to begin with, but over time becoming critical of them and their coping strategies. Parents mentioned others not understanding their child's condition, which also contributed to their fears (Britton and Moore, 2002a; Britton and Moore, 2002b; Gaughan et al., 2014; Jordan et al., 2016; Rossato et al., 2007; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008). As well as being judged themselves, parents were concerned about their child being judged, as described by Yuwen et al. (2017): "Parents felt that their children were judged by other people… in situations such as the older child sitting in a stroller" (p. e27). Feeling judged and misunderstood like this may contribute to parents' reluctance to seek external support, both emotional and practical (Jordan et al., 2016).

Alongside this fear of judgment from others, parents seemed to judge themselves. Parents described their children's difficulties leading to a sense of failure in their role as parent (MacIver et al., 2010; McNeill, 2004; Rossato et al., 2007; Smart and Cottrell, 2005; Waite-Jones and Madill, 2008; Yuwen et al., 2017) and tended to blame themselves. Parents placed significant pressure upon themselves to know their child well enough to assess their pain and the best course of action; at the same time, their inability to resolve the pain led to feelings of inadequacy (Smart and Cottrell, 2005). This was exacerbated by parents' role in the management of their child's condition, which sometimes entailed inflicting more pain through physiotherapy exercises, and so on. Worse yet, these treatments were not always perceived to help the child, and sometimes caused side-effects (Britton and Moore, 2002a; Gaughan et al., 2014; MacIver et al., 2010; Navarro et al., 2018; Rabbitts et al., 2017; Yuwen et al., 2017). This was summarised by one mother in MacIver et al.: "I feel like nothing you do is really helping. And everything you do hurts" (p. 1276). This, in turn, contributed to parents' judgment of themselves as inadequate or lacking, (Gaughan et al., 2014). Parents

spoke of betraying their child and felt guilt, horror, and trauma from the treatments they had to provide (Yuwen et al.).

**Theme 4: Seeking normality even whilst adapting to a 'new normal'**

A significant proportion of parents' distress appeared to arise from a sense of their child being less than normal, and somehow inferior to 'normal' children; parents described jealousy when comparing their child or family with others perceived as normal, and the impact this had for them socially (Britton and Moore, 2002b; Jordan et al., 2007; Le et al., 2019; Waite-Jones and Madill, 2008; Yuwen et al., 2017). There was a sense of grief here: "Parents reported a mourning of the loss of a happy, healthy, carefree child; this sense of mourning exacerbated by parental comparison of their ill adolescent with healthy siblings or peers" (Jordan et al., 2007, p. 54). Parents tried to maintain normal activities (Rossato et al., 2007; Sallfors and Hallberg, 2003) and described wanting their child to lead a normal life (Rossato et al., 2007). In some ways, these efforts to maintain normal activities in the face of their child's condition were another example of parents' efforts to regain some control, as discussed in Theme 1.

Even as they struggled with these matters, parents undertook a simultaneous process of adjusting to a new normal, as described in Waite-Jones and Madill (2008): "Eventually fathers adjusted such that their family life became 'normal'… Len explained that 'It has been there that long now it is part of your routine'" (p. 596). As part of this process, parental roles changed and they began to parent their child in new ways (Britton and Moore, 2002b; Carter, 2002; Gaughan et al., 2014; Jordan et al., 2016; MacIver et al., 2010). MacIver et al. described parents having to change not only their behaviour but also their perceptions of what makes a good parent; this process perhaps resolved or eased some of the distress relating to judging themselves as inadequate, discussed in Theme 3. In addition, parents spoke about changes in priorities in light of their child's condition, with heavier focus on the health, wellbeing, and happiness of the child and the family (Jordan et al., 2016).

This adaptation was seen as a gradual process rather than an immediate change (Jordan et al., 2016; MacIver et al., 2010; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008), as parents tussled with the implications of living with a child with persistent pain, and developed ways of coping in their new, often changeable, situations.

**Theme 5: Focusing on the child vs. awareness of the impact on the wider family**

This theme considers the need for additional attention for the child in pain, weighed against the needs of the family as a whole, and the ways parents attempted to find a functional balance. The child's pain necessitated them being the centre of attention to a point, and yet parents were conscious of the impact this had on the family as a whole. For example, as Brodwall et al. (2018) discussed, "Many parents deviated from their daily routines during pain episodes, for instance, by making special food or gathering in front of the television." (p. 4). Parents were aware of a change in the nature of their relationship with the child with pain; the activities which they could enjoy with their child were changed (Jordan et al., 2016), with some fathers in Jordan et al.'s study reporting strengthening of the relationship as a result, whilst others reported it was weakened. The impact on the parents' marriage was also acknowledged, with parents reporting disagreements regarding treatment options, uneven division of the burden of care for the child, and an impact on their sex lives (MacIver et al., 2010; Rossato et al., 2007; Waite-Jones and Madill, 2008; Yuwen et al., 2017).

Even as they made exceptions and changes to the family's way of functioning for the child with pain, there was an awareness of the impact this had for other members of the family in terms of fatigue and limitations to the parents' social lives (Britton and Moore, 2002a; Britton and Moore, 2002b; Brodwall et al., 2018; Gaughan et al., 2014; Jordan et al., 2016; Jordan et al., 2007; Le et al., 2019; MacIver et al., 2010; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008; Yuwen et al., 2017). For example, parents described having to divide the family at weekends or on holiday, with the child with pain spending time with one parent doing more 'sedate' activities, whilst other children went with the second parent for

more ‘challenging’ activities (Britton & Moore, 2002b). Parents were conscious of the impact on the child’s siblings, for example in terms of reduced availability of time and attention (Britton and Moore, 2002a; Britton and Moore, 2002b; Brodwall et al., 2018; Gaughan et al., 2014; Jordan et al., 2016; Yuwen et al., 2017). The knowledge of this discrepancy between siblings was another contributing factor to parents’ distress, and they endeavoured to make up for it in other ways – for example, in Jordan et al. (2007): “I don’t want my boys to feel that when they look back, that their life was so different because of their sister. I try not to let them have to do things ... I think that I probably overdo that” (p. 53).

Parents discussed the impact of having a child with persistent pain on their careers; the unpredictable nature of the children’s conditions meant parents often worked reduced hours or required flexible working, which had an impact upon the family’s financial situation (Le et al., 2019). Some parents found it difficult to balance their simultaneous roles as parent/carer and employee/earner (Jordan et al., 2016), with parents being aware of the impact of their child’s changeable condition upon their proficiency at work (McNeill, 2004). Some parents (mainly mothers) reported having had to give up working altogether (MacIver et al., 2010; Rossato et al., 2007; Sallfors and Hallberg, 2003). Yuwen et al. (2017) also considered the difficult balance between parents needing to take time off work to care for their child and attend appointments, whilst also needing to work additional hours to provide financially for the family, and in particular for added costs associated with the child’s condition.

**Theme 6: Dichotomy: The push and pull of raising a child with persistent pain**

Running through the first five themes is a narrative of being pulled in two directions at once. However, there are additional examples of this sense of ‘push and pull’, beyond the themes already discussed. This final theme, then, reflects the conflict and inconsistency which was evident across the papers reviewed, in numerous aspects of parents’ experiences of caring for a child with persistent pain.

Parents spoke about the importance of interaction with and support from others in similar situations (Gaughan et al., 2014; Le et al., 2019; Rabbitts et al., 2017; Sallfors and Hallberg, 2003; Yuwen et al., 2017) and yet, found it difficult to hear about the struggles of others in similar situations: “Fathers who had shared experiences with others in similar situations found that learning about the experiences of others made their own experience of chronic pain more negative” (Jordan et al., 2016, p. 2470).

With a similar sense of ‘push and pull’, parents spoke about being protective of the child and/or the family unit around the child (Britton and Moore, 2002b; Gaughan et al., 2014; Jordan et al., 2016; MacIver et al., 2010; McNeill, 2004; Rossato et al., 2007; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008; Yuwen et al., 2017), and yet, described trying to balance this with not being over-protective or over-sympathetic, for fear of leading to hypochondria (Smart and Cottrell, 2005; Waite-Jones and Madill, 2008). Here, parents were understandably concerned about their child and wished to take good care of them, but at the same time did not wish to ‘spoil’ the child or risk making matters worse by coddling them.

With a similar sense of conflicting interests, parents spoke about fighting for access to support and resources (Jordan et al., 2007; Navarro et al., 2018; Sallfors and Hallberg, 2003), whilst at the same time, the number of appointments their children attended was perceived in some ways as a burden requiring careful management (Le et al., 2019; Sallfors and Hallberg, 2003; Waite-Jones and Madill, 2008; Yuwen et al., 2017). Parents simultaneously recognised their need for support and resources for their child and described fighting to access this support, whilst appearing resentful of the imposition on their family.

## Discussion

This thematic synthesis of 17 papers relating to parenting a child with persistent pain produced a synthesis of six main themes: (1) Seeking control in an uncontrollable situation; (2) Being let down by experts and becoming their own expert; (3) Fearing judgment whilst

judging themselves; (4) Seeking normality even whilst adapting to a 'new normal'; (5) Focusing on the child vs. awareness of the impact on the wider family; and (6) Dichotomy: The push and pull of raising a child with persistent pain. Together, these themes offer understanding of parents' experiences of caring for a child with persistent pain. In particular, the review highlights how this experience is characterised by a series of dichotomies; parents are frequently pulled in two directions at once or pursue two competing objectives. This understanding may offer some explanation as to the distress experienced by parents of children with persistent pain.

Research on the experience of parenting a child with a long-term health condition already acknowledges difficulties which are inherent within this role; for example, a systematic review by Cousino and Hazen (2013) looked at parenting stress across a number of long-term conditions. The authors concluded that both general parenting stressors and illness-specific stressors were of relevance to these parents, with greater responsibility for management of the condition being associated with higher levels of stress. However, to date, no review has considered the experiences of parents of children with persistent pain, which is a somewhat unique experience.

Persistent pain differs from many long-term conditions insofar as it is not generally life-threatening (indeed, the present review excluded papers which reported on parents of children with pain secondary to a potentially terminal condition, such as cancer), and yet can have a profound impact on daily functioning for child and family. A review by Santacroce (2003) concluded that uncertainty relating to a child's health condition is linked with post-traumatic stress symptoms. Santacroce posited that parents attempt to manage this uncertainty in two main ways, namely through seeking information about the child's condition and also through avoiding social encounters or information which might highlight negative aspects of uncertainty. Santacroce compared these strategies to hyperarousal and avoidance symptoms, respectively, found in Post-Traumatic Stress Disorder (PTSD). These strategies appear to fit

with the dichotomy discussed in Theme 6 above, whereby parents desire support from others who have faced similar situations, whilst simultaneously finding information from their peers distressing.

Theme 3, which related to parents feeling that they were judged by others, whilst also judging themselves, fits within the wider literature on health-related stigma. Goffman (1963) defines stigma as the judgment that an individual has an attribute or attributes which makes them different, and therefore somehow less, than others; Link and Phelan (2001) suggest that stigma also involves an element of discrimination. Stigma is sometimes separated into different categories, whereby *enacted* refers to behaviours driven by negative beliefs towards the individual, whilst *felt* refers to the individual's sense of being devalued; *internalised* stigma, meanwhile, is stigma the individual directs towards themselves due to taking on societal beliefs and stereotypes (Major et al., 2018).

Wakefield et al. (2018) discuss the role of stigma in adolescent persistent pain, concluding from their focus group that sources of stigma included healthcare providers, school staff, family members, and peers. Though their work was preliminary, Wakefield et al. drew on similar work with adults with persistent pain which has demonstrated links between stigma and higher levels of anxiety and stress, disruptions in relationships, and social isolation. Being the parents of a child with a health condition is also associated with stigma; for example, Gray (1993) found that parents of autistic children perceived themselves to be stigmatised by their child's condition. The findings of this review suggest that parents of children with persistent pain also perceive stigma towards themselves.

**Clinical Implications**

There is some evidence of the utility of psychological interventions for parents of children with long-term illness, as a recent Cochrane review concluded (Law et al., 2019). For clinical psychologists (and others) supporting parents of children with persistent pain, a more thorough understanding of experiences faced by these parents may be beneficial in

offering appropriate intervention. For example, the themes of dichotomy and seeking control in an uncontrollable situation are likely to fit well with an acceptance and commitment therapy (ACT) approach (Hayes et al., 1999). ACT-based work promotes psychological flexibility and values-focused action. ACT is recognised as a valuable approach for individuals living with persistent pain, encouraging them to pursue what is important in their lives, in spite of the pain. In their systematic review, Hann and McCracken (2014) found ACT to be efficacious in increasing general functioning and decreasing distress in adults with persistent pain. It is logical, then, that ACT may be useful for people affected by a loved one's persistent pain – especially given issues highlighted in the present review, whereby uncertainty and 'abnormality', as well as responses such as seeking control and fear of judgment, make significant contributions to distress.

Indeed, some research relating to the utility of ACT with parents of children with persistent pain has already been published; Kanstrup et al. (2016) reported on their pilot study of an ACT-based intervention for adolescents with persistent pain and their parents. Improvements for parents were noted in pre- and post-intervention measures of pain reactivity and psychological flexibility, though there was no change in anxiety, depression, or overall emotional functioning. Given evidence of the impact of parental responses on children's level of disability, pain intensity, emotional wellbeing, and school functioning (Donnelly et al., 2020), such interventions are of potential importance to both parents and their children.

In addition to ACT, a National Clinical Guideline by the Scottish Government highlights family cognitive behavioural therapy (CBT) as a possible intervention for moderate to severe persistent pain (The Scottish Government, 2018). A randomised controlled trial by Palermo et al. (2016) found that internet-delivered CBT for adolescents with persistent pain and their parents contributed to improvements in outcomes. Palermo et

al. concluded that their CBT intervention offered a number of benefits for both parents and adolescents.

Another pertinent issue for consideration by professionals working with this group, as identified in the first theme, is feeling let down by experts, whom parents frequently experienced as dismissing their children. Of particular relevance is parents' perception that a referral to psychology meant that doctors did not believe them about their child's pain; this highlights the care that must be taken in explaining the rationale behind such referrals and the kind of support that psychology may provide. There may be a need for psychology to ensure that colleagues and referring teams are clear about the role of psychology and what constitutes an appropriate referral, to allow better communication to parents about such referrals. Several papers discussed parents' difficulties with communication from their healthcare teams, for example in Britton and Moore (2002b) where the authors concluded that healthcare professionals can contribute to parents' distress rather than supporting them to cope.

**Limitations**

The findings of this meta-synthesis are of course limited by the papers reviewed. Notably, the vast majority of papers were from North America and Western Europe, with only one paper coming from Brazil and one being conducted online meaning location of participants was unknown. The experiences of parents in these studies, therefore, are shaped by Western society's opinions on the role of parents; findings may not be generalisable to non-Western settings where experiences may be markedly different. A systematic review by Orhan et al. (2018) concluded that experiences of persistent pain differ between cultures in various aspects, including coping strategies, illness perceptions, and self-efficacy. The papers included in Orhan et al.'s review focused almost exclusively on adults; however, it is reasonable to expect that there may be similar differences in children, particularly given the

evidence that children's experiences of pain are shaped by their parents' attitudes and beliefs (Donnelly et al., 2020).

An additional limitation is the scarcity of research with fathers, which was discussed in several papers; as Britton and Moore (2002b) pointed out, 'research with parents' often translates to 'research with mothers', meaning that perspectives and experiences of fathers are neglected and poorly understood. This has implications for the ability of services to offer appropriate support to parents of children with persistent pain. Three papers in this review focused exclusively on experiences of fathers (Jordan et al., 2016; McNeill, 2004; Waite-Jones & Madill, 2008), going some way to addressing this imbalance, but future work including fathers, and particularly comparing and contrasting the experiences of mothers and fathers, would be of value. MacFadyen et al. (2011) discuss some benefits of involving fathers in research, including a clearer picture of how the child's condition is affecting the family. MacFadyen et al. (2011) also offer suggestions on ways of engaging fathers in research, which may aid future researchers in this area.

As with all approaches to the synthesis of qualitative data, there are methodological limitations to consider. For example, Sandelowski et al. (1997) argued that qualitative research is by its nature resistant to being summarised, given its emphasis on the importance of context; they suggest that in summing up qualitative findings, the integrity of the original research is lost. Sandelowski et al. also highlight the difficulties with conflating the diverse approaches which can constitute qualitative research; indeed, in the present review, a variety of techniques of data collection and analysis were noted. However, the aim of thematic synthesis as described by Thomas and Harden (2008) is not to summarise the papers included but rather to 'go beyond' description and critique, to develop a new theory or understanding of the phenomenon of interest, thus creating a synthesis which is more than the sum of its parts. Therefore, whilst some context and detail is inevitably lost through the analytic

process, this is not necessarily in opposition to the primary aim of developing new conceptual understanding.

**Recommendations for Future Research**

As highlighted above, no comprehensive review of quantitative findings relating to parents' experiences of raising a child with persistent pain has yet been completed. This area may benefit from future research attention, in order to gain a fuller understanding of the impact of paediatric persistent pain on parents. Additionally, as discussed, there is a scarcity of research which includes or focuses upon the experiences of fathers, and what little research there is suggests that experiences of mothers and fathers can differ quite considerably. There is merit, therefore, in developing understanding of these differences and potential implications for support. In general, additional research into the experiences of these parents would be of value. In particular, research which aims to understand how parents seek to balance the host of competing demands they face, as discussed above, may inform the development of more tailored support. Furthermore, an exploration of parents' experiences of being referred to psychology may be of benefit in informing referral pathways. This may be valuable given the distress reported by parents who perceived that such referrals meant they were disbelieved about their child's pain.

**Conclusions**

In conclusion, this thematic synthesis identified six themes which summarised experiences of parenting a child with persistent pain. A narrative running throughout these themes was the idea of 'dichotomy', whereby parents consistently have to manage competing demands and occupy seemingly contradictory positions. Implications for therapeutic work and other interventions are considered, and recommendations of directions for future research are made.

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Yuwen, W., Lewis, F. M., Walker, A. J. & Ward, T. M. (2017) Struggling in the dark to help my child: Parents' experiences in caring for a young child with juvenile idiopathic arthritis. *Journal of Pediatric Nursing,* 37**,** e23-e29. 10.1016/j.pedn.2017.07.007.

## Tables and Figures

**Table 1: Results of SPiDER Tool**

| **Element** | **Details** |
|---|---|
| Sample | Parents (or those taking on the parenting role) |
| Phenomenon of Interest | Paediatric persistent pain |
| Design | Interviews, focus groups, open-ended questionnaires, diary studies |
| Evaluation | Experiences, perceptions, attitudes, views, opinions |
| Research type | Qualitative |

**Table 2: Search Terms**

| **Concept** | **Search Terms** | **MeSH Headings (CINAHL, Medline)** | **APA Subject Headings (PsychInfo, PsycArticles)** |
|---|---|---|---|
| Parents | "parent*" OR "mother*" OR "father*" OR "caregiv*" OR "grandpa*" OR "grandm*" OR "grandfather*" | Parent-Child Relations OR Parental Attitudes OR Parenting OR Parents OR Caregivers OR Mother-Child Relations OR Mothers OR Father-Child Relations OR Fathers OR Grandparents | Parental Attitudes OR Parental Involvement OR Parenting OR Parenting Skills OR Parenting Style OR Parent Child Relations OR Caregivers OR Grandparents |
| Children | Child* OR Pediatr* OR paediatr* OR Adolescen* OR "young pe*" OR youth* OR teen* | Child OR Pediatrics OR Adolescence OR Young Adult | Pediatrics OR Chronically Ill Children |
| Persistent Pain | "chronic N/3 pain" OR "persistent N/3 pain" OR "recurrent N/3 pain" OR "long-term N/3 pain" OR "fibromyalgia" OR "arthritis" OR "headache" OR "migraine" OR "stomach ache" OR "ear ache" OR | Chronic Pain OR Complex Regional Pain Syndromes OR Reflex Sympathetic Dystrophy OR Neuralgia OR Fibromyalgia OR Arthritis, Juvenile OR Headache OR Migraine Disorders OR Earache | Chronic Pain OR Fibromyalgia OR "Complex Regional Pain Syndrome (Type I)" OR Neuralgia OR Back Pain OR Arthritis OR Headache OR Migraine Headache OR |

| | | | |
|---|---|---|---|
| | “complex regional pain syndrome” OR “CRPS” OR “neuropath*” OR “back pain” OR “cerebral palsy” OR “hurts” | OR Back Pain OR Low Back Pain OR Cerebral Palsy | Neuropathic Pain OR Cerebral Palsy |

**Table 3: Papers Excluded at Full-Text Screening Stage, With Reasons**

| Author/s (Year of publication) | Title | Reason for exclusion |
|---|---|---|
| Alaee, Shahboulaghi, Khankeh & Kermanshahi (2015) | Psychosocial challenges for parents of children with cerebral palsy: a qualitative study | Not focused on pain |
| Anonymous (2014) | Down the medical rabbit hole | Not research |
| Appelbaum & Smolowitz (2012) | Appreciating life: being the father of a child with severe cerebral palsy | Not focused on pain |
| Aras, Aras, Sahin & Yanerdag (2011) | The effect of pain complaint on quality of life in mothers of children with cerebral palsy | Not available in English |
| Aras, Aras, Sahin & Yanerdag (2011) | Quality of life in mothers of children with cerebral palsy | Not available in English |
| Backman, Smith, Smith, Montie & Suto (2007) | Sometimes I can, sometimes I can't': The influence of arthritis on mothers' habits | Not paediatric pain |
| Barlow & Ellard (2006) | The psychosocial well-being of children with chronic disease, their parents and siblings: An overview of the research evidence base | Not original research (review - but not focused on pain) |
| Barlow (1998) | Parents' experience of caring for children with juvenile chronic arthritis | Not available |
| Barlow, Cullen-Powell & Cheshire (2006) | Psychological well-being among mothers of children with cerebral palsy | Not qualitative research |

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|---|---|---|
| Barlow, Harrison & Shaw (1998) | The experience of parenting in the context of juvenile chronic arthritis | Data coded into pre-determined categories |
| Bennett, Huntsman & Lilley (2000) | Parent perceptions of the impact of chronic pain in children and adolescents | Not qualitative research |
| Britton & Moore (2002) | Views from the inside, part 3: How and why families undertake prescribed exercise and splinting programmes and a new model of the families' experience of living with juvenile arthritis | Specifically about an intervention |
| Burkhard (2011) | The lived experience of mothers caring for an adolescent or young adult with severe cerebral palsy | Thesis |
| Burkhard (2013) | A different life: Caring for an adolescent or young adult with severe cerebral palsy | Not focused on pain |
| Bursch & Zelter (2001) | Pain: A Story | Not parents' experiences |
| Carter, Arnott, Simons & Bray (2017) | Developing a sense of knowing and acquiring the skills to manage pain in children with profound cognitive impairments: Mothers' perspectives | Cognitive impairment |

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|---|---|---|
| Carter, McArthur & Cunliffe (2002) | Dealing with uncertainty: Parental assessment of pain in their children with profound special needs | Cognitive impairment |
| Chaturvedi & Kanakalatha (1988) | Pain in children of chronic pain patients | Not parents' experiences |
| Cooper (2002) | Parents as care managers: the experiences of those caring for young children with cerebral palsy | Not research |
| Cuneo (1997) | Psychological implications of childhood arthritis on adolescent and parent adjustment | Not qualitative research |
| Davis, Shelly, Waters, Boyd, Cook, Davern & Reddihough (2009) | The impact of caring for a child with cerebral palsy: Quality of life for mothers and fathers | Not focused on pain |
| Dunford, Thompson & Gauntlett-Gilbert (2014) | Parental behaviour in paediatric chronic pain: A qualitative observational study. | Observational only |
| Evans, Meldrum, Tsao, Fraynt & Zeltser (2010) | Associations between parent and child pain and functioning in a pediatric chronic pain sample: A mixed methods approach | Emphasis on children's experiences, not parents |
| Glasscock (1997) | The experience of being a mother of a child with cerebral palsy: A phenomenologial study | Not focused on pain |

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|---|---|---|
| Glasscock (2000) | A phenomenological study of the experience of being a mother of a child with cerebral palsy | Not focused on pain |
| Gomez-Ramirez, Gibbon, Berard, Jurencak, Green, Tucker, Shiff & Guzman (2016) | A recurring rollercoaster ride: A qualitative study of the emotional experiences of parents of children with juvenile idiopathic arthritis | Data coded into pre-determined categories |
| Gorodzinsky, Tran, Medrano, Fleischman, Anderson-Khan, Ladwig & Weisman (2012) | Parents' initial perceptions of multidisciplinary care for pediatric chronic pain | Specifically about experiences of one service |
| Guite, Russell, Homan, Tepe & Williams (2018) | Parenting in the context of children's chronic pain: Balancing care and burden | Not research |
| Hallisy (2015) | Empowerment: A pain caregiver's perspective | Not research |
| Ho, Goldschneider, Kashikar-Zuck, Kotagal, Tessman & Jones (2008) | Healthcare utilization and indirect burden among families of pediatric patients with chronic pain | Not qualitative research |
| Hunfeld, Perquin, Duivenvoorden, Hazebroek-Kampschreur, Passchier, van Suijlekom-Smit & van der Wouden (2001) | Chronic pain and its impact on quality of life in adolescents and their families | Not qualitative research |

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|---|---|---|
| Hunfeld, Perquin, Hazebroek-Kampschreur, Passchier, van Suijlekom-Smit & van der Wouden (2002) | Physically unexplained chronic pain and its impact on children and their families: The mother's perception | Not qualitative research |
| Jensen, Patel, Listernick, Charrow & Lai (2019) | Lifespan development: Symptoms experienced by individuals with Neurofibromatosis Type 1 Associated Plexiform Neurofibromas from childhood into adulthood | Parents' views not separate |
| Jongudomkarn, Aungsupakorn & Camfield (2008) | Families in northeast Thailand: Living with a child in chronic pain | Children with cancer |
| Jordan (2010) | Parenting an adolescent with chronic pain: Impact on parents and association with adolescent functioning | Not original research (review - but not systematic, not focused on parents, & doesn't include many of the papers identified for the current review) |
| Kemper, Sarah, Silver-Highfield, Xiarhos, Barnes & Berde (2000) | On pins and needles? Pediatric pain patients' experience with acupuncture | Not parents' experiences |

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|---|---|---|
| Kurtuncu, Akhan, Yildiz & Demirbag (2015) | Experiences shared through the interviews from fifteen mothers of children with cerebral palsy | Not focused on pain |
| Lafrenaye, Dumas, Duhamel & Bourgault (2010) | La symbolique des parents en regard de la douleur de leur enfant atteint d'une maladie chronique | Not available in English |
| Lauruschkus, Nordmark & Hallstrom (2017) | Parents' experiences of participation in physical activities for children with cerebral palsy - protecting and pushing towards independence | Not parents' experiences |
| Le, Norris, Reid, Scott, Hartling & Ali (2017) | Development and usability evaluation of an art and narrative-based knowledge translation tool for parents with a child with pediatric chronic pain: Multi-method study | Specifically about experiences of a resource |
| Leksell, Hallberg, Horne, Ernberg, & Hedenberg-Magnusson (2017) | Parenting a child with juvenile idiopathic arthritis, orofacial pain and dysfunction: A qualitative study | Not focused on pain |
| Logan, Guite, Sherry & Rose (2006) | Adolescent-parent relationships in the context of adolescent chronic pain conditions | Not qualitative research |
| MacIver, Jones & Nicol (2014) | Parental experiences of paediatric chronic pain management services | Specifically about experiences of one service |

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|---|---|---|
| McNeill (2007) | Fathers of children with a chronic health condition - beyond gender stereotypes | Not focused on pain |
| Noel, Beals-Erickson, Law, Alberts & Palermo (2016) | Characterizing the pain narratives of parents of youth with chronic pain. | Data coded into pre-determined categories |
| Ogunlana, Oyewole, Falola, Davis, Lateef & Adepoju (2019) | Psychosocial problems among mothers of children with cerebral palsy attending physiotherapy outpatient department of two selected tertiary health centres in Ogun state: A pilot study | Not focused on pain |
| Palermo & Chambers (2005) | Parent and family factors in pediatric chronic pain and disability: An integrative approach | Not research |
| Palermo, Slack, Zhou, Aaron, Ficher & Rodriguez (2019) | Waiting for a Pediatric Chronic Pain Clinic Evaluation: A Prospective Study Characterizing Waiting Times and Symptom Trajectories. | Parents' views not separate |
| Power, Muhit, Heanoy, Karim, Galea, Badawi & Khandaker (2019) | Depression, anxiety and stress among caregivers of adolescents with cerebral palsy in rural Bangladesh | Not qualitative research |
| Rabbitts, Aaron, Fisher, Lang, Bridgwater, Tai & Palermo (2017) | Chronic pain after pediatric surgery: A qualitative study with children, parents, and healthcare providers | Not research |

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|---|---|---|
| Reid, Lander, Scott & Dick (2010) | What do the parents of children who have chronic pain expect from their first visit to a pediatric chronic pain clinic? | Not qualitative research |
| Reiter-Purtill, Gerhardt, Passo, Taylor, Vannatta & Noll (2002) | Child-rearing practices of caregivers with and without a child with juvenile rheumatoid arthritis: The perspectives of caregivers and professional | Not research |
| Roizenblatt, Tufik, Goldenberg, Pinto, Hilario & Feldman (1997) | Juvenile fibromyalgia: clinical and polysomnographic aspects | Not qualitative research |
| Roth (2018) | Ethnography of integrative pain management at a large urban pediatric hospital | Parents' views not separate |
| Ruskin, Campbell, Stinson & Kohut (2018) | Changes in Parent Psychological Flexibility after a One-Time Mindfulness-Based Intervention for Parents of Adolescents with Persistent Pain Conditions | Not parents' experiences |
| Santos de Araujo Dantas, Pontes, Dantas de Assis & Collet (2012) | Families abilities and difficulties in caring for children with cerebral palsy | Not available in English |
| Sherry & Weisman (1988) | Psychologic aspects of childhood reflex neurovascular dystrophy | No analysis |
| Sieberg & Manganella (2015) | Family beliefs and interventions in pediatric pain management | Not research |

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|---|---|---|
| Sieberg, Williams & Simons (2011) | Do parent protective responses mediate the relation between parent distress and child functional disability among children with chronic pain? | Not qualitative research |
| Silver (2004) | The smallest sufferers: Parents and caregivers must truly understand children's pain to treat it effectively | Not research |
| Simoes, Silva, dos Santos, Misko & Bousso (2013) | The parents' experience in taking care of their children with cerebral palsy | Not available in English |
| Simons & Sieberg (2015) | Parents - To help or hinder pain memories in children | Not research |
| Singogo, Mweshi & Rhoda (2015) | Challenges experienced by mothers caring for children with cerebral palsy in Zambia | Not focused on pain |
| Ståhle-Öberg & Fjellman-Wiklund (2009) | Parents’ experience of pain in children with cerebral palsy and multiple disabilities - An interview study | Cognitive impairment |
| Tutelman, Chambers, Urquhart, Fernandez, Heathco, Noel, Flanders, Guilcher, Schulte, Stinson, MacLeod & Stern (2019) | When "a headache is not just a headache": A qualitative examination of parent and child experiences of pain after childhood cancer | Children with cancer |

| | | |
|---|---|---|
| Uziel, Friedland, Jaber, Press, Buskila & Hashkes (2007) | Living with children with growing pains: how does it affect the parents? | Not qualitative research |
| Van Slyke & Walker (2006) | Mothers' responses to children's pain | Not qualitative research |
| van Vlierberghe, Goubert, Bijttebier, Mertens & Crombez (2004) | De invloed van pijncatastroferen op somatische klachten en disfunctioneren bij kinderen en jongeren met chronische pijn: Een vragenlijststudie | Not available in English |
| Vetter, Bridgewater & McGwin (2012) | An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? | Not qualitative research |
| Vetter, Bridgewater, Ascherman, Madan-Swain, McGwin Jr & McGwin (2014) | Patient versus parental perceptions about pain and disability in children and adolescents with a variety of chronic pain conditions | Not qualitative research |
| Violon (1985) | Family etiology of chronic pain | Not research |
| von Baeyer & Whitehead (2006) | Effects of parent attention versus distraction on abdominal discomfort in children: A new method and new findings | Not qualitative research |
| Vowles, Cohen, McCracken & Eccleston (2010) | Disentangling the complex relations among caregiver and adolescent responses to adolescent chronic pain | Not qualitative research |

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|---|---|---|
| Waldman & Perlman (2019) | My child has a disability… Do I have to worry about an opioid addiction too? | Not research |
| Walker & Greene (1989) | Children with recurrent abdominal pain and their parents: More somatic complaints, anxiety, and depression than other patient families? | Not qualitative research |
| Walker, Williams, Smith, Garber, van Slyke & Lipani (2006) | Parent attention versus distraction: Impact on symptom complaints by children with and without chronic functional abdominal pain | Not qualitative research |
| Wallace, McCracken, Weiss & Harbeck-Weber (2015) | The role of parent psychological flexibility in relation to adolescent chronic pain: Further instrument development | Not qualitative research |
| Wallace, Woodford & Connelly (2016) | Promoting psychological flexibility in parents of adolescents with chronic pain: Pilot study of an 8-week group intervention | Specifically about one intervention program |
| Wallrath, Rubel, Ohls, Demiralay & Hechler (2019) | Bottom-up or top-down?: The role of child and parent chronic pain and anxiety in the context of parental catastrophizing and solicitousness | Not qualitative research |
| Walters (1993) | The effect of appraisal, coping, and family variables on stress outcomes of pediatric chronic pain patients | Not available |

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|---|---|---|
| Weiss, Junghans-Rutelonis, Aaron, Harbeck-Weber, McTate, Luedtke & Bruce (2019) | Improving Distress and Behaviors for Parents of Adolescents With Chronic Pain Enrolled in an Intensive Interdisciplinary Pain Program | Not qualitative research |
| Whittingham, Wee, Sanders & Boyd (2013) | Sorrow, coping and resiliency: Parents of children with cerebral palsy share their experiences | Not focused on pain |
| Williams, Smith, Bruehl, Gigante & Walker (2009) | Medical evaluation of children with chronic abdominal pain: Impact of diagnosis, physician practice orientation, and maternal trait anxiety on mothers' responses to the evaluation | Not qualitative research |
| Williamson, Walters & Shaffer (2002) | Caregiver models of self and others, coping, and depression: Predictors of depression in children with chronic pain | Not qualitative research |
| Wilson, Lewandowski & Palermo (2011) | Fear-avoidance beliefs and parental responses to pain in adolescents with chronic pain | Not qualitative research |
| Wilson, Moss, Palermo & Fales (2014) | Parent pain and catastrophizing are associated with pain, somatic symptoms, and pain-related disability among early adolescents | Not qualitative research |
| Wolff, Darlington, Hunfeld, Verhulst, Jaddoe, Hofman, Passchier & Tiemeier (2010) | Determinants of somatic complaints in 18-month-old children: The Generation R Study | Not qualitative research |

| | | |
|---|---|---|
| Wolters, Burns, Martin, Baldwin, Dombi, Toledo-Tamula, Dudley, Gillespie & Widemann (2015) | Pain interference in youth with neurofibromatosis type 1 and plexiform neurofibromas and relation to disease severity, social-emotional functioning, and quality of life | Not qualitative research |
| Woods & Ostrowski-Delahanty (2017) | Psychometric Properties of the Psychosocial Assessment Tool-Chronic Pain Version in Families of Children With Headache | Not qualitative research |
| Zabalia, Jacquet, Grasmenil & Wood (2013) | Pediatric pain assessment: A pragmatic analysis of dialogues in the interactions of healthcare providers, children and their parents | Not parents' experiences |

**Table 4: Details of Papers Included**

| ID | Author (year) | Country | Condition (if specific) | Focus of research | Participants | Sample Size | Methods | Analysis |
|---|---|---|---|---|---|---|---|---|
| 1* | Britton and Moore (2002a) | UK | Juvenile Idiopathic Arthritis | Journey to diagnosis | Parents, patients, siblings | 9 families | Semi-structured interviews; video diaries; written diaries | "Standard ethnographic processes" |
| 2* | Britton and Moore (2002b) | UK | Juvenile Idiopathic Arthritis | Experiences | Parents, patients, siblings | 9 families | Semi-structured interviews; video diaries; written diaries | "Standard ethnographic processes" |

| ID | Author (year) | Country | Condition (if specific) | Focus of research | Participants | Sample Size | Methods | Analysis |
|---|---|---|---|---|---|---|---|---|
| 3 | Brodwall et al. (2018) | Norway | Functional Gastrointestinal Disorders | Experiences | Parents | 14 families (5 fathers, 10 mothers) | Semi-structured interviews | Qualitative content analysis |
| 4 | Carter (2002) | UK | Any pain lasting 3 months or longer, with or without formal diagnosis | Medical encounters | Parents, grandparent, sibling | 3 families | Journals; interviews | Thematic analysis |
| 5 | Gaughan et al. (2014) | USA | Chronic neuropathic pain (Complex Regional Pain Syndrome) | Journey caring for child | Parents | 9 families (8 mothers, 5 fathers) | Open-ended interviews | Qualitative content analysis |
| 6 | Jordan et al. (2016) | UK | Pain for a period of at least 3 months | Experiences | Fathers | 6 | Semi-structured interviews | Interpretative phenomenological analysis |

| ID | Author (year) | Country | Condition (if specific) | Focus of research | Participants | Sample Size | Methods | Analysis |
|---|---|---|---|---|---|---|---|---|
| 7 | Jordan et al. (2007) | UK | Pain for a minimum duration of 3 months | Parental experiences | Parents & carers | 15 families (11 mothers, 5 fathers, 1 grandmother) | Focus groups | Interpretative phenomenological analysis |
| 8 | Le et al. (2019) | Canada | Chronic pain (no definition given but recruited through chronic pain clinic) | Parental experiences | Parents | 13 families (12 mothers, 1 father) | Semi-structured interviews | Thematic analysis |
| 9 | MacIver et al. (2010) | UK | Chronic pain (musculoskeletal or neuropathic) | Impact for parents | Parents | 10 families (10 mothers, 2 fathers) | Interviews | Thematic analysis |
| 10 | McNeill (2004) | Canada | Juvenile Rheumatic Arthritis | Fathers' experiences | Fathers | 22 | Semi-structured interviews | Grounded theory |

| ID | Author (year) | Country | Condition (if specific) | Focus of research | Participants | Sample Size | Methods | Analysis |
|---|---|---|---|---|---|---|---|---|
| 11 | Navarro et al. (2018) | Unclear (data from online forums) | Complex Regional Pain Syndrome | Parental online communication | Parents | 39 users | Online forums | Thematic analysis |
| 12 | Rabbitts et al. (2017) | USA | Major surgery 3-12 months prior | Long-term pain and recovery after surgery | Patients, parents, healthcare providers | 15 families (13 mothers, 1 father, 1 grandmother) | Semi-structured interviews | Thematic analysis |
| 13 | Rossato et al. (2007) | Brazil | Juvenile Idiopathic Arthritis | Family experiences | Parents, patients, siblings | 12 families (12 mothers, 2 fathers) | Semi-structured interviews | Grounded theory |
| 14 | Sallfors and Hallberg (2003) | Sweden | Juvenile Chronic Arthritis | Parental experiences | Parents | 22 families (16 mothers, 6 fathers) | Open interviews | Grounded theory |

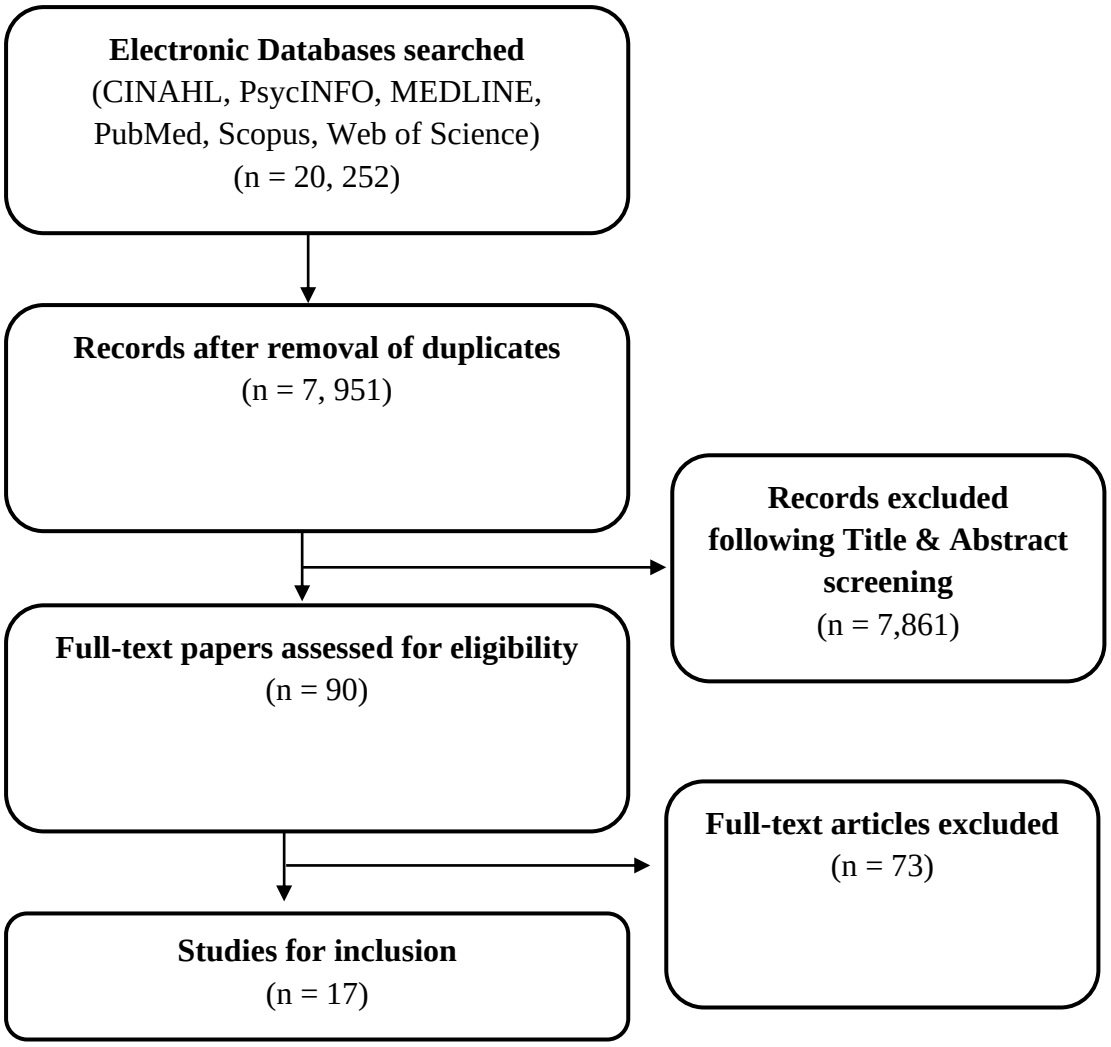
| ID | Author (year) | Country | Condition (if specific) | Focus of research | Participants | Sample Size | Methods | Analysis |
|---|---|---|---|---|---|---|---|---|
| 15 | Smart and Cottrell (2005) | UK | Recurrent abdominal pain (physically unexplained) | Going to the doctors | Mothers | 28 families | Semi-structured interviews | Thematic analysis |
| 16 | Waite-Jones and Madill (2008) | UK | Juvenile Idiopathic Arthritis | Fathers' experiences | Fathers | 8 | Semi-structured interviews | Grounded theory |
| 17 | Yuwen et al. (2017) | USA | Juvenile Idiopathic Arthritis | Parental experiences | Parents | 8 families (8 mothers, 1 father) | Semi-structured interviews | Inductive content analysis |

* Papers 1 and 2 relate to the same sample.

**Table 5: Results of Critical Appraisal using CASP Qualitative Checklist**

| Paper | Clear statement of aims | Qualitative | Research design | Recruitment strategy | Data collection | Consideration of | Consideration of ethical | Sufficiently rigorous | Clear statement of | Consideration of value | Total score |
|---|---|---|---|---|---|---|---|---|---|---|---|
| **1** | 3 | 3 | 3 | 3 | 2 | 3 | 2 | 2 | 3 | 3 | **27** |
| **2** | 3 | 3 | 3 | 3 | 2 | 3 | 2 | 2 | 3 | 3 | **27** |
| **3** | 3 | 3 | 2 | 3 | 3 | 2 | 3 | 2 | 3 | 2 | **26** |
| **4** | 3 | 3 | 2 | 3 | 3 | 1 | 3 | 3 | 3 | 2 | **26** |
| **5** | 3 | 3 | 3 | 3 | 3 | 3 | 2 | 3 | 2 | 2 | **27** |
| **6** | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 2 | **29** |
| **7** | 3 | 3 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 3 | **23** |
| **8** | 3 | 3 | 3 | 2 | 3 | 1 | 2 | 3 | 2 | 3 | **25** |
| **9** | 3 | 3 | 1 | 3 | 2 | 1 | 3 | 3 | 3 | 3 | **25** |
| **10** | 3 | 3 | 1 | 3 | 3 | 2 | 3 | 3 | 2 | 3 | **26** |
| **11** | 3 | 3 | 2 | 3 | 3 | 2 | 3 | 3 | 3 | 3 | **28** |
| **12** | 3 | 3 | 2 | 3 | 3 | 2 | 3 | 3 | 3 | 3 | **28** |
| **13** | 3 | 3 | 2 | 2 | 2 | 1 | 2 | 3 | 3 | 2 | **23** |
| **14** | 3 | 3 | 1 | 3 | 3 | 1 | 2 | 3 | 3 | 3 | **25** |
| **15** | 3 | 3 | 2 | 3 | 2 | 2 | 2 | 3 | 3 | 2 | **25** |
| **16** | 2 | 2 | 1 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | **26** |
| **17** | 3 | 3 | 1 | 3 | 2 | 1 | 3 | 3 | 3 | 3 | **25** |

**Figure 1: PRISMA Flow Diagram (Moher et al., 2009)**



# Appendices

## Appendix 1-A: Development of analytic themes from codes and descriptive themes

The below table reflects the process of development from initial codes (developed from all 17 papers included) to descriptive themes, and then to analytic themes. As can be seen in the table, the descriptive themes did not map directly onto the analytic themes – with several of the descriptive themes being represented within each analytic theme. Rather, the descriptive themes reflect a stage in the iterative process of analysis; their development allowed familiarisation with and deeper understanding of the data. This thereby facilitated the third-order interpretations necessary for development of the final analytic themes.

| Code | Descriptive Theme | Analytic Theme |
|---|---|---|
| "In limbo" due to the pain | Pain takes over | Seeking control in an uncontrollable situation |
| (Lack of) control | | |
| Constantly alert, on guard, vigilant | | |
| Being assertive (is uncomfortable) | The impact of other people | |
| Role of parents in treatment | Parents as a unit | |
| One parent takes the lead | | |
| Want to 'trade places' with the child | | |
| Uncertainty, unpredictability | Stress and difficult emotional responses | |
| Want to make it better | | |
| Availability of information, resources | | |
| Helplessness, hopelessness | | |
| Denial | | |
| Don't know what to do | | |
| Focus on practical things | Moving forwards in positive ways | |
| Developing medical knowledge | | |
| Taking responsibility | | |
| Advocacy on behalf of the child | | |
| Focus on family (instead of on pain) | | |

| | | |
|---|---|---|
| Having to be strong, tough, stay positive | | |
| Silver linings | | |
| Gaining perspective | | |
| Getting on with things, being pragmatic | | |
| Relationships with medical profession | The impact of other people | Being let down by experts and becoming their own expert |
| Let down by services | | |
| Trust in healthcare teams | | |
| HCPs' knowledge of best practice | | |
| (Lack of) understanding of the patient/family's experience | | |
| Validation, feeling invalidated | | |
| Having to explain the condition/diagnosis/symptoms | | |
| Engagement with services, treatment | Parents as a unit | |
| Parents are experts on their own child | | |
| Navigating systems | Stress and difficult emotional response | |
| Frustration | | |
| Empowerment | Moving forward in positive ways | |
| Social burden of pain | The impact of other people | Fearing judgment whilst judging themselves |
| Being believed (or not) | | |
| | | |
| "They don't look ill/in pain/disabled" | | |
| Rejection of family's coping strategies | | |
| Being/feeling/fearing being judged | | |
| Communication between family and services | | |
| Other people's awareness/understanding | | |
| Lack of recognition of severity | | |
| Self-doubt | Stress and difficult emotional responses | |
| Sense of failure as a parent | | |

| | | |
|---|---|---|
| Seeing the child suffer | | |
| Guilt/blame | | |
| Feeling like a broken record | | |
| Inflicting more pain/suffering | | |
| Relief at diagnosis | Moving forwards in positive ways | |
| Life changed due to the child's condition | Pain takes over | Seeking normality even whilst adapting to a 'new normal' |
| Jealousy; comparison with "normal" children/families | Impact of other people | |
| Impact on parental identity | Parents as a unit | |
| Changes in parental roles | | |
| Holistic parental role | | |
| Wanting child to have a normal life | Stress and difficult emotional responses | |
| Not what I expected | | |
| Difficulty acknowledging changes | | |
| Unfair | | |
| New priorities | Moving forwards in positive ways | |
| Process of adjustment, acceptance, adaptation | | |
| Learning to live with it | | |
| Adjusting to a new normal | | |
| Trying to maintain normal activities | | |
| Pain affects the whole family | Pain takes over | Focusing on the child vs. awareness of impact on the wider family |
| Impact on leisure time, social lives | | |
| Life centres around child, their pain | | |
| Financial implications | | |
| Fatigue/exhaustion | | |
| Impact on parents' careers | | |
| Family members feeling excluded/rejected | Parents as a unit | |
| Siblings getting enough attention | | |
| Family functioning | | |
| Closeness between parent(s) and child | | |

| | | |
|---|---|---|
| Impact on relationships within the family | | |
| Pulling together as a family | | |
| Child's response to pain | | |
| Parents recognise pain even when denied by the child | | |
| `Communication within the family | | |
| Resentment | Stress and difficult emotional responses | |
| Confusing for siblings | | |
| Impact on child is difficult | | |
| 'Costs' of caring | | |
| Feeling alone, social isolation | | |
| Fighting for support/resources | Pain takes over | Dichotomy: The push and pull of raising a child with persistent pain |
| Burden of so many appointments | | |
| No choice | | |
| Importance of peer support | Impact of others | |
| Social support | | |
| Sharing experiences with others in similar situations makes it worse | | |
| Support turns into criticism | | |
| Self support | Parents as a unit | |
| Dislike seeking external support | | |
| Careful not to be over-sympathetic; fear causing hypochondria | | |
| Protectiveness of child/family | | |
| Support from partner | | |
| Balance within parents' relationship; completement each other | | |
| Marital tension, disagreements | | |
| Differences between mothers and fathers | | |
| Parents lack insight into each other's perspective | | |
| Hiding feelings (from partner/child/friends) | | |

| | | |
|---|---|---|
| Showing/expressing feelings | | |
| Family has an impact on pain | | |
| Catastrophising, thinking the worst | Stress and difficult emotional responses | |
| Practical difficulties accessing treatment/support | | |
| Difficulties securing diagnosis | | |
| Making difficult decisions | | |
| No outlet for feelings | | |
| “Genuine physical disease” = more real | | |
| Search for physical explanation and treatment | | |
| Recognition of psychological factors in pain | Moving forwards in positive ways | |
| Belief in a cure/miracle | | |
| Hope | | |
| Religion, faith | | |

**Appendix 1-B: Author Guidelines of Chosen Journal for Submission 'Journal of Health Psychology'**

Manuscript Submission Guidelines: Journal of Health Psychology

This Journal is a member of the Committee on Publication Ethics

**Please read the guidelines below then visit the Journal's submission site** http://mc.manuscriptcentral.com/jhealthpsychology **to upload your manuscript. Please note that manuscripts not conforming to these guidelines may be returned.**

Only manuscripts of sufficient quality that meet the aims and scope of Journal of Health Psychology will be reviewed.

Please ensure that your manuscript is suitable for publication and completely free of errors before you submit. Please pay particular attention to SAGE guidelines on Authorship and the SAGE Correction Policy.

There are no fees payable to submit or publish in this journal.

As part of the submission process you will be required to warrant that you are submitting your original work, that you have the rights in the work, and that you have obtained and can supply all necessary permissions for the reproduction of any copyright works not owned by you, that you are submitting the work for first publication in the Journal and that it is not being considered for publication elsewhere and has not already been published elsewhere.

Please see our guidelines on prior publication and note that ***Journal of Health Psychology*** **may accept submissions of papers that have been posted on pre-print servers**; please alert the Editorial Office when submitting (contact details are at the end of these guidelines) and include the DOI for the preprint in the designated field in the manuscript submission system. Authors should not post an updated version of their paper on the preprint server while it is being peer reviewed for possible publication in the journal. If the article is accepted for publication, the author may re-use their work according to the journal's author archiving policy. If your paper is accepted, you must include a link on your preprint to the final version of your paper.

**1. What do we publish?**

**1.1 Aims & Scope**

Before submitting your manuscript to Journal of Health Psychology, please ensure you have read the Aims & Scope.

**1.2 Article Types**

The Editorial Board of the **Journal of Health Psychology** considers for publication:

(a) Reports of empirical studies likely to further our understanding of health psychology
(b) Critical reviews of the literature
(c) Theoretical contributions and commentaries
(d) Intervention studies
(e) Brief reports
(e) Signed editorials (about 1000 words) on significant issues.

**Intervention studies**

Publication guidelines for intervention studies are published in Volume 15, Issue 1, pp. 5-7.The journal normally publishes papers reporting intervention studies of up to 8,000 words allowing 500 words per table and figure.

The Journal of Health Psychology welcomes research reports regardless of the direction or strength of the results. However the JHP will only consider reports of clinical trials that have been pre-registered at http://www.clinicaltrials.gov/ or http://www.controlled-trials.com/

Please consult the Editorial concerning “Publication Guidelines for Intervention Studies in the Journal of Health Psychology” by David F. Marks J Health Psychol January 2010 vol. 15 no. 1 5-

7: http://www.sagepub.com/content/15/1/5.full.pdf+html The criteria for publication include the application of the CONSORT, TREND and PRISMA statements.

**Brief reports**

The Journal also publishes Brief Reports of up to 3,000 words. Brief Reports should include an abstract of 100 words, and may include a table or figure in lieu of 500 words of the 3,000-word maximum.

**Article length and house style**

Articles should be as short as is consistent with clear presentation of subject matter. The word count for articles is 8,000 words, including footnotes and a reference list. Articles over the word count should be ran by the Editor first. Tables and figures count as 500 words each which should be attached as separate pages at the end. "INSERT HERE" signs should be noted within the text. The title should indicate exactly, but as briefly as possible, the subject of the article. It is essential that your literature review is completely up to date. Please check recent issues of the **Journal of Health Psychology** and other key journals to ensure that any relevant papers are cited. Papers that fail to do this will be rejected. An Abstract should be at the start of the manuscript and not exceed **100 words** (in spite of what is stated on the ScholarOne website) accompanied by **five** keywords should be selected from the list provided on the JHP ScholarOne website. References are not numbered but appear in alphabetical order by first author surname.

To enable blind, impartial review, all documentation must be anonymized. A common error is to include the author's name in the Word document title, as in:

Smith (blind copy).doc

Such manuscripts will be rejected for re-submission in fully blinded fashion.

**1.3 Writing your paper**

The SAGE Author Gateway has some general advice and on how to get published, plus links to further resources.

***1.3.1 Make your article discoverable***

When writing up your paper, think about how you can make it discoverable. The title, keywords and abstract are key to ensuring readers find your article through search engines such as Google. For information and guidance on how best to title your article, write your abstract and select your keywords, have a look at this page on the Gateway: How to Help Readers Find Your Article Online

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**2. Editorial policies**

**2.1 Peer review policy**

Journal of Health Psychology operates a strictly blinded peer review process in which the reviewer's name is withheld from the author and, the author's name from the reviewer. The reviewer may at their own discretion opt to reveal their name to the author in their review but our standard policy practice is for both identities to remain concealed.

As part of the submission process you will be asked to provide the names of [X no.] peers who could be called upon to review your manuscript. Recommended reviewers should be experts in their fields and should be able to provide an objective assessment of the

manuscript. Please be aware of any conflicts of interest when recommending reviewers. Examples of conflicts of interest include (but are not limited to) the below:

- The reviewer should have no prior knowledge of your submission
- The reviewer should not have recently collaborated with any of the authors
- Reviewer nominees from the same institution as any of the authors are not permitted

Please note that the Editors are not obliged to invite/reject any recommended/opposed reviewers to assess your manuscript.

**2.2 Authorship**

All parties who have made a substantive contribution to the article should be listed as authors. Principal authorship, authorship order, and other publication credits should be based on the relative scientific or professional contributions of the individuals involved, regardless of their status. A student is usually listed as principal author on any multiple-authored publication that substantially derives from the student's dissertation or thesis.

**2.3 Acknowledgements**

All contributors who do not meet the criteria for authorship should be listed in an Acknowledgements section. Examples of those who might be acknowledged include a person who provided purely technical help, or a department chair who provided only general support.

Any acknowledgements should appear first at the end of your article prior to your Declaration of Conflicting Interests (if applicable), any notes and your References.

***2.3.1 Third party submissions***

Where an individual who is not listed as an author submits a manuscript on behalf of the author(s), a statement must be included in the Acknowledgements section of the manuscript and in the accompanying cover letter. The statements must:

- Disclose this type of editorial assistance – including the individual's name, company and level of input
- Identify any entities that paid for this assistance
- Confirm that the listed authors have authorized the submission of their manuscript via third party and approved any statements or declarations, e.g. conflicting interests, funding, etc.

  Where appropriate, SAGE reserves the right to deny consideration to manuscripts submitted by a third party rather than by the authors themselves.

**2.4 Funding**

Journal of Health Psychology requires all authors to acknowledge their funding in a consistent fashion under a separate heading. Please visit the *Funding Acknowledgements* page on the SAGE Journal Author Gateway to confirm the format of the acknowledgment text in the event of funding, or state that: This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

**2.5 Declaration of conflicting interests**

It is the policy of Journal of Health Psychology to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles.

Please ensure that a 'Declaration of Conflicting Interests' statement is included at the end of your manuscript and on the title page, after any acknowledgements and prior to the

references. If no conflict exists, please state that 'The Author(s) declare(s) that there is no conflict of interest'. For guidance on conflict of interest statements, please see the ICMJE recommendations here

Please see the ICMJE Form for Disclosure of Potential Conflicts of Interest for more information about what items should be referenced in a Conflict of Interest statement.

**2.6 Research ethics and patient consent**

Medical research involving human subjects must be conducted according to the World Medical Association Declaration of Helsinki

Submitted manuscripts should conform to the ICMJE Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals, and all papers reporting animal and/or human studies must state in the methods section that the relevant Ethics Committee or Institutional Review Board provided (or waived) approval. Please ensure that you have provided the full name and institution of the review committee, in addition to the approval number.

For research articles, authors are also required to state in the methods section whether participants provided informed consent and whether the consent was written or verbal. Information on informed consent to report individual cases or case series should be included in the manuscript text. A statement is required regarding whether written informed consent for patient information and images to be published was provided by the patient(s) or a legally authorized representative. Please do not submit the patient's actual written informed consent with your article, as this in itself breaches the patient's confidentiality. The Journal requests that you confirm to us, in writing, that you have obtained written informed consent but the written consent itself should be held by the authors/investigators themselves, for example in a patient's hospital record. The confirmatory letter may be uploaded with your submission as a separate file.

Please also refer to the ICMJE Recommendations for the Protection of Research Participants

**2.7 Reporting guidelines**

These guidelines relate to level of specificity, labels, participation, gender, sexual orientation, racial and ethnic identity, disabilities and age. Authors should also be sensitive to issues of social class, religion and culture.

The relevant EQUATOR Network reporting guidelines should be followed depending on the type of study. For example, all randomized controlled trials submitted for publication should include a completed CONSORT flow chart as a cited figure and the completed CONSORT checklist should be uploaded with your submission as a supplementary file. Systematic reviews and meta-analyses should include the completed PRISMA flow chart as a cited figure and the completed PRISMA checklist should be uploaded with your submission as a supplementary file. The EQUATOR wizard can help you identify the appropriate guideline.

**2.8 Research data**

At SAGE we are committed to facilitating openness, transparency and reproducibility of research. From the 1st July 2020 *Journal of Health Psychology* requires authors to share only those data described in the publication and to submit a Data Sharing Statement alongside their submission. This should appear as a distinct sub-section at the end of the Method section of the manuscript.

The data must be uploaded to the SAGE Track submission system and will be uploaded to Figshare on publication. Please see section 3.4 for information on MIRD data sharing, data uploading and required files and the relevant Editorial for further details.

#### 2.8.1 Data sharing statement

Data sharing statements must indicate the following: whether individual de-identified participant data (including data dictionaries) are shared; what data in particular are shared; additional, related documents that are available (e.g. study protocol and statistical analysis plan). The shared data should be useable and interpretable and include the following features:

1. If the data are in the form of a **statistical dataset**, variables must be labelled clearly, and variables that are stored as labelled numeric values must have associated value labels. The version of the software used to create the dataset must be stipulated (to clarify potential back-compatibility issues).
2. For data stored as a **spreadsheet, or delimited text**, an associated text file containing variable labels and, where appropriate, value labels for labelled numeric data.
3. **Missing data codes** should be documented, together with numbers of missing values for each variable. Ideally, missing data should be left blank, not assigned a pseudo-numeric code.
4. **Measurement units** and **measurement times** (where appropriate).
5. The dataset should be accompanied by a **codebook** giving means of continuous variables and frequencies of categorical variables, together with numbers of valid cases. This allows the use to check that they have read the data correctly into whatever software they are using.
6. Clearly spell out the **analytic procedures** upon which the submitted claims rely, and where possible provide access to all relevant analytic materials.
7. **Note** explaining the datasets will be available in Figshare and as supplementary material on the SAGE Journals platform.

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## 3. Publishing Policies

### 3.1 Publication ethics

SAGE is committed to upholding the integrity of the academic record. We encourage authors to refer to the Committee on Publication Ethics' International Standards for Authors and view the Publication Ethics page on the SAGE Author Gateway

#### *3.1.1 Plagiarism*

Journal of Health Psychology and SAGE take issues of copyright infringement, plagiarism or other breaches of best practice in publication very seriously. We seek to protect the rights of our authors and we always investigate claims of plagiarism or misuse of published articles. Equally, we seek to protect the reputation of the journal against malpractice. Submitted articles may be checked with duplication-checking software. Where an article, for example, is found to have plagiarised other work or included third-party copyright material without permission or with insufficient acknowledgement, or where the authorship of the article is contested, we reserve the right to take action including, but not limited to: publishing an erratum or corrigendum (correction); retracting the article; taking up the matter with the head

of department or dean of the author's institution and/or relevant academic bodies or societies; or taking appropriate legal action.

### 3.1.2 Prior publication

If material has been previously published it is not generally acceptable for publication in a SAGE journal. However, there are certain circumstances where previously published material can be considered for publication. Please refer to the guidance on the SAGE Author Gateway or if in doubt, contact the Editor at the address given below.

### 3.2 Contributor's publishing agreement

Before publication, SAGE requires the author as the rights holder to sign a Journal Contributor's Publishing Agreement. SAGE's Journal Contributor's Publishing Agreement is an exclusive licence agreement which means that the author retains copyright in the work but grants SAGE the sole and exclusive right and licence to publish for the full legal term of copyright. Exceptions may exist where an assignment of copyright is required or preferred by a proprietor other than SAGE. In this case copyright in the work will be assigned from the author to the society. For more information please visit the SAGE Author Gateway

### 3.3 Open access and author archiving

Journal of Health Psychology offers optional open access publishing via the SAGE Choice programme. For more information please visit the SAGE Choice website. For information on funding body compliance, and depositing your article in repositories, please visit *SAGE Publishing Policies* on our Journal Author Gateway.

### 3.4 Transparency, Openness and Replication Policy

From the 1st July 2020, *Journal of Health Psychology* requires all authors to make their data fully accessible for all empirical research submitted to the journal for publication, and will only consider manuscripts which follow an open publication model with M = Mandatory, I = Inclusion (of), R = Raw, D = Data (MIRD). According to the MIRD model, all contributions of new qualitative and quantitative studies must fully document and share the raw data collected by the author(s) or their data collection team together with full details of the analytical procedures used. All data and analytical procedures must be sufficiently well described to enable a third party with the appropriate level of expertise to replicate the data analyses.

Authors must include their raw data and disclose the key aspects of the research design to every extent possible. The raw data and associated contextual information will be sent to reviewers, revised alongside the paper in every round and published alongside the paper (as an appendix or online supplement). In addition to publishing the raw data with the article, the data must be shared through a digital repository. Authors have to use data citation practices that identify a dataset's author(s), title, date, version, and a persistent identifier, for example a Digital Object identifier (DOI).

The MIRD data sharing principle will be applied to all empirical studies, not only clinical trial report data:

1. As of 1 July 2020, manuscripts concerning clinical trials and other empirical studies that are submitted to *Journal of Health Psychology* must contain a data sharing statement as delineated in section 2.8 Research Data.

2. Any clinical trial that begins enrolling participants and is intended for later submission to *Journal of Health Psychology* must include a data sharing plan in the trial's registration.

It is *Journal of Health Psychology* policy that authors submit detailed information on empirical analysis alongside their written article. Authors should upload *at least* the first four files listed below when they submit their article.

- data set
- syntax file(s) from the software that has been used for the analysis;
- explanatory memo: explaining enclosed files/material and their content including help with regard to the analysis, which is important when non-standard techniques have been used; this may also apply to qualitative work; also some information on the software used for the analysis, including its version, is required;
- log file(s): output with results from the software that has been used for the analysis;
- Additional data analysis, including robustness analyses

Authors must provide a separate readme PDF listing all included files and documenting the purpose and format of each file provided, as well as instructing a user on how a replication can be conducted.

Making datasets publicly available is mandated by *Journal of Health Psychology* policy. Authors should ensure that they are uploading to the *Journal of Health Psychology* SAGE Track submission site, all data to do with their article. Once the article is accepted and published, it will be automatically uploaded to the Figshare repository.

The manuscript will not be moved through to Peer Review, or to Production until the editor is satisfied that all relevant data has been submitted alongside the manuscript. If cited data are restricted (e.g. classified, require confidentiality protections, were obtained under a non-disclosure agreement, or have inherent logistical constraints), authors must notify the editor at the time of submission. The editor shall have full discretion to follow the journal's policy on restricted data, including declining to review the manuscript of granting an exemption with or without conditions. The editor shall inform the author of this decision prior to review.

In addition to sharing the raw data, *Journal of Health Psychology* requires authors to delineate clearly the analytic procedures upon which their published claims rely and, where possible, provide access to all relevant analytic materials.

### 3.4.1 Replication studies

*Journal of Health Psychology* encourages the submission of replication studies regardless of whether or not the findings are statistically significant. Normally replication studies fall within of one or more of the following types:

***Theoretical replication:*** The submitted article argues that the original theoretical model is missing at least one key element. The missing element(s) are addressed and included in the empirical analysis;

***Technical replication*:** The submitted article identifies faults in the original research design or analysis, thereby arguing that the original results might not hold; and/or

***Concept replication****:* The submitted article questions the validity of the original study. An alternative measurement or operationalisation is proposed which yields different substantive results.

### *3.4.2 Preregistration of Studies and Analysis Plans*

Researchers conducting experimental studies are encouraged to consider pre-registering their research design in advance with an established registry. *Journal of Health Psychology* will publish papers where authors indicate the conducted research was preregistered with an analysis plan in an independent, institutional registry (e.g., http://clinicaltrials.gov/) of studies involves registering the study design, variables, and treatment conditions. Including an analysis plan involves specification of sequence of analyses or the statistical model that will be reported.

For preregistered studies, the following requirements apply:

1. Authors must, in acknowledgments or the first footnote, indicate that research was preregistered in an independent, institutional registry (with name and link to its location) with an analysis plan;
2. The author must:
    1. confirm in the text that the study was registered prior to conducting the research with links to the time-stamped preregistration(s) at the institutional registry, and that the preregistration adheres to the disclosure requirements of the institutional registry or those required for the preregistered badge with analysis plans maintained by the Center for Open Science.
    2. report all pre-registered analyses in the text, or, if there were changes in the analysis plan following preregistration, those changes must be disclosed with explanation for the changes clearly distinguish in text analyses that were preregistered from those that were not, such as having separate sections in the results for confirmatory and exploratory analyses (these changes are added as a separate document linked to the text of the main paper)

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**4. Preparing your manuscript for submission**

Please ensure that your manuscript is suitable for publication and completely free of errors before you submit. Please pay particular attention to SAGE guidelines on Authorship and the SAGE Correction Policy.

**4.1 Formatting**

The preferred format for your manuscript is Word. LaTeX files are also accepted. Word and (La)Tex templates are available on the Manuscript Submission Guidelines page of our Author Gateway.

**4.2 Language and terminology**

Authors must follow the Guidelines to Reduce Bias in Language of the Publication Manual of the American Psychological Association (6th ed). These guidelines relate to level of specificity, labels, participation, gender, sexual orientation, racial and ethnic identity, disabilities and age. Authors should also be sensitive to issues of social class, religion and culture.

The language used in your manuscript should be inclusive and language that might be deemed sexist or racist should not be used. All submissions should avoid the use of pejorative terms and insensitive or demeaning language and submissions that use unacceptable language will be returned by the editor.

Useful websites to refer to for guidance

We recommend that authors consider looking at the below guidance:

- APA guidelines on Bias Free Language
- Words Matter
- Authors are encouraged to refer to land use any language guidelines that relate specifically to their research

**4.3 Artwork, figures and other graphics**

For guidance on the preparation of illustrations, pictures and graphs in electronic format, please visit SAGE's *Manuscript Submission Guidelines*

Figures supplied in colour will appear in colour online regardless of whether or not these illustrations are reproduced in colour in the printed version. For specifically requested colour reproduction in print, you will receive information regarding the costs from SAGE after receipt of your accepted article.

**4.4 Supplemental material**

This journal is able to host additional materials online (e.g. datasets, podcasts, videos, images etc) alongside the full-text of the article. For more information please refer to our guidelines on submitting supplementary files

**4.5 Reference style**

Journal of Health Psychology adheres to the SAGE Harvard reference style. View the SAGE Harvard guidelines to ensure your manuscript conforms to this reference style.

If you use *EndNote* to manage references, you can download the SAGE Harvard EndNote output file.

**4.6 English language editing services**

Authors seeking assistance with English language editing, translation, or figure and manuscript formatting to fit the journal's specifications should consider using SAGE Language Services. Visit SAGE Language Services on our Journal Author Gateway for further information.

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**5. Submitting your manuscript**

*Journal of Health Psychology* is hosted on SAGE Track, a web based online submission and peer review system powered by ScholarOne™ Manuscripts.

Visit http://mc.manuscriptcentral.com/jhealthpsychology to login and submit your article online.

IMPORTANT: Please check whether you already have an account in the system before trying to create a new one. If you have reviewed or authored for the journal in the past year it is likely that you will have had an account created. For further guidance on submitting your manuscript online please visit ScholarOne Online Help.

**5.1 ORCID**

As part of our commitment to ensuring an ethical, transparent and fair peer review process SAGE is a supporting member of ORCID, the Open Researcher and Contributor ID. ORCID provides a unique and persistent digital identifier that distinguishes researchers from every other researcher, even those who share the same name, and, through integration in key research workflows such as manuscript and grant submission, supports automated linkages between researchers and their professional activities, ensuring that their work is recognized. The collection of ORCID iDs from corresponding authors is now part of the submission process of this journal. If you already have an ORCID iD you will be asked to associate that

to your submission during the online submission process. We also strongly encourage all co-authors to link their ORCID ID to their accounts in our online peer review platforms. It takes seconds to do: click the link when prompted, sign into your ORCID account and our systems are automatically updated. Your ORCID iD will become part of your accepted publication's metadata, making your work attributable to you and only you. Your ORCID iD is published with your article so that fellow researchers reading your work can link to your ORCID profile and from there link to your other publications.
If you do not already have an ORCID iD please follow this link to create one or visit our ORCID homepage to learn more.

**5.2 Information required for completing your submission**

You will be asked to provide contact details and academic affiliations for all co-authors via the submission system and identify who is to be the corresponding author. These details must match what appears on your manuscript. The affiliation listed in the manuscript should be the institution where the research was conducted. If an author has moved to a new institution since completing the research, the new affiliation can be included in a manuscript note at the end of the paper. At this stage please ensure you have included all the required statements and declarations and uploaded any additional supplementary files (including reporting guidelines where relevant).

**5.3 Permissions**

Please also ensure that you have obtained any necessary permission from copyright holders for reproducing any illustrations, tables, figures or lengthy quotations previously published elsewhere. For further information including guidance on fair dealing for criticism and review, please see the Copyright and Permissions page on the SAGE Author Gateway
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**6. On acceptance and publication**

**6.1 SAGE Production**

Your SAGE Production Editor will keep you informed as to your article's progress throughout the production process. Proofs will be made available to the corresponding author via our editing portal SAGE Edit or by email, and corrections should be made directly or notified to us promptly. Authors are reminded to check their proofs carefully to confirm that all author information, including names, affiliations, sequence and contact details are correct, and that Funding and Conflict of Interest statements, if any, are accurate. Please note that if there are any changes to the author list at this stage all authors will be required to complete and sign a form authorising the change.

**6.2 Online First publication**

Online First allows final articles (completed and approved articles awaiting assignment to a future issue) to be published online prior to their inclusion in a journal issue, which significantly reduces the lead time between submission and publication. Visit the SAGE Journals help page for more details, including how to cite Online First articles.

**6.3 Access to your published article**

SAGE provides authors with online access to their final article.

**6.4 Promoting your article**

Publication is not the end of the process! You can help disseminate your paper and ensure it is as widely read and cited as possible. The SAGE Author Gateway has numerous resources

to help you promote your work. Visit the Promote Your Article page on the Gateway for tips and advice.

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**7. Further information**

Any correspondence, queries or additional requests for information on the manuscript submission process should be sent to the Journal of Health Psychology editorial office as follows:

David Marks PhD: editorjhp@gmail.com

**Section Two: Research Paper**

# The Impact of Complex Regional Pain Syndrome (CRPS) on Personal and Social Identity

Word Count: 7961

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Prepared for: Psychology & Health

## Abstract

**Objective:** This qualitative study explored the experiences of individuals living with complex regional pain syndrome (CRPS), and its impact on their identity.

**Design:** The research design was informed by interpretative phenomenological analysis. Six participants completed online semi-structured interviews. These were transcribed verbatim, with individual and cross-case analysis conducted.

**Results:** Four key themes were identified: (1) It's taken time to feel like 'me' again; (2) CRPS alienates me from other people; (3) Having this condition is shameful; and (4) The importance of control to a sense of self.

**Conclusion:** Onset of CRPS symptoms leads to a sudden change in an individual's abilities and lifestyle, impacting on their work, hobbies, and relationships. These changes are at the cost of their sense of self, and adapting to the changes is a process which takes time. Participants found having CRPS shameful and felt that it alienated them from others. The findings offer understanding to health professionals, including psychologists, working with this group to adjust to their diagnosis.


Key words: *Qualitative, Complex Regional Pain Syndrome, Identity, Pain, Interpretative Phenomenological Analysis*

## Introduction

Complex regional pain syndrome (CRPS) is a neurological condition causing pain in a localised area (Goh, Chidambaram, & Ma, 2017). The incidence rate is estimated at 5.5-26.2/100,000 person-years (de Mos et al., 2007; Sandroni, Benrud-Larson, McClelland, & Low, 2003), with women being affected around 3-4 times more commonly than men (Castillo-Gusmán et al., 2015; de Mos et al., 2007). Onset of CRPS generally follows an injury – for example fracture or surgery – although the Royal College of Physicians (RCP) acknowledges that in around 9% of cases, no precipitating organic injury is identified (RCP, 2018). Generally symptoms remain confined to one limb, but in some cases they can 'spread' (van Rijn et al., 2011).

Symptoms of CRPS are variable and can include pain, swelling, skin discolouration, changes in sweating and/or skin temperature, changes in hair and/or nail growth, and motor dysfunction (Birklein, Ajit, Goebel, Perez, & Sommer, 2018). In a study of more than 1,000 patients with CRPS, pain was typically described as 'stabbing', 'burning', or 'dragging', and was exacerbated by touch, by physical effort, and at night-time (Ott & Maihöfner, 2018). Diagnosis is on the basis of clinical assessment, according to set criteria (Merskey & Bogduk, 1994); no conclusive diagnostic test is available. CRPS is divided into subtypes I and II, dependent upon the presence of major nerve damage, with Type I (without major nerve damage) being the more common (Harden et al., 2010).

CRPS is not a straightforward condition and has been subject to much controversy. The condition was first described in detail in 1864 (Mitchell, Morehouse, & Keen, 1982), with Mitchell later coining the term 'causalgia' meaning 'burning pain'; various other names have been used (Sebastin, 2011) before the current nomenclature was adopted. Borchers and Gershwin (2014) acknowledge that CRPS is, even now, "a syndrome steeped in confusion and often inaccuracy" (p. 243). Though it goes some way to reconciling the various names

for the condition, the name ‘CRPS’ has itself been criticised as being vague and inaccurate (Schott, 2007). It is suggested that, given the variability between patients with CRPS – who can in theory have entirely different sets of symptoms, excepting the pain, yet receive the same diagnosis – the condition is perhaps not one homogenous disease, but rather something of a ‘catch-all’ diagnosis given to patients with, “a ‘funny’ pain in a ‘funny-looking’ limb” (p. 148, Schott, 2007).

There is currently no cure for CRPS, and research suggests that although symptoms often reduce with time, only around 5% of patients are symptom-free at twelve months post-onset (Bean, Johnson, Heiss-Dunlop, & Kydd, 2016). Treatment may include physiotherapy, medication, and/or psychological therapy, and should be tailored to individual presentation (Bharwani, Dirckx, & Huygen, 2017) – though the National Institute for Health and Care Excellence (NICE) do not make any recommendations for management, citing insufficient evidence (O'Connell, Wand, McAuley, Marston, & Moseley, 2013). Psychological evaluation and support is important in managing the impact of CRPS upon psychological wellbeing, particularly mood; Lee et al. (2014) found that around 70% of patients with CRPS meet diagnostic criteria for depression and are at risk of suicidal ideation. Indeed, anecdotally, CRPS is often referred to as “the suicide disease” within the patient community (Binkley & Katznelson, 2020).

The need for research into CRPS from a lived experience perspective has been highlighted; for example, in their narrative review, Johnston, Oprescu, and Gray (2015) concluded that such research may enhance healthcare providers’ understanding of CRPS and inform future treatment guidelines. A recent qualitative study by Beales et al. (2021) explored the lived experience of CRPS, identifying four key themes including the life-changing impact of CRPS; variable experiences of care; making sense of CRPS; and lessons learnt from living with CRPS. In terms of the ‘life-changing impact of CRPS’, Beales et al. briefly discussed

the “isolating nature of CRPS and the loss of identity” and “the loss of the former healthy self and the pain-free self” (p. 11). This consideration of identity and self follows on from extensive research on the impact of long-term health conditions upon identity, though Beales et al. did not explore identity in depth. Long-term illness has been described as ‘biographical disruption’ (Bury, 1982) and, as summarised by Oris et al. (2018), individuals living with a long-term illness must integrate their condition into their identity to allow continued wellbeing.

Beales et al.’s (2021) observation regarding the loss of healthy and pain-free selves is consistent with the work of Charmaz (1983), who described the loss of former self-image in long-term illness, and the lack of opportunity to develop a valued new image – Charmaz suggested this occurs cumulatively, leading to ever-increasing impact on personal identity. In addition, reduced ability to work, pursue hobbies, and maintain relationships may also have an effect on individuals’ identity (Charmaz, 1983). The role of identity has been highlighted in recovery in a variety of conditions including mental illnesses (Wisdom, Bruce, Saedi, Weis, & Green, 2008), diabetes (Luyckx et al., 2008), and chronic fatigue syndrome (CFS) (Larun & Malterud, 2007). Identity is argued to be essential to living a meaningful, purposeful, and coherent life, contributing to psychological wellbeing (Thoits, 1986).

As Charmaz suggests, long-term illness may impact identity through disruption of relationships and valued activities; Wisdom et al. (2008) considered how internalisation of negative social responses to an illness or diagnosis may also alter the way people see themselves. For example, Adams, Pill, and Jones (1997) discussed the social stigma of an asthma diagnosis, and noted the lengths to which some of their participants went in avoiding confrontation with their diagnosis, including avoiding previously enjoyed activities. In other words, to avoid what they perceived to be a stigmatised social identity as an asthmatic, these participants avoided activities which were part of their valued identity, thereby compounding

the effect of their illness upon their identity. Humans have evolved to live in social groups, and thus, our identities as members of society are integral to our sense of self (Jetten et al., 2017), with the social groups and intimate relationships people belong to contributing to their identity (Stets & Burke, 2000). Recent research by Packham, Wainio, and Wong (2020) investigated the impact of CRPS on intimate relationships. Their participants described the renegotiation of social relationships, changes in their roles within relationships, and the impact of their condition on their self-perceived attractiveness to partners. This is of course an important aspect of identity, since one's identity is at least partially constructed, developed, and maintained in relationships with other people (Stryker & Burke, 2000).

In addition to this impact on relationships, Vlaeyen, Morley, and Crombez (2016) considered persistent pain (that is, pain lasting longer than three months, which is one of CRPS's major symptoms) generally, concluding that it interferes with daily activities and 'life tasks', leading to loss of roles and challenges to sense of self. Further, a review by Yu, Norton, Harrison, and McCracken (2015) on identity in people with persistent pain concluded that negative self-evaluation affects daily functioning, whereas a sense of self aside from pain is associated with better daily functioning. However, CRPS is relatively unique among pain conditions (Lee et al., 2014); in particular, unlike many persistent pain conditions, CRPS is associated with visible changes, as discussed above. Jacoby, Snape, and Baker (2005) discussed how visible difference can contribute to the impact of a condition upon identity, and yet it is unclear how this may be experienced by CRPS patients. Additionally, CRPS has been associated with neglect-like symptoms, as seen in stroke, for example with patients failing to attend to or care for their affected limb (Lewis, Kersten, McCabe, McPherson, & Blake, 2007); whilst stroke survivors have been found to experience a negative sense of self post-stroke (Ellis-Hill & Horn, 2000), it is again unclear whether or how this may be experienced by CRPS patients.

Furthermore, CRPS is the subject of some disbelief and controversy in the medical world, with some healthcare professionals believing that it is not a legitimate diagnosis, or disbelieving patients about the presence and/or intensity of their symptoms (Beales et al., 2021; Chang, McDonnell, & Gershwin, 2019; Schott, 2007). Beales et al. reported that this disbelief contributed to patients' feelings of isolation. This disbelief is common among pain conditions (Newton, Southall, Raphael, Ashford, & LeMarchand, 2013) and is certainly not unique to CRPS; for example Asbring (2001) discusses similar issues with regard to CFS and fibromyalgia, concluding that this has implications for patients' experiences. However, how this factor may interplay with the other differences discussed above is not clear.

The present study, therefore, aimed to explore participants' experiences of living with chronic CRPS, and how living with CRPS affects their identity. As argued by Johnston et al. (2015), an improved understanding of the experience and impact of chronic CRPS will be valuable to clinical psychologists and other health professionals supporting these patients. The research question was: "How does living with chronic CRPS impact upon personal and social identity?"

## Method

### Design

Given the exploratory nature of the research, interpretative phenomenological analysis (IPA) was selected as an appropriate means of studying participants' experiences of living with CRPS, and the impact this has upon their identity. IPA is considered a relevant approach within health psychology, allowing researchers to gain greater understanding of individuals' experiences, beyond a purely biomedical approach (Brocki & Wearden, 2006). IPA allows for an exploration of lived experience, and individuals' reflections upon this experience, without reducing it to predefined categories (Smith, Flowers, & Larkin, 2009). IPA has its roots in phenomenology (the examination of human experience) and hermeneutics

(the theory of interpretation); thus, IPA is concerned with experiences and how these are interpreted, both first-hand (by the participants) and second-hand (by the researcher/s). This is known as the *double hermeneutic* of IPA (Smith et al., 2009).

In addition, IPA is an idiographic approach, meaning there is a focus on the particular. This is in contrast with many psychological methods, which tend to focus more on the group or population level, aiming to establish general understandings of human behaviours. More specifically, the idiographic nature of IPA means that there is a focus on the details of the phenomena of interest, and that these are interpreted and understood from the perspective of being experienced by particular people, in particular contexts (Smith et al., 2009). The epistemology of IPA, and the one I adopted, is that people are able to reflect on and provide insights (through their accounts) to their thoughts, beliefs, attitudes, and behaviours. Drawing on elements of social constructionism, in which knowledge production is viewed as an inherently social activity, IPA acknowledges the role of the social context in which accounts are produced and that of the researcher in producing and interpreting participant accounts. Thus, IPA is neither purely 'realist' nor social constructionist, but occupies a middle-ground, having an epistemology that can be best characterised as 'critical realism'.

**Participants**

Individuals were eligible to participate if they (a) were aged eighteen years or older; (b) had had a diagnosis of CRPS for a minimum of twelve months (confirmed by self-report); (c) were able to take part in an online interview in English; and (d) were able to give informed consent to participate, i.e. did not suffer from significant cognitive impairment. Participants were recruited internationally online via a two-pronged approach. Firstly, a number of potential participants were identified from an internet search for online articles about living with CRPS. Those who gave an email address or contact form were approached

and invited to participate in an interview. A total of five individuals were contacted in this way, three of whom agreed to participate. Secondly, an additional three participants were recruited via advertisements on social media sites; an advert was shared by relevant charities on Facebook, as well as by the researcher on Twitter. Interested parties were requested to contact the researcher by email.

A total of six participants (five female) aged 20-37 (mean = 29.7 years) took part in the study. Four were from the US, one from Canada, and one from the UK. Three participants were employed (though often with reduced or flexible hours), two were unemployed, and one was a full-time student. Duration of symptoms ranged from 2-13 years (mean = 5.6 years). All participants identified as white with the exception of Amy[1], who identified as white/Hispanic. Demographic details are presented in Table 1 below.

<Insert Table 1 about here>

**Procedure**

Potential participants expressed interest by contacting the researcher via email; they were then emailed the Participant Information Sheet and Consent Form (see Ethics section). A minimum of 48 hours was allowed for the participant to read the information provided, and ask any questions, before a convenient time to complete an interview was agreed. Participants were asked to ensure they were somewhere comfortable, quiet, and private for the anticipated duration of the interview. At the beginning of each interview, the researcher checked that the participant had read the information sheet, and again gave opportunity for the participant to ask questions. Informed consent was given verbally with participants being asked to agree to each item on the consent form before giving overall consent to participate. At the end of their

[1] Not her real name – all participants were assigned pseudonyms, which are used throughout this paper.

interview, participants were given opportunity to ask any additional questions of the researcher, following which they were sent the Participant Debrief (see Ethics section).

**Data Collection**

Semi-structured interviews ranging in length from 63 to 157 minutes (mean = 95 minutes) were conducted by the researcher. All interviews were completed via video conferencing (Skype and Microsoft Teams). The rationale for this was twofold; firstly, the timing of the research meant that social distancing due to COVID-19 was in force; and secondly, since the majority of the participants lived internationally, face-to-face interviews were not practicable. All interviews were video-recorded using in-program recording features particular to the software.

An interview guide was developed, informed by previous research relating to the impact of long-term health conditions on identity (Adams et al., 1997; Asbring, 2001; Larun & Malterud, 2007; Lempp, Scott, & Kingsley, 2006; Piot-Ziegler, Sassi, Raffoul, & Delaloye, 2010). Input was sought from healthcare professionals working with individuals with CRPS – one clinical psychologist and one physiotherapist. Two CRPS charities were also contacted to seek feedback on the interview guide from individuals living with CRPS; unfortunately neither charity responded. Care was taken to consider the unique aspects of CRPS including the visual changes often present and the controversy around nomenclature. Questions fell into three broad areas: *Background*, including demographic questions as well as questions about official diagnosis and journey to diagnosis (e.g. What is your official diagnosis and when did you receive this?); *Understanding of CRPS*, including questions about the individual's understanding of their condition, and the understanding of those around them including family and healthcare professionals (e.g. How would you explain CRPS to someone who hasn't heard of it before?); and *Impact of/response to CRPS*, including questions about how CRPS has affected the way they see themselves and the way others see them (e.g. Do you

think you have changed as a result of CRPS – if so, in what ways?) This was used to guide the conversation with each participant; cues were also taken from participants in terms of what was most important to them, and topics they wished to discuss (Smith, Flowers & Larkin, 2009). (See Ethics section for full interview guide)

**Data Analysis**

Analysis began with verbatim transcription of each interview – including noting pauses, non-verbal communications (e.g. laughter), and gestures (e.g. nodding). The process of transcription contributes to immersion in, and therefore familiarity with, the data (Smith et al., 2009). Each transcript was then considered in turn, according to the steps outlined by Smith et al. The first transcript was read and re-read (step one), which allowed the analyst to "enter the participant's world" (Smith et al., p. 82). This progressed into the initial notation phase (step two), during which the researcher began to identify specific ways in which the participant speaks about, thinks about, and understands the issue. The product of this phase of analysis was a deeper engagement with the participant's recounting of their experience, and a series of initial comments which began to draw out the participant's meaning-making and highlight the way language was used to convey this meaning.

Next came the development of emerging themes (step three), which began during the previous stage of analysis, as connections between notations began to form. In this stage, then, the aim was to reduce the volume of data whilst maintaining the complexity of the participant's narrative; here there lies an analytic shift, as the analyst began to work primarily with the notations rather than the transcript itself, though as Smith et al. state these notes should be closely tied to the original transcript. An example of the development of notations and emergent themes can be seen in Appendix 2-A. Next, these emergent themes were brought together, again through an iterative process which involved grouping and re-grouping of seemingly related emergent themes, until an overall understanding was arrived at (step

four). The final outcome for the first transcript, then, was a number of themes which encapsulate the essence of the participant's experiences insofar as they relate to the research question.

These stages were repeated for each of the remaining transcripts (step five), starting afresh each time – that is, bracketing the themes identified in previous transcripts so as to allow new themes to emerge for each subsequent participant. Themes for each participant, along with a supporting quotation for each theme, can be seen in Table 2.

<Insert Table 2 about here>

Finally, once themes had been generated for all six transcripts, these were compared and contrasted (step six), with themes being used to support and further illuminate each other. At this stage the analysis became more interpretative, as inferences were made about how participants' experiences may link with each other, where similarities and differences may lie, and how elements from one case may aid understanding of another. The overall outcome, then, was a number of themes which relate to the data set as a whole, with varying levels of interpretation and induction. These themes offer one way of interpreting how participants' identities were impacted by living with CRPS. Table 3 shows contributions made by each participant to the final overall themes identified.

<Insert Table 3 about here>

**Ethical Considerations**

The research project was reviewed by Lancaster University Faculty of Health and Medicine's Research Ethics Committee (REC) and ethical approval was granted. See Ethics section for the full ethics application.

To safeguard their anonymity, each participant has been assigned a pseudonym which is used throughout the reporting of this research project. Every effort has been made to ensure that all potentially identifying information has been removed from quotations used.

However, participants were made aware that there is a chance they may be recognised by people who know them, and those recruited from their online writing were additionally made aware that they may be recognised by people who have read their articles.

Completing interviews online, and with overseas participants, presented the question of how to manage any distress which arose during interviews. Following advice from the REC, a plan was made with each participant prior to commencing their interview with regards to how to manage any distress – for example, establishing whether there was someone at home with the participant, or someone they would like the researcher to contact, should they become upset. In the event, this situation did not arise, with no distress beyond what could be managed within the interview.

**Quality of the Data**

The principles of Yardley (2000) were followed to improve quality and validity of the research. Yardley highlights the importance of sensitivity to context; commitment and rigour; transparency and coherence; and impact and importance. Emergent themes and overall themes were reviewed by the research supervisors to improve validity, with themes being amended based on feedback from supervisors.

In keeping with the 'transparency and coherence' element of Yardley's (2000) guidelines, a reflexive stance was taken throughout, recognising the fact that all analysis and interpretations will be impacted by the researcher's own beliefs and biases (Smith et al., 2009). As someone who has never personally experienced persistent pain, nor any of the neurological symptoms associated with CRPS, the researcher had limited insight into the realities of life with such a condition. Their perceptions and preconceptions were, inevitably, shaped by societal beliefs and biases relating to individuals with persistent pain. In addition, having previously worked in a physical health psychology service which supported individuals with persistent pain, the researcher did approach the interviews with some prior

understanding of the potential impacts of living with persistent pain and related symptoms. There was a risk, then, that these preconceptions could contribute to the researcher unintentionally influencing participants, for example through leading questions. Therefore, efforts were made to ask open questions, and a reflective diary was used to aid 'bracketing' of any assumptions. For example, notes were made throughout the research process, considering the researcher's decisions, reactions, and responses to various stages of the project. Thus, biases and expectations became conscious and explicit rather than implicit, which meant their impact upon analysis could be managed. Further consideration is given to this matter in the Critical Appraisal.

## Findings

This process of analysis yielded four themes: (1) It's taken time to feel like 'me' again; (2) CRPS alienates me from other people; (3) Having this condition is shameful; and (4) The importance of control to a sense of self. These themes are discussed in greater detail below.

### Theme 1: It's taken time to feel like 'me' again

All six participants discussed a process of change in their sense of self, whereby CRPS stopped them feeling like themselves; over time, this seemed to gradually resolve, as participants developed ways of incorporating parts of their 'old self' into their new situation. Participants were even able to identify 'benefits' to having CRPS, including learning about themselves and identifying their strengths.

The onset of symptoms of CRPS was often associated with a sudden change in activities, including work and hobbies, which for some participants were key parts of their identity – this seemed especially true for Rachel, who did gymnastics as a hobby as well as working in the field. She described this as "my identity" and "my life"; the loss of both her work and hobby, then, required a huge adjustment for Rachel and caused significant distress:

“I couldn’t watch gymnastics on TV for quite a while”. Rachel spoke about the difficulties inherent in dating, and uncertainty about what to tell potential partners about herself: “You tell someone, ‘Oh, I used to be athletic, I used to be this and that’ and then you’re like, ‘But now I don’t really do much…’” Rachel’s dilemma here reflects the extent to which she felt the loss of her identity associated with her work and hobby – to the point where she did not know what else to tell people about herself.

David, too, appeared to experience a sudden shift in his sense of self upon having to give up work; he described how, “Work was my life”, and that earning a good salary and being successful were important to him. Understandably, then, David struggled to adjust from being independent to his fiancée having to care for him whilst also singlehandedly earning enough money for the two of them since David was no longer able to work. This shift may have been particularly difficult for David to reconcile, given society’s expectations of men being the main breadwinners.

In addition to the impact on their activities, participants noted changes in their mindsets – for instance, becoming more pessimistic; in relation to this, David spoke about how at one point he had been unable to recognise himself: “I couldn’t find any shred of myself at that time… I… thought that I had lost who I was.”

However, with time, participants felt they arrived at a place of feeling more like themselves again, often by incorporating ‘old’ parts of themselves or finding ways to adapt to their new circumstances – for instance, Beth spoke about factors she had to take into consideration to spend time with her friends: “If I go on nights out and stuff, I have to know I'm going with people that I can literally drag off the dance floor, to go and sit down”. Her acceptance that socialising looked different than it may have pre-CRPS meant that she was still able to enjoy spending time with her friends, thus retaining an important part of her identity as a student. Louisa, meanwhile, spoke about the process of coming to terms with her

new self – “I’m just learning now how to love myself again” – which she attributed to a mindfulness course she was completing. Louisa was able to identify parts of her ‘old’ self which were still present, such as previous voluntary work being mirrored in current fundraising for CRPS charities.

Participants felt that having CRPS had taught them about themselves, and also that they were more compassionate towards other people as a result, as described by Amy: “Now I’m more aware of it [invisible illness or disability], and I understand the pain that other people can be going through”. Participants described feeling that they were better people, due to the struggles they had been through, and several participants described being grateful to CRPS for this, going so far as calling it a “blessing in disguise” (Louisa, Amy, and Jen). It was important to several participants to use their experiences to help others, which took various forms; some wrote online about their experiences, whilst fundraising and awareness-raising were also mentioned. Rachel described feeling “humbled” that she was able to help other people in similar situations to hers; helping others appeared to allow participants to feel more like their old selves, particularly for those who had spent time volunteering or otherwise helping others prior to their diagnosis.

**Theme 2: CRPS alienates me from other people**

Alongside challenges to their sense of self, participants felt alienated by their condition, feeling they were markedly different from others, and that other people – both medical professionals as well as friends, family, and strangers – were unable to understand. There was a sense of the stigma of being seen as ‘crazy’ or having their symptoms attributed to mental health difficulties rather than a ‘real’ physical condition. In addition, participants were further alienated by their reduced ability to ‘join in’, which meant that their social circles were diminished.

Amy, Jen, and Beth spoke about the process of obtaining a diagnosis leaving them feeling that they were 'crazy' or somehow imagining the pain, compounded by people (both health professionals and family members) who assumed they were faking their symptoms. As Beth explained, "You start going through in your head, like- are you crazy? Because… you can feel this pain, but everyone's telling you there's no reason". It was a relief, then, to finally be given a diagnosis – even when the diagnosis given did not offer much in the way of hope, additional information, or reliable treatment plan. Regardless, the initial response from these participants was one of relief to have a physical, rather than mental or emotional, explanation for their symptoms, as Jen described: "I felt relief and validation – like, I'm not crazy!" The use of the word 'crazy' in this way speaks to the stigma which still surrounds mental health conditions, and the way in which society takes physical health conditions more seriously and sees them as more legitimate than mental health diagnoses.

Meanwhile, Louisa felt that her responses to the diagnosis and efforts to access treatment led others to think she was 'crazy' unless she carefully monitored and managed her responses. She described becoming upset in her doctor's office at the difficulty she was facing in finding an effective way of managing her symptoms; in response, she was told that crying makes her 'look bad' and 'look crazy'. Louisa stated that she had, "learnt not to cry in [doctors'] offices when possible", reflecting that she felt the need to closely control her emotional responses in order to be taken seriously. Again, this reflects the stigma in society towards mental health difficulties, and the notion that anyone becoming emotional must be 'crazy'.

In addition to these feelings, and accusations, of being 'crazy', a lack of understanding from family and friends led to participants feeling isolated or less able to socialise. Losing friends following diagnosis was a common experience, in part due to physical restrictions including fatigue meaning that they were no longer being able to join in

with the same activities or socialise in the same ways. Rachel described how her social circle "dwindled", which she found hard as she is a very sociable person. Beth felt that her friends thought she didn't try hard enough to join in with them, resulting in her feeling left out of group activities she would previously have been invited to: "I think they perceive me as someone that doesn't come out and doesn't do things with them and doesn't try hard enough." In contrast, Jen felt that not being invited to activities she couldn't participate in was a sign of respect, and did not take this personally: "If I don't get invited to something, I know it's because they're thinking about my disease. Like when I see my friends go kayaking, they know I'm not gonna do it. So they don't even try to invite me, and I'm totally cool with it." Jen felt able to invite her friends to things that she *could* do and appeared happy with this dynamic within her relationships.

**Theme 3: Having this condition is shameful**

In addition to participants' feelings of alienation from others, it seemed that there was something shameful about CRPS, with some participants expressing their dislike of the condition, their affected limb(s), and by extension themselves – this theme seemed most salient for Louisa, David, Jen, and Beth. This shame is perhaps linked with the issue discussed in theme two above, whereby participants felt they were 'crazy' when awaiting a diagnosis; although physical health conditions are more widely understood and taken more seriously than mental health conditions, there is still a certain amount of stigma surrounding disabilities, particularly invisible disabilities. This is illustrated in the way Beth felt she had to "come clean" about her diagnosis to people, like it was some kind of dirty secret; she stated, "I can't really hide it from people and just be like, 'Oh, I'm normal!'" – thereby suggesting that she was somehow abnormal. The contribution made by society's view of disability was overtly recognised by Beth when she spoke about internalised stigma she experienced in relation to issues such as using mobility aids or other accommodations: "It came back to that

stigmatisation, internally, that was like, ‘Oh, am I disabled enough to need all these adaptions?’” This reluctance to see herself as ‘disabled enough’ to need such aids as crutches or a disabled parking permit suggests Beth experienced shame about her condition and the accommodations she required. Similarly, Jen spoke about the shame of having to ask for help, and the feelings of humiliation this provoked in her: “It’s humiliating, but you gotta [ask for help] when you need to. I want to say there’s no shame in it, but you do feel shame”. Again, there is a suggestion here of internalised stigma which tells disabled people that it is shameful to need help.

Several participants stated that they ‘hated’ or ‘strongly disliked’ their affected limbs, suggesting shame about changed abilities and/or appearance; David expressed that he would call his affected limb names and say things like, “just chop it off”. He stated that CRPS made it easier for him to hate himself, whilst Louisa acknowledged that her “strong dislike” of her affected limbs probably connected with why she strongly dislikes herself at times, too. These emotions did appear linked with changes in appearance or visible difference, for example Louisa mentioned that she thought her affected limbs were “funny-looking” whilst Jen discussed feeling uncomfortable wearing certain clothes that meant the changes were visible – “I’m still a little embarrassed, like with the atrophy”. Further, Jen described being horrified by what she saw in the mirror, sharing, “I thought I was the scary monster people tell their kids about”. This statement powerfully captures the shame that participants seemed to feel about the physical representations of their CRPS. There is a juxtaposition here between participants seeming to wish their condition was visible so it would be taken more seriously (and indeed, being relieved to be given a diagnosis of a physical condition), and yet feeling abject shame and horror at the ways in which their condition *was* visible to others.

**Theme 4: The importance of control to a sense of self**

Besides the shame they felt, a sense of control felt important to several of the participants. Having CRPS caused some participants, understandably, to feel they had lost control of their bodies and their lives, prompting them to fight to regain some control, whilst in other cases CRPS had actually provided a means of taking back some control. This theme appeared salient for Louisa, Amy, Jen, and Rachel.

Participants spoke about ways in which they fought to maintain a sense of control, for example by planning for the future or working hard to maintain a positive outlook. Jen described herself as a "control freak" and discussed how planning for the future had helped her to feel in control. For Jen, feeling like a burden or asking for help was unacceptable; at times when she did have to ask for help, this was experienced as humiliating. It seemed that asking for help was an explicit admission of not being in control, hence Jen finding it so intolerable; there are clear links here with the shame discussed in theme 3. Similarly, Louisa tried hard to maintain a positive outlook; there was a sense here of Louisa fostering an optimistic persona and almost forcing herself to focus on the positives or, in her own words, "Fake it [happiness] 'til you make it". This, too, seemed to be about control –Louisa seemed to feel if she could control her outlook and the personality she presented, she would feel more in control of her overall situation.

Amy, meanwhile, felt that her diagnosis with CRPS had allowed her to make changes in her life and take back control: "I think CRPS, as horrible as it was, it woke me up. It was like, hey, you are in control, you can change the course of your life!" For example, Amy spoke about previously doing a job she didn't enjoy and not having time for her passions. She believed CRPS had acted as a wake-up call, prompting her to quit her job and become self-employed in a role that aligned more closely with her values; in essence, she felt more authentically herself now than she was prior to her diagnosis: "I wasn't really in alignment with what my passions were. I went to school for something creative, and yet I was

working in this boring office job!" In contrast, Rachel described feeling quite out-of-control in her life, which was not turning out how she had expected; she compared herself to her younger brother, and to her own expectations of herself, and felt things were very different for her than she had hoped: "I thought I would be married by now, have kids- or if I wasn't, I would be on my own, more independent, have my own place.". This being 'out of control', and not where she expected to be, seemed to contribute to a sense of Rachel not quite being sure who she was anymore.

## Discussion

This project aimed to understand the ways in which CRPS impacts upon individuals' identity. In particular, the key themes highlighted by participants related to a process of change in their sense of self, which has taken time; a feeling of alienation from others; the shame associated with their condition; and the importance of control in their sense of self.

In the first theme, participants discussed the loss of their sense of self, and the process of re-incorporating elements of their 'old' self into their new circumstances, whereby they came to feel like themselves again. Charmaz (1995) discussed the process of adaptation, arguing this was one way in which individuals sought to live with a long-term health condition resulting in impairment or loss of bodily function. Charmaz defined 'adaptation' as the process by which people adjust their life and sense of self to accommodate their limitations, thereby reunifying their body and self; this does seem to reflect the processes described in the first theme, and this research thereby provides examples of the ways in which adaptation may occur. For example, Louisa mentioned fundraising for CRPS charities where in the past she may have done voluntary work; in adapting her activities to her new circumstances in this way, Louisa was able to retain an important aspect of her sense of self.

In a similar vein, the work of Bury (1982) considered long-term illness as 'biographical disruption', insofar as "the structures of everyday life and the forms of

knowledge which underpin them are disrupted" (p. 169). This kind of disruption was evident for the participants in the present study, who described interruptions to their work, hobbies, and relationships. However, Bury's work was completed with individuals with rheumatoid arthritis, hence his description of the onset of chronic illness being insidious rather than sudden; this is in direct opposition to the experience of participants in the present study, many of whom described a sudden onset of symptoms (with the exception of David and Beth, whose CRPS both developed following a pre-existing condition and was therefore perceived as less sudden). It is interesting to note, then, that the experience of biographical disruption described by Bury appears to be consistent with the experiences of participants involved in the present study, despite very different onsets of the conditions studied. This is perhaps a logical outcome, given that a sudden onset would seem to incur more biographical disruption than a more gradual change. This re-negotiation of participants' identities echoes findings of Packham et al. (2020), whose participants referenced CRPS leading them to have to re-negotiate their social relationships; re-negotiation in one form or another is perhaps a common element of the experience of living with CRPS.

In addition to this process of redeveloping their sense of self, participants spoke about shame related to their condition. They discussed ways in which they disliked themselves and their affected limb(s), as well as shaming themselves for needing help or becoming more dependent upon others. There appeared to be shame and internalised stigma associated with having what was largely an invisible condition, with participants questioning at times whether they were 'crazy'. The theory of stigma proposed by Goffman (2009) described stigma as 'spoiled identity' – with an individual becoming stigmatised when something marks them out as different from the norm. This is certainly the case for such rare conditions as CRPS, and indeed any disability or long-term health condition is stigmatising to a degree (Susman, 1994). It is perhaps not surprising, then, that shame and stigma were

identified as a theme in this study; this research provides insight into the particular elements of living with CRPS which are perceived to be shameful, including changes in appearance and loss of independence.

Invisible disabilities are associated with a stigma of their own, whereby the fact that people's condition cannot be seen means that the symptoms they describe can be ignored; appropriate, necessary accommodations and exemptions from obligations may also be ignored (Charmaz, 2019). Individuals with invisible long-term conditions or disabilities may therefore be reluctant to disclose their diagnosis, through fear of being ignored or discredited (Charmaz, 2019). This was reflected by participants in the present study; for example, Beth was reluctant to apply for a disabled parking permit as she was not a full-time wheelchair user. In an interesting juxtaposition here, alongside this internalised stigma relating to living with a largely invisible condition, participants in the present study were simultaneously distressed about and ashamed of the visible symptoms of their condition, including swelling, skin discolouration, and atrophy. Kent and Thompson (2014) discuss the development and maintenance of shame in visible difference, which they argue can be linked with perceived confirmation of existing negative beliefs about the self. This would certainly seem to fit for David, who reported that the CRPS made it easier for him to find things about himself to dislike.

In addition to the shame they felt about their condition, participants spoke about CRPS alienating them from others, due in part to the physical limitations brought about by their condition. There may be a parallel here whereby participants also feel alienated from their own bodies, as described by Svenaeus (2015); this would certainly fit with the feelings of shame and stigma discussed above. This feeling of alienation is consistent with the social model of disability, which suggests that disability arises at least partly through barriers in society which preclude participation of differently-abled individuals (Oliver, 2013). Thus,

participants' symptoms may not necessarily have been so disabling (and by extension, alienating) if society were more accessible. These barriers also act at a psycho-emotional level, serving to remind disabled people that they are different (Reeve, 2014), and perhaps thereby reinforcing internalised stigma. The implications of understanding disability in this way include the need for change at a systemic level in order to make society accessible to all, and to do so in a way which does not further contribute to psycho-emotional disablism (Reeve, 2004).

As well as this alienation, the final theme considered the importance of control in participants' sense of self. Stets and Serpe (2013) discuss the way in which individuals seek control over their identities, contrasting obligatory identities (e.g. parent, partner, employee) with voluntary identities (e.g. friend, athlete, choir member). As Stets and Serpe highlight, voluntary identities are often selected because an individual benefits from them in some way. There is an element of control insofar as an individual can choose their voluntary identities, whereas obligatory identities may be more socially determined. Furthermore, having more voluntary identities appears to be associated with higher self-esteem, mastery, and lower distress than does having more obligatory identities (Thoits, 2003). And yet, it is voluntary identities which seemed to be most impacted in participants' experiences of CRPS – for example, Rachel discussed no longer being able to participate in gymnastics, which had previously been a central part of her identity. This loss of control over their voluntary identities can be understood to contribute to participants' distress.

**Clinical Implications**

Findings of the present study, alongside previous research on identity (Larun & Malterud, 2007; Luyckx et al., 2008; Wisdom et al., 2008), suggest this is an important area for health professionals to consider when working with people with CRPS.

In particular, participants spoke about the importance of incorporating elements of their 'old' selves into their new situation; this was a process which could perhaps be facilitated and supported through therapeutic intervention. Acceptance and Commitment Therapy (ACT), with its focus on 'valued direction' (Wilson, Sandoz, Kitchens, & Roberts, 2010), may be a relevant framework, particularly for individuals whose distress relates to a sense of loss of self or loss of valued activities. A core component of ACT is identification of values and commitment to acting in line with these values; thus, intervention using an ACT framework could aid individuals in recognising their values and developing ways to live in line with these, despite limitations due to CRPS. Indeed, in a systematic review of randomised controlled trials with adults with persistent pain, ACT has been found to be efficacious in enhancing functioning and reducing distress (Hann & McCracken, 2014).

Alternatively, particularly for individuals whose distress relates to the perceived shame of their condition, there may also be benefits to a Compassion Focused Therapy (CFT) -informed approach. As Leaviss and Uttley (2015) discussed in their systematic review, CFT is a promising therapeutic model with individuals whose distress relates to shame and self-criticism. CFT supports individuals to take a more compassionate stance towards themselves, which seems particularly relevant for participants who dislike themselves or make self-deprecating remarks. As Kılıç et al. (in-press) concluded in their systematic review, therapies focused on self-compassion led to improved outcomes for individuals with long-term physical health conditions; improvements were noted in anxiety, depression, stress, and sleep problems.

**Limitations and Future Research**

As with all research, the present study was subject to several limitations, which highlight potential areas for future focus, or improvements which could be made in future research. Firstly, online recruitment meant that participants' diagnoses could not be verified.

It is possiblethat one or more participants may not have met formal diagnostic criteria for CRPS; future research could reduce this risk by recruiting through formal medical channels, which would allow verification of diagnosis. This was not possible for the present study due to COVID-19 restrictions.

Another limitation of the present study relates to the relatively homogenous sample involved, as required for IPA; for example, all participants were between the ages of 20 and 37. The present study does not, therefore, offer any insight into experiences of older people with CRPS. Given changes in identity which can occur with age anyway (for example as discussed by Weiss and Lang (2012)), a potential area of interest for future research may be in exploring the impact of CRPS on identity in older individuals, particularly since highest incidence rates of CRPS have been reported in women aged 61-70 (de Mos et al., 2007). Experiences of this group may prove different, and additional clinical implications would need to be considered when working with older individuals with CRPS. Furthermore, the participants in this sample were overwhelmingly white, with only one participant identifying otherwise, and all were from Western countries. Again, future research involving non-White and/or non-Western participants may be beneficial, particularly given cultural differences in how pain is understood – for example, as Orhan et al. (2018) identified in their systematic review, there is some evidence of cross-cultural differences in coping strategies, illness perceptions, and self-efficacy with chronic musculoskeletal pain.

**Conclusion**

This study used IPA to explore six participants' experiences of living with CRPS, in particular the impact this has on their identity. Participants spoke about the time taken to feel like 'themselves' again; a sense of alienation from others; shame and stigma related to living with CPRS; and the importance of control in their sense of self. These findings have implications for support offered by healthcare professionals to people living with CRPS.

Future research to broaden the scope of the present study, for example to older adults and those from differing cultural backgrounds, would be beneficial.

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## Tables & Figures

**Table 1: Participants' demographic details**

| Pseudonym | Gender | Age | Location | Location of CRPS | Duration of Symptoms | Employment status |
|---|---|---|---|---|---|---|
| Louisa | Female | 32 | Canada | Arm, spread to leg | 3.5 years | Employed (reduced hours) |
| David | Male | 34 | USA | Foot | 2 years | Unemployed |
| Rachel | Female | 26 | USA | Foot | 5 years | Unemployed |
| Amy | Female | 37 | USA | Hand | 8 years | Self-employed |
| Jen | Female | 29 | USA | Arm, spread to head | 13 years | Employed (flexible) |
| Beth | Female | 20 | UK | Foot | 2 years | Student |

**Table 2: Themes identified for each participant**

| Participant | Themes | | | | | |
|---|---|---|---|---|---|---|
| **Louisa** | **I cannot show emotional responses, or people think I am crazy**<br>"Dr H told me to stop crying because it makes me look bad. … She also told me that I was crazy. A lot. I look really crazy when I'm in doctors' offices, cos I'm really desperate for hope." | **I dislike the CRPS, and therefore myself**<br>"I strongly dislike them [affected limbs]. And I think they're funny-looking to … I don't like them, but they're still attached!" | **I've had to reinvent myself**<br>"I've had to reinvent myself, so to say, to be comfortable again." | **My role is to help others (but no-one helps me)**<br>"I'm the wings who cover everybody, and make sure everything's okay for everybody else, and no-one puts me under their wings." | **I have to focus on the positives**<br>"That's the way I have to look at everything, is the glass is half-full" | |
| **David** | **A shift in priorities**<br>"The focus is off me. The focus is more, I guess you would say I've become more of a hippy… I want to help people." | **Worthless, I hated myself**<br>"I used to call it [affected limb] names. I said it was a chicken leg or a dead leg or a dead foot." | **People didn't understand**<br>"It's hard to really explain it where people get it – other than them going, 'It can't be that bad'. Um, it sure is, it sure is that bad." | **I mustn't be a burden**<br>"At the beginning I would put on a happy face, even though I was in pain, and not share that because I didn't want anyone else to have to be burdened by it, 'cos it was mine." | **CRPS broke me to make me anew**<br>"I think this whole experience has made me a stronger and more open individual." | |
| **Rachel** | **This is not how I expected life to be**<br>"I wanted to be on my own, more independent, I wanted to work or… Travel more, or at least have my own money and not rely | **It's hard to keep the sociable part of me with CRPS**<br>"Now I'm like, 'Nope, I'm done' around like 5:00 or 6:00pm, and that's definitely changed my schedule." | **Not being able to do gymnastics meant losing a huge part of myself**<br>"When I was told I can no longer do gymnastics or be involved in anything of that sort, I had a meltdown. I cried. | **Guilt that my suffering is minimal compared to others**<br>"I don't have that fire feeling that I did those two nights [when I was first diagnosed]. And people have it every day, so I feel guilty | **I'm still myself but in new ways**<br>"I think I came to terms with it early and it was like okay, this is what I have, I just wanna know what to do with it, how to live with it, | **Other people don't really understand**<br>"I'm like trying to kind of prove to them [family members] what it is, and that I'm not faking it. And it's kind of like, I shouldn't have to, but in your head |

| | | | | | | |
|---|---|---|---|---|---|---|
| | on my mom. Just be my own person." | | That was kind of like my identity through elementary to high school, college…" | sometimes, being like, I don't really know what you're living with." | and what comes next." | you're like, 'Please believe me!'" |
| **Amy** | **I'm in control**<br>"I feel like now that I have the tools and the resources… I know what works for me, it gives me that empowerment." | **I felt like I was going crazy**<br>"When they tell you, 'Oh it's your nervous system, or it's your brain telling you…' and it's like, great, so I'm crazy? … I must be crazy, because obviously my hand's fine, the X-ray says it." | **Our struggles become our strengths**<br>"It's been such a struggle, but because of it, I've become stronger and I've learned so much from it." | **I'm curious**<br>"I'm all about getting to the root cause 'cos I don't want to just, you know, cover up the symptoms, I want long term health. And I'm very curious about everything." | **I feel different than other people with CRPS**<br>"I've shared my story [with the CRPS community online] and people got really mad at me, I got a lot of hate emails." | |
| **Jen** | **I make other people uncomfortable**<br>"I've had people break up with me because of my disease. It's a lot, okay, it was a lot back then cos I wasn't as great as I am now." | **I'm a control freak**<br>"When you can't walk and you need someone to shower you, and help you in the bathroom – humiliating. Absolutely humiliating." | **I thought I was the scary monster people tell their kids about**<br>"I used to look at myself in the mirror, when I first got diagnosed, and I was horrified… And I used to cry, and throw things at my mirror thinking I was a monster." | **I've become the person I always wanted to be**<br>"I was a very cold person before ,and now I'm very warm and positive. So it was a blessing in disguise." | **CRPS does not define me**<br>"I truly believe that I am not my disease, it does not define me. Yes, I have it. And yes, I have to take care of it and rearrange certain parts of my life for it, but it's not who I am." | |
| **Beth** | **I have to "come clean" about my condition**<br>"This is something that I have to be pretty honest with straight away because I can't really | **I felt like I was going crazy**<br>"There was always a thing in the back of my mind going, 'Do I have it? Do I actually have this? | **I'm disabled and I'm trying to be okay with that**<br>"I was like, 'Am I sick enough to need all these adaptations?' or like, 'Am I disabled | **People don't understand**<br>"Like, the amount of people that are like, 'What do you mean, storms hurt?'" | **I always have to consider the CRPS**<br>"When you go on nights out and stuff, so many people wear heels and smarter shoes and stuff, and | **I'm still me, but in adapted ways**<br>"Everyone tells me I'm a determined person and I'm just kind of like- I guess I get on with things, no matter what." |

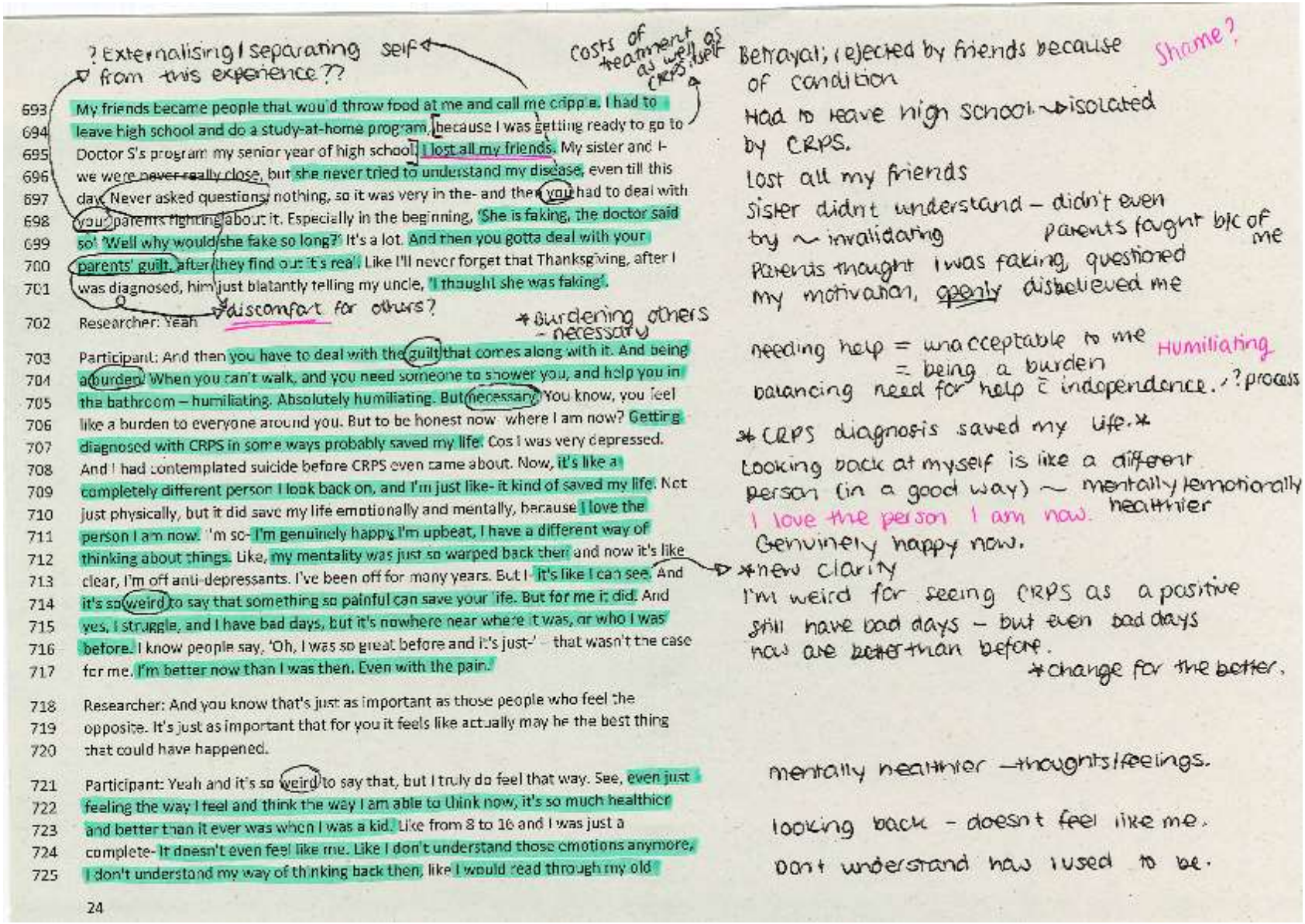
| | | | | | | |
|---|---|---|---|---|---|---|
| | hide it from people and just be like, ‘Oh, I’m normal!’” | Or is it just me being crazy?’” | enough to need all these?’… It is like, oh, I do actually need this.” | | I’m just standing there in my trainers.” | |

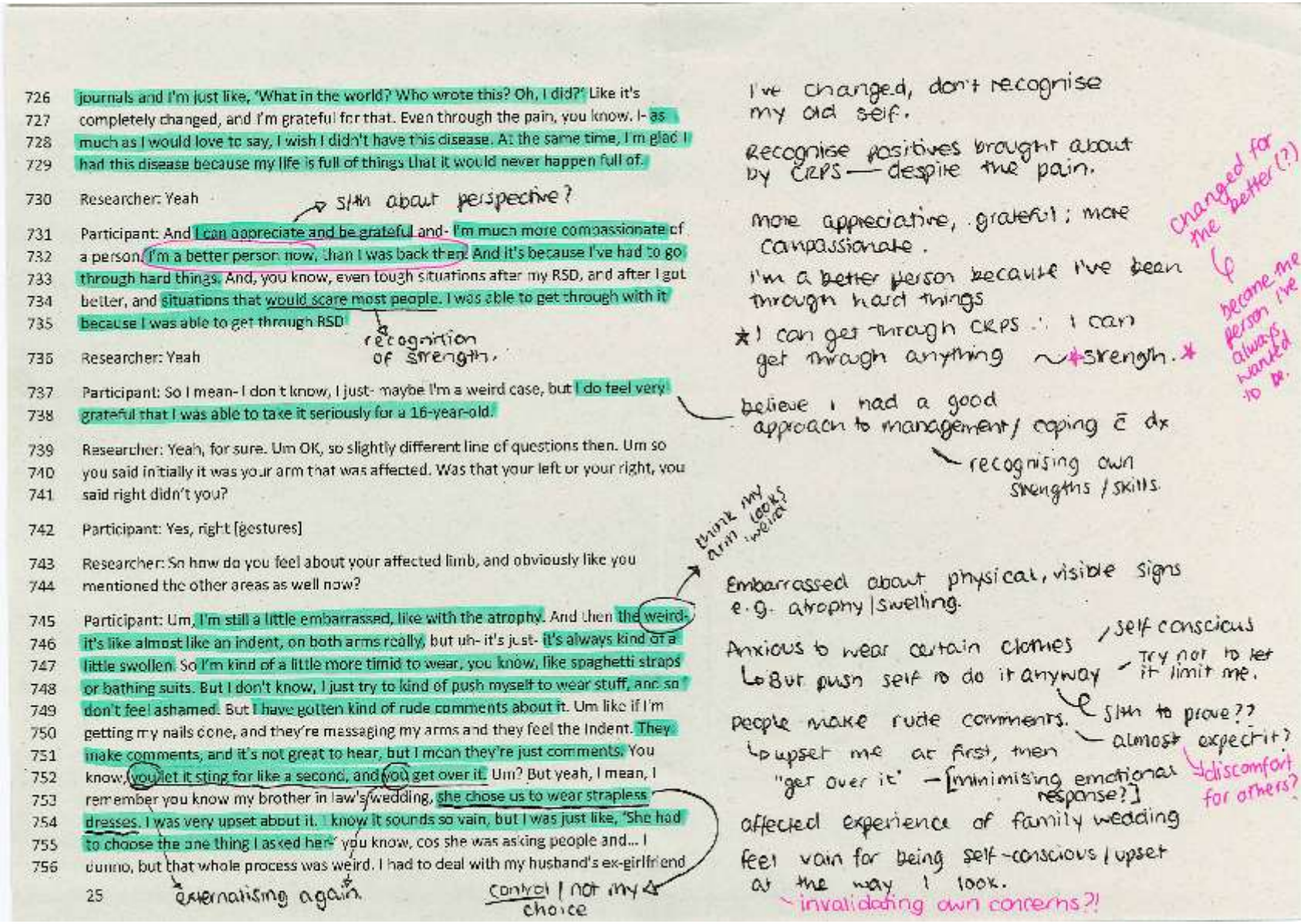
**Table 3: Contributing themes from each participant to overall themes**

| | Louisa | David | Rachel | Amy | Jen | Beth |
|---|---|---|---|---|---|---|
| **Theme 1: It's taken time to feel like 'me' again** | • I've had to reinvent myself | • CRPS broke me to make me anew | • I'm still myself but in new ways | • Our struggles become our strengths | • I've become the person I always wanted to be | • I'm still me but in adapted ways<br>• I always have to consider the CRPS |
| **Theme 2: CRPS alienates me from other people** | • I cannot show emotional responses, or people think I am crazy | • People didn't understand | • Other people don't really understand | • I felt like I was going crazy | • I make other people uncomfortable | • I felt like I was going crazy<br>• People don't understand |
| **Theme 3: Having this condition is shameful** | • I dislike the CRPS and therefore myself | • Worthless, I hated myself | — | — | • I thought I was the scary monster people tell their kids about | • I have to "come clean" about my condition<br>• I'm disabled and I'm trying to be okay with that |
| **Theme 4: The importance of control to a sense of self** | • I have to focus on the positives | — | • This is not how I expected life to be | • I'm in control now | • I'm a control freak | — |

# Appendices

## Appendix 2-A: Example of notations and emerging themes for one participant





726 journals and I'm just like, 'What in the world? Who wrote this? Oh, I did?' Like it's
727 completely changed, and I'm grateful for that. Even through the pain, you know. I- as
728 much as I would love to say, I wish I didn't have this disease. At the same time, I'm glad I
729 had this disease because my life is full of things that it would never happen full of.

730 Researcher: Yeah

731 Participant: And I can appreciate and be grateful and- I'm much more compassionate of
732 a person. I'm a better person now, than I was back then. And it's because I've had to go
733 through hard things. And, you know, even tough situations after my RSD, and after I got
734 better, and situations that would scare most people. I was able to get through with it
735 because I was able to get through RSD.

736 Researcher: Yeah

737 Participant: So I mean- I don't know, I just- maybe I'm a weird case, but I do feel very
738 grateful that I was able to take it seriously for a 16-year-old.

739 Researcher: Yeah, for sure. Um OK, so slightly different line of questions then. Um so
740 you said initially it was your arm that was affected. Was that your left or your right, you
741 said right didn't you?

742 Participant: Yes, right [gestures]

743 Researcher: So how do you feel about your affected limb, and obviously like you
744 mentioned the other areas as well now?

745 Participant: Um, I'm still a little embarrassed, like with the atrophy. And then the weird-
746 it's like almost like an indent, on both arms really, but uh- it's just- it's always kind of a
747 little swollen. So I'm kind of a little more timid to wear, you know, like spaghetti straps
748 or bathing suits. But I don't know, I just try to kind of push myself to wear stuff, and so I
749 don't feel ashamed. But I have gotten kind of rude comments about it. Um like if I'm
750 getting my nails done, and they're massaging my arms and they feel the indent. They
751 make comments, and it's not great to hear, but I mean they're just comments. You
752 know, you let it sting for like a second, and you get over it. Um? But yeah, I mean, I
753 remember you know my brother in law's wedding, she chose us to wear strapless
754 dresses. I was very upset about it. I know it sounds so vain, but I was just like, 'She had
755 to choose the one thing I asked her-' you know, cos she was asking people and... I
756 dunno, but that whole process was weird. I had to deal with my husband's ex-girlfriend

25

I've changed, don't recognise my old self.

Recognise positives brought about by CRPS — despite the pain.

more appreciative, grateful; more compassionate.

I'm a better person because I've been through hard things

★I can get through CRPS ∴ I can get through anything ~ *strength.*

Believe I had a good approach to management/coping ∈ dx

recognising own strengths / skills.

Changed for the better (?) & become the person I've always wanted to be.

think my arm looks weird

Embarrassed about physical, visible signs e.g. atrophy | swelling.

Anxious to wear certain clothes / self conscious

But push self to do it anyway — Try not to let it limit me.

people make rude comments. — s/th to prove??

upset me at first, then — almost expect it?

"get over it" — [minimising emotional response?] — discomfort for others?

affected experience of family wedding

feel vain for being self-conscious / upset at the way I look.

~invalidating own concerns?!

externalising again

control / not my choice

**Appendix 2-B: Author Guidelines of Chosen Journal for Submission 'Psychology & Health'**

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**Section Three: Critical Appraisal**

Word Count: 3977

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The critical appraisal aims to allow further reflections on the research process. I will first give a summary of the findings of each paper, considering links between the two, and implications for practice. I will then explore the strengths and limitations of the research paper, as well as some reflections on the process of carrying out this study. I will also consider the impact of the COVID-19 pandemic upon my research.

## Overview of findings

The research paper explored the lived experience of people with complex regional pain syndrome (CRPS), and in particular the impact of living with this condition on identity. An interpretative phenomenological analysis (IPA) approach was taken (Smith, Flowers, & Larkin, 2009). Six semi-structured interviews were completed and transcribed verbatim. Analysis yielded four themes: (1) It's taken time to feel like 'me' again; (2) CRPS alienates me from other people; (3) Having this condition is shameful; and (4) The importance of control to a sense of self. These findings were discussed in terms of their place within the extant literature, as well as potential implications for psychological support for these individuals.

A systematic review and thematic synthesis of qualitative papers relating to parenting a child with persistent pain identified six themes: (1) Seeking control in an uncontrollable situation; (2) Being let down by experts and becoming their own expert; (3) Fearing judgment whilst judging themselves; (4) Seeking normality even whilst adapting to a 'new normal'; (5) Focusing on the child vs. awareness of the impact on the wider family; and (6) Dichotomy: The push and pull of raising a child with persistent pain. Again, these findings were considered in terms of their contribution to the literature and their potential implications for support offered to parents of children with persistent pain.

Despite the differences between the two papers in terms of the population of interest, there were commonalities between the findings. The theme of control was raised in both

papers; in the research paper, participants highlighted the importance of control, at least in some respects, to their sense of self – for example, Louisa[2] spoke about her need to maintain a positive approach, which seemed to be a means of retaining control. Meanwhile, in the literature review, parents spoke about seeking control in a situation which felt wildly out-of-control for them.

The role of control in how people cope with long-term health conditions and persistent pain has been recognised for some time; for example, as Williams and Koocher (1998) discussed, loss of control due to long-term illness is a key contributor to psychological distress amongst both patients and family members. Further, as Bates and Rankin-Hill (1994) concluded, individuals with an internal locus of control (that is, perceived control of wellbeing is being located within, rather than externally) appear more able to successfully cope with persistent pain. Bates and Rankin-Hill suggest that interventions targeting locus of control could help individuals living with persistent pain to cope more positively with their symptoms. There are implications here, then, for professionals supporting people with CRPS, who may consider supporting individuals to regain a sense of control as a means of decreasing distress and improving quality of life.

As well as the issue of control, both papers identified themes relating to shame and judgment; in the research paper, this related to participants' sense of shame about their condition, for example in terms of their physical appearance or needing help from others or from mobility aids and other adaptations. In the literature review, parents spoke about fearing and feeling judgment from others but at the same time, judged themselves in relation to their competence as parents. These two findings both appear to relate to the experience of stigma,

[2] Not her real name – pseudonyms are used throughout, consistent with the research paper.

which Goffman (1963) defined as “an undesired differentness” (p. 5). This definition would fit with participants in the research paper feeling different from others, and with parents in the literature review feeling different from both their own and others’ expectations of themselves, as well as being conscious of their child’s difference. Goffman’s definition has been expanded to include felt stigma – feelings of shame related to being different, and feeling that discrimination may occur because of this – and enacted stigma – actual experiences of discrimination (Scambler & Hopkins, 1986). More recently, the work of Link and Phelan (2001) identified components of stigma including labelling of difference, stereotyping, separation of ‘them’ and ‘us’, status loss, and discrimination. Link and Phelan also recognised power as an important component in stigma, insofar as it is inherently linked with power differences, with those who are stigmatised inevitably having less power than those who stigmatise. There may be links here with the issues relating to control discussed above; for instance, people may feel out of control in response to their reduced power, and seek to regain or reassert control as a means of increasing their power and thereby reducing stigma.

There are important societal implications in terms of reducing stigma associated with disability; a systematic review by Smythe, Adelson, and Polack (2020) found that education and training were effective in reducing enacted stigma experienced by children with disabilities and their families. This review may be a good starting point in identifying ways to address stigma within society, although the authors did focus on research completed in low- and middle-income countries, so it is unclear how this would apply in the countries of participants in the research paper (USA, Canada, and UK). Older research has identified several strategies for reducing stigma towards children with physical disabilities, including direct contact with the stigmatised group (Brown & Hewstone, 2005), and participation in adapted physical activity (Goodwin, Thurmeier, & Gustafson, 2004). Again, although this work may offer good starting points, it is unclear how they would apply to adults.

## Strengths and limitations

I will now consider some strengths and limitations of the research paper, with reflections on what I may do differently in future. It is important to note, however, that most of these points are not purely strength or limitation, but rather each point has its own costs and benefits.

### Participants

The sample size (six participants) is perhaps on the smaller side, though not an unusually small sample for an IPA study; research with samples of six participants or fewer has been published – for example, Levy and Cartwright (2015) published an IPA study in the target journal with five participants. As Smith et al. (2009) state, "[3-6 participants] should provide sufficient cases for the development of meaningful points of similarity and difference between participants, but not so many that one is in danger of being overwhelmed". IPA focuses more on detailed accounts of individual experiences rather than on generalisability; therefore, smaller sample sizes are not necessarily a problem. Nonetheless, a larger sample may have permitted a greater complexity and richness to the data gathered, though this would have needed careful balancing to avoid the volume of data becoming unwieldy.

### Recruitment

In addition to the sample size, the fact of recruitment taking place online meant I only reached people who have access to the internet. This in itself may have excluded people who would have been interested in participating, and may go some way to accounting for the younger age range of participants, despite CRPS being more common in older adults (de Mos et al., 2007). Furthermore, half of the participants were recruited from their writing online about living with CRPS (a pragmatic decision to aid with recruitment, given the circumstances discussed below relating to COVID-19); these people, then, were already relatively comfortable discussing their diagnosis and experiences relating to it. Those who

volunteered in response to social media adverts were also willing to be interviewed about their CRPS. In contrast, those who saw the adverts and did not respond may have been less open and reflective about their condition and less willing to discuss their experiences – hence not responding. This is a strength as well as a limitation, as it meant that participants were able to be open and honest with me in interviews, allowing depth and richness to the data gathered. However, the experiences of these individuals may differ from those of people who were less willing to participate; all participants in the research paper had, to some degree, reached a point of acceptance with their diagnosis, and were even able to identify some positive consequences. Perhaps people who are still struggling with their diagnosis to a greater degree would be less likely to come forward to discuss it with a stranger.

**Data collection**

Conducting research interviews was a new experience for me, which therefore involved a learning curve. In the initial interviews, I was perhaps not quite as familiar with my interview guide as I could have been. This unfortunately meant I missed some opportunities to explore a participant's response further, as I checked what came next on the schedule. This improved as data collection progressed, as I became more familiar with the interview guide and therefore more able to keep my focus on the participant. My confidence also improved as I completed more interviews, meaning I was more relaxed and more perceptive to the subtleties of participants' responses. In future, 'pilot' interviews may facilitate familiarity and confidence with the interview schedule; an expert by experience may be able to provide valuable feedback here on my delivery of questions and so on.

One point that I did note whilst transcribing the interviews was that quite a lot of time was taken up at the start of the interview in asking participants details such as their age, family circumstances, and a brief history of their diagnosis. This part of the conversation aided in rapport-building and giving the participant time to settle into the interview

somewhat. However, this is information which could potentially have been collected prior to the interview, thereby allowing more time in the interview itself for more in-depth discussion of the impact of CRPS, and more flexibility to follow up interesting points. In future, I might consider using a brief form for participants to complete pre-interview, to gather demographic details, date of diagnosis, and a brief summary of their journey from symptom onset to diagnosis. A similar process was used by Beales et al. (2021) in their qualitative study of people with CRPS in Australia.

The use of online interviews is also an interesting point; I had not planned to collect data in this way, as discussed below, though I had intended to offer it as an option alongside face-to-face interviews. As O'Connor and Madge (2016) set out, there are both strengths and limitations to online data collection. Notably, in terms of accessibility, online interviews meant that people who may not have been able to travel for an in-person interview were able to participate and have their views heard (Bowker & Tuffin, 2004). In addition, video-recording through the online calling platforms (as opposed to audio-recording face-to-face interviews, as initially planned) meant that a greater depth of information was available during transcription, including participants' facial expressions and gestures as well as their actual words.

Despite these benefits, completing the interviews online had its drawbacks; firstly, this approach meant that individuals who didn't have access to suitable technology, or a private space to complete an interview, may not have felt able to participate. Additionally, older people may also have been excluded by the use of online interviews (O'Connor & Madge, 2016). This could perhaps be mitigated to some extent by offering telephone interviews as an alternative option; I did consider this, but given the international sample and limitations in terms of budget, this was not feasible. Secondly, it is perhaps more difficult to build rapport in an online interview than it might have been face-to-face, and so it is possible

that a slightly lower level of detail and disclosure may have been attained than would have been possible with in-person interviews. Overall, though, I felt that online interviews worked well in this context and I would certainly consider using them again in future – I would say I have come to agree with Deakin and Wakefield (2013) that online interviews are a viable method of data collection in and of themselves, not just, "an alternative or secondary choice when face-to-face interviews cannot be achieved" (p.3).

**IPA approach**

As well as the above considerations relating to my specific choices and limitations within this project, the research was also subject to general strengths and limitations of an IPA approach. As with all IPA-based studies, findings are not considered to be generalisable beyond samples with similar characteristics and contexts to those of the participants involved. Again, this is not purely a limitation, since the aim of IPA as an idiographic approach is to explore the particular experiences of a small group of participants, rather than to generate theories relating to whole populations. As Brocki and Wearden (2006) discuss, part of the role of IPA is to explore subjective experiences, and participants' accounts of making sense of these experiences. This approach, Brocki and Wearden argue, can usefully supplement quantitative data, allowing for greater exploration and understanding of the complexity of human experience. Nonetheless, it is important to bear in mind the limited generalisability of the findings.

**Involvement of experts by experience**

As noted in the research paper, I contacted two CRPS charities to seek feedback and suggestions from an expert by experience on my interview guide. Unfortunately, neither charity responded, and due to time constraints I was unable to look elsewhere, so I was not able to gain any feedback from experts by experience – although I did review material written online by people with CRPS when designing the interview guide. Feedback was offered by

two healthcare professionals working with individuals with CRPS (a physiotherapist and a clinical psychologist), which went some way to ensuring relevant areas were addressed. For example, based on the feedback from the physiotherapist, questions about participants' perceptions of their affected limb were included, to explore any potential dysmorphia or perceptual disturbances which were felt to be linked to identity. However, this is obviously not equivalent to having input from individuals with CRPS, who may have been able to highlight further areas for inquiry or suggest better ways of approaching certain topics. Future research, then, would benefit from involvement of experts by experience to ensure that relevant areas are being addressed in research.

On reflection, one way of addressing this significant limitation of the present research may have been to ask one or more of the participants recruited via their online writing to act as "consultants", for example by providing feedback on my research question, participant materials, and interview guide. I had enough interest through social media recruitment to still have sufficient participants even without the online writers taking part in the interviews. The main reason I did not take this approach was probably anticipatory anxiety around recruitment, given the substantial delays my thesis had already encountered, as discussed below. Prior to submitting this work for publication, I plan to share the themes with participants to gain feedback and reflection, which can be used to adjust or rework the themes if necessary.

## Suggestions for future research

In addition to the suggestions made in the main research paper, relating to the age and cultural background of participants, there is scope for future research exploring the efficacy and acceptability of psychological interventions for people with CRPS. To date, the majority of research relating to treatment and management of CRPS seems to investigate medical treatments such as medication and surgery. A recent paper by Murray, Harrison,

Goebel, and Twiddy (2020) did explore the impact of a pain management programme (PMP) for individuals with CRPS; however, this study explored the impact of the PMP on patients' decision-making around medication options. Thus, there is still a need for a broader understanding of how patients experience PMPs, and other psychologically informed interventions, in terms of living with and/or managing their CRPS.

## Reflexivity

As discussed in the main research paper, reflexivity is an important element in IPA research, contributing to the acknowledgement and 'bracketing' of preconceptions, assumptions, and biases which may otherwise impact upon the interview process and/or the analysis (Smith et al., 2009; Yardley, 2000). I will now share some of my reflections from throughout the research process.

In terms of choosing a topic for my thesis project, I knew that I wanted to study something related to physical health, based on my previous assistant psychologist role within a physical health psychology team and my aspiration to work in this area, once qualified. Persistent pain has long been an area of interest for me, and I was intrigued by the idea of exploring people's experiences of living with pain and how this affected them. As I began to explore the literature around this area, CRPS stood out as a condition with very little existing research, which I found appealing given my passion for helping people to have their stories heard. A little more background reading, and discussion with my research supervisors, enabled me to identify identity as a specific area of interest, and the project developed from there.

As discussed above, I had not completed research interviews prior to this study, and therefore had a degree of anxiety about the process. I discussed this with peers who had worked on similar projects, and also recorded some thoughts and reflections in my research journal. In July 2020, I wrote:

> "I'm feeling quite anxious about my first interview this week – research interviews are not something I've done before. I'm trying to compare it mentally to therapy, as I have often noticed feeling anxious before the first session with a new client, too. Although the contexts are different, I'm sure that similar skills will be helpful – containment, empathy, genuine interest in participants' stories."

Reflecting in this way allowed me to consider the specific elements of the process which were causing me anxiety, and think about ways of managing this anxiety to reduce the impact on my data collection. In particular, the parallel with therapy was helpful for me – as a trainee clinical psychologist in my final year of training, I was comparatively comfortable in therapy sessions by this stage. Thus, highlighting to myself the transferable skills provided me with reassurance that I would be able to manage in the interviews.

At this stage, I had been completing remote therapy by video link on placement for a few months, and so was perhaps feeling more confident about the technology than I may otherwise have been. Nonetheless, I was conscious of the differences between therapy and research interviews, and anxious that I would accidentally slip into 'therapy-mode' during an interview. I noticed from early in the process of data collection that there was a temptation during interviews to revert to this 'therapy-mode', perhaps because I am more familiar with the role of therapist than interviewer. Remaining conscious of this temptation helped me to maintain a balance. As I reflected in late July 2020, towards the end of my interviews:

> "It's hard not to slip into therapist role when participants are sharing difficult experiences and their emotional reactions. Trying to balance this by remaining empathic and validating. I've found this particularly hard with Jen and her descriptions of her self-image – a topic that often comes up therapeutically.

> Knowing she has her own therapist definitely helped but even so – not an easy position to hold, for me anyway!"

As well as noticing these processes which occurred prior to and during interviews, I also became conscious of participants seeming extremely grateful for my interest in CRPS – this was a novel situation for most. Several participants mentioned feeling dismissed and invalidated in their interactions with healthcare professionals; thus, my interest in CRPS was a welcome change for them. Unfortunately, these experiences were not unusual, with similar findings being reported by Johnston, Oprescu, and Gray (2015) in their narrative review. I found these experiences sad to hear, as I believe strongly that an important part of healthcare is taking people seriously and listening to their concerns. This also meant that I felt a lot of pressure to 'do them justice' in my write-up and any dissemination of my research; I was anxious to understand their views and experiences 'properly', and make their participation seem worthwhile. As I considered in my research journal following my interview with Jen:

> "It's nice to hear how much participants appreciate my research in CRPS, but at the same time pretty sad when you think it comes from, in some cases, years of feeling unheard and invalidated by health professionals… Feeling quite a lot of pressure to do them all justice and make their contributions worthwhile!"

In particular, I noticed as I began my analysis that it was difficult for me to begin to develop from initial notes to emerging themes. I felt that I wanted to honour my participants' stories, and that by reworking their words in this way I would not be doing this. This conflict is acknowledged by Smith et al. (2009), highlighting that this is part of the process and that the end result will be "a product of both of your [researcher and participant] collaborative efforts" (p. 92). I found this a reassuring perspective, which enabled me to begin to become more interpretative in my analysis.

## Impact of COVID-19

My ethics application was a lengthy process, exacerbated by academic strikes and then university closures for holiday periods. Originally, my research was going to be hosted through an NHS trust, recruiting through their pain clinic. It was immensely frustrating, then, to obtain NHS ethical approval just as the COVID-19 pandemic took hold in the UK. Upon contacting the Research and Development team in the trust, I was informed that all non-COVID research had been suspended indefinitely. This was understandable, given the seriousness of the situation, but disheartening nonetheless. This prompted a re-design of the project and a new ethics application via the University to allow online recruitment.

This adaptation had benefits as well as drawbacks. Most notably, online recruitment meant that I had a much larger number of potential participants. Recruiting online allowed me to reach potential participants internationally, meaning a greater breadth of experiences were covered, with participants having experiences of a variety of healthcare systems. However, this means of recruitment meant that I was unable to verify participants' diagnosis (Hewson, Yule, Laurent, & Vogel, 2003); it is possible that one or more participants did not have a confirmed diagnosis of CRPS. Furthermore, it is unclear whether a sample of participants solely from the UK, as originally planned, would have produced similar findings; there is potential that participants' experiences with a lack of universal healthcare in the US, for example, will have had impacted findings.

Just as COVID had an impact on myself (and this research), participants were also living through unprecedented times. As individuals with pre-existing health conditions, a couple mentioned some anxiety about the current situation, due to feeling they would be at increased risk of complications should they catch the virus. This background anxiety may have coloured participants' responses to my questions, for example making them more attuned to potential 'threats'. On reflection, it may have been interesting to explore the impact of COVID for participants – particularly given early evidence about the extent of 'ableism' in

response to the pandemic and the impact this has had for disabled individuals (Lund, Forber-Pratt, Wilson, & Mona, 2020) – though this was probably beyond the scope of the current study.

## Conclusions

This thesis explores the impact of persistent pain, from the perspective of parents of children with transdiagnostic persistent pain, and fromthat of adults with CRPS. As with all research, there are strengths and limitations of this study; these may inform and direct future research. A reflexive stance has been taken throughout the research, contributing to reliability and validity. The impact of completing research during a global pandemic is also considered.

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# Section Four – Ethics Section

Word Count: 3,522 (excluding appendices)

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**Ethics Form**

Faculty of Health and Medicine Research Ethics Committee (FHMREC)
Lancaster University
Application for Ethical Approval for Research
Guidance on completing this form is also available as a word document

**Title of Project**: The Impact of Complex Regional Pain Syndrome (CRPS) on Personal and Social Identity

**Name of applicant/researcher**: Jess Smith

**ACP ID number (if applicable)*:** N/A **Funding source (if applicable)** N/A

**Grant code (if applicable):** N/A

***If your project has *not* been costed on ACP, you will also need to complete the Governance Checklist [link].**

**Type of study**

☐ Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. **Complete sections one, *two* and four of this form**

☒ Includes *direct* involvement by human subjects. **Complete sections one, *three* and four of this form**

**SECTION ONE**

**1. Appointment/position held by applicant and Division within FHM:**
Trainee Clinical Psychologist; Division of Health Research

**2. Contact information for applicant**:
**E-mail**: j.smith26@lancaster.ac.uk
**Telephone**: 07575 950959
**Address**: c/o Doctorate in Clinical Psychology, Furness College, Lancaster University, LA1 4YG

**Names and appointments of all members of the research team (including degree where applicable):**
Dr Craig Murray, Senior Lecturer, Doctorate in Clinical Psychology, Lancaster University
Dr Fiona Eccles, Lecturer, Doctorate in Clinical Psychology, Lancaster University
Dr Richard Johnson, Consultant Clinical Psychologist, Salford Royal NHS Foundation Trust

**3. If this is a student project, please indicate what type of project** by marking the relevant box/deleting as appropriate:
DClinPsy Thesis ☒

**4. Project supervisor(s), if different from applicant**: Dr Craig Murray; Dr Fiona Eccles; Dr Richard Johnson

**5. Appointment held by supervisor(s) and institution(s) where based (if applicable)**: See above (question 2).

**SECTION THREE**

**Complete this section if your project includes *direct* involvement by human subjects.**

**1. Summary of research protocol in lay terms (indicative maximum length 150 words):**
Complex Regional Pain Syndrome (CRPS) is a pain condition with an estimated incidence of 26.2 cases per 100,000 person-years (de Mos et al., 2007). Symptoms can include pain, swelling, tremors, and changes in hair and nail growth, amongst others. There is no known cure.

Research has found that long-term health conditions have an impact on the individual's personal and social identities – that is, how they see themselves, and how they perceive they are seen by others (e.g. Siegel & Lekas, 2002). This, in turn, can contribute to feelings of distress. CRPS differs from other health conditions in several important ways, and so it is unclear whether and how it may affect patients' identities.

The proposed research project therefore plans to conduct interviews investigating the impact of living with CRPS on people's personal and social identities; it is hoped this will contribute to an understanding of what it is like to live with CRPS, and thereby to improvements to care provided to these individuals.

**2. Anticipated project dates (month and year only**)
Start date: July 2020 End date: July 2021 (data collection proposed to be completed by end August 2020; projected end date allows time for examination, viva, any amendments, and preparation for publication if appropriate)

**Data Collection and Management**
*For additional guidance on data management, please go to* Research Data Management *webpage, or email the RDM support email:* rdm@lancaster.ac.uk

**3. Please describe the sample of participants to be studied** *(including maximum & minimum number, age, gender):*
A minimum of three and maximum of six participants will be recruited. This is based on the Interpretative Phenomenological Analysis (IPA) approach used, an idiographic approach which recommends small, homogenous samples with a focus on individual voices. Studies with these numbers of participants are considered appropriate for a doctoral-level research project (Smith, Flowers & Larkin, 2009). With regards publication, it is not unheard of for single-case studies to be published (e.g. Eatough & Smith, 2006), and studies with small numbers of participants are frequently published (e.g. Wilde & Murray, 2009).

To be eligible to participate, individuals must:
Be aged 18 years or over;
Having had a formal diagnosis (given by a qualified medical practitioner, as opposed to self-diagnosed) of Complex Regional Pain Syndrome (CRPS) for a minimum of 12 months (self-

reported, as it will not be possible to verify this);
Be able to complete an interview in English (as sufficient funding is not available for interpreters);
Be able to give informed consent to participate (individuals with major cognitive impairment (for example severe learning disability) will therefore be excluded).

**4. How will participants be recruited and from where?** *Be as specific as possible. Ensure that you provide the full versions of all recruitment materials you intend to use with this application (e.g., adverts, flyers, posters).*
Two recruitment paths may be used:
1. Potential participants have been identified from an internet search for blogs and blog posts relating to CRPS, and contacted via a scoping email – this has only been possible where contact details have been shared on the blogs identified. It has been highlighted that ethical approval has not yet been granted, and these individuals have been asked to express potential interest only. Three expressions of interest have been received so far. Further information (e.g. Participant Information Sheet) will be shared once ethical approval is granted, to allow them to make an informed decision.

If sufficient participants are not recruited via path one above, the below will be used as a next step in recruitment:
2. Adverts will be posted on social media (including the Recruitment Poster) and shared with relevant CRPS charities/organisations, inviting interested individuals to make contact for further information about the research project. The Participant Information Sheet will then be shared, to enable them to make an informed decision about whether to participate.

Recruitment will be closed at the end of August, unless the minimum number of participants (3) have not been recruited. Any additional individuals making contact after this point will be thanked for their interest and informed that recruitment has now been closed.

If sufficient participants cannot be recruited via these means, blog posts relating to CRPS written by participants recruited in step 1 above may also be included in the analysis, where participants have given consent for this. The use of blog posts alone is not considered sufficient due to their limited tailoring to the research question.

Given the timescale of the project, repeat interviews (i.e. multiple interviews with the same participant) are not considered appropriate; this is because IPA would generally require there to be a significant amount of time between interviews whereby developments or change is likely to occur.

**5. Briefly describe your data collection and analysis methods, and the rationale for their use.**
Data will be collected through an individual interview with each participant. Interviews will be conducted via telephone or the participant's video calling software of choice (e.g. Microsoft Teams, Skype, Zoom, or similar). Given that participants will be recruited online, it is assumed that they will have access to the internet. An interview guide will be used to

guide the questions asked; it is anticipated that interviews will last around 60-90 minutes per participant. Interviews will be video- or audio-recorded for later transcription and analysis. As discussed above, if the minimum number of participants cannot be recruited, blog posts written by participants may also be included in the analysis, where consent is given to do so.

Given the research question's focus on participants' experiences, an Interpretative Phenomenological Analysis (IPA) approach will be taken to data analysis. Interviews will be transcribed by the student researcher, and the transcripts used to create codes or themes of common experiences.

**6. What plan is in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc.)?** *Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.*
For the duration of the research project, data will be stored electronically on the university's secure encrypted server or in university-approved cloud-based storage.

Data (including typed transcripts of interviews) will be retained by the Doctorate in Clinical Psychology programme's research administration team for a period of ten years. Data will be transferred to the administration team using a secure university-approved procedure. Following the retention period, the data will be deleted by the administration team under the supervision of the research supervisors.

**7. Will audio or video recording take place?** ☐ no ☒ audio ☒ video
**a. Please confirm that portable devices (laptop, USB drive etc) will be encrypted where they are used for identifiable data.** *If it is not possible to encrypt your portable devices, please comment on the steps you will take to protect the data.*
Dependent upon the software chosen by the participant, video may be recorded using in-application features (e.g. recording directly within Microsoft Teams). These files will be stored on the university's secure encrypted server or University-approved secure cloud storage.

Should this not be an option (that is, if the interview is completed in a program without this facility), audio will be recorded using a digital audio recorder; this device cannot be encrypted, and data will therefore be transferred, as soon as practicable following completion of each interview, to the university's secure encrypted server or University-approved secure cloud storage. It is anticipated that this should be possible immediately following completion of each interview, however should there be any delay the audio recorder will be kept with the researcher at all times between completion of interview and upload of data.

**b. What arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?**
Video and/or audio recordings of interviews will be stored on the university's secure encrypted server or university-approved secure cloud storage until the research has been examined; at this point, the audio files will be deleted.

Video and/or audio recordings of consent will be stored by the programme research administration team for a period of ten years; these will be stored separately from other data including transcriptions of interviews.

Please answer the following questions *only* if you have not completed a Data Management Plan for an external funder.

**8a. How will you share and preserve the data underpinning your publications for at least 10 years, e.g. PURE?** Data (including transcripts of interviews) will be stored in electronic format by the Doctorate in Clinical Psychology programme's administration team. Data will be transferred electronically using a secure method that is supported by the university. It will then be stored on the university's secure encrypted server, or in university-approved secure cloud storage, as per usual course procedures.

**8b. Are there any restrictions on sharing your data?**

Due to the small sample size, even after full anonymisation there is a risk that participants may be identified from their interviews. Therefore, full transcripts will only be shared on request with genuine researchers.

**9. Consent**

**a. Will you take all necessary steps to obtain the voluntary and informed consent of the prospective participant(s) or, in the case of individual(s) not capable of giving informed consent, the permission of a legally authorised representative in accordance with applicable law?** yes

**b. Detail the procedure you will use for obtaining consent?**

Participants will be provided with the Participant Information Sheet and the Consent Form, a minimum of 48 hours before the interview is scheduled to take place. Participants will be given opportunity to ask any questions before consenting to interview. Once any questions have been answered to the participant's satisfaction, the Consent Form will be read aloud, one statement at a time, and the participant asked to verbally agree to each section. This process will be audio-recorded separately from the rest of the interview. Recordings of consent will be stored separately from all other data including interview recordings and transcripts, on the university's secure encrypted server or in university-approved secure cloud storage. Written signatures will not be requested due to the remote nature of the interview and the difficulties participants may have accessing printers, scanners etc. to facilitate this.

**10. What discomfort (including psychological e.g. distressing or sensitive topics), inconvenience, or danger could be caused by participation in the project?** *Please indicate plans to address these potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.*

No major discomfort or distress is anticipated from participation in this project; however, it is possible that participants will find it upsetting to talk about their health condition and the impact that this has had on their life. As far as possible, this will be managed within the interview by the interviewer, who is a trainee clinical psychologist. Should a participant become distressed during their interview, they will be offered the opportunity to pause or discontinue the interview. A safety plan will be agreed at the beginning of the interview,

which will include someone (e.g. a friend or family member) the researcher can contact should the participant become distressed. If necessary, participants will be directed to appropriate sources of support. All participants will also be provided with a debrief sheet to direct them to appropriate resources and sources of support (including their GP/family doctor, and charities relevant to CRPS) should they experience any distress after the interview has ended.

Participants will be able to withdraw their participation at any time before or during the interview, and for two weeks following completion of the interview. Beyond this point, transcription and analysis will have commenced, and it may not be possible to retrieve data. Participants who withdraw consent during the interview will be asked whether they wish to withdraw data already collected, in which case all recordings will be deleted immediately. If participants are happy for data collected so far to be retained, they will be reminded of their right to withdraw their data within two weeks.

**11. What potential risks may exist for the researcher(s)?** *Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).*
No direct risks to the researcher are anticipated due to the remote nature of the interviews. However, it is possible that the nature of the material discussed during interviews may cause distress; this will be managed via regular supervision with the research supervisors.

**12. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.**
No direct benefits to participants are anticipated. However, it is hoped that participants will find taking part interesting and rewarding. In addition, the research may contribute to improved care for people with CRPS in the future.

**13. Details of any incentives/payments (including out-of-pocket expenses) made to participants:**
Participants will not be paid to take part. Given the remote methods of data collection, it is not anticipated that any expenses will be incurred. It is possible that not all potential participants will have access to video calling equipment (e.g. webcam/microphone), however given the recruitment strategy which focuses on online activity, this is considered unlikely.

**14. Confidentiality and Anonymity**
*a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications?*
yes

*b. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.*
Interviews will be conducted remotely via the most convenient means for the participant, which may include Microsoft Teams, Zoom, Skype, or similar. New accounts will be created

for any platforms used, and these accounts will only be used for the purposes of this research project. Participants will be informed that the internet cannot be guaranteed to be a completely secure means of communication.
Interviews will be anonymised as they are transcribed by the student researcher, with potential identifying information such as names and locations removed.
Anonymised quotations from interviews will be used within the final report; total confidentiality therefore cannot be assured. It is possible, given the relative rarity of CRPS, that participants could be recognised by people who know them from the quotations used. Participants will be made aware of this risk before they consent to participate.
In addition, if blog posts are included in the analysis, it will not be possible to assure anonymity due to the public nature of blogs (it would be possible to identify the blogs through an internet search). Participants will be made aware of this fact before consenting to participate, and will be given the option to consent to interview only and *not* to inclusion of their blog posts in analysis. This will apply only to participants recruited through their published blogs/blog posts relating to CRPS.

**15. If relevant, describe the involvement of your target participant group in the *design and conduct* of your research.**
Feedback on participant materials (including adverts, information sheet, consent form, debrief and interview guide) was provided by clinicians – a clinical psychologist and a physiotherapist – working with individuals with CRPS. CRPS charities were also contacted to seek feedback from individuals with CRPS, but unfortunately none were forthcoming.

**16. What are the plans for dissemination of findings from the research?** If you are a student, include here your thesis.
Data will be seen only by members of the research team including the student and supervisors.

In terms of dissemination, the research project will form part of the applicant's thesis. A presentation on the thesis project will be given to members of the Doctorate in Clinical Psychology department and other interested parties.

In addition, publication will be sought in appropriate academic and/or professional journal(s) so that findings can contribute to improvements in the care of the target population.

**17. What particular ethical considerations, not previously noted on this application, do you think there are in the proposed study?**
Given the remote nature of interviews, consideration has been given to the best approach to take with regards to any issues of risk which may be raised during the interviews, e.g. participants disclosing thoughts of harming themselves or others. This is not considered a likely outcome but is possible if the interview raises distressing feelings for the participant. The interviewer would seek to manage this within the interview e.g. by making a safety plan with the participant at the start of the interview, including whom the researcher might contact in the case of distress.

In terms of the potential to include blog posts and the implications this will have for maintenance of participants' anonymity, participants will be made aware of these implications, and offered the option to consent to interview only, to use of their blog posts only, or to interview *and* use of their blog posts. This will be stated clearly in the audio/video recorded consent. In addition, in the write-up of the research, any blog posts used in the analysis will not be referred to as 'blogs'; instead, these will be referred to as 'written reports of experiences'.

**SECTION FOUR: signature**

**Applicant electronic signature**: Jess Smith Date 13/05/2020

Student applicants: please tick to confirm that your supervisor has reviewed your application, and that they are happy for the application to proceed to ethical review ☒

**Project Supervisor name** (if applicable): Dr Craig Murray Date application discussed 13/05/2020

**Submission Guidance**

1. **Submit your FHMREC application by email to Becky Case (**fhmresearchsupport@lancaster.ac.uk) as two separate documents:

i. **FHMREC application form.**

Before submitting, ensure all guidance comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line.*

ii. **Supporting materials.**

Collate the **following materials for your study, if relevant, into a single word document:**

a. **Your full research proposal (background, literature review, methodology/methods, ethical considerations).**
b. Advertising materials (posters, e-mails)
c. Letters/emails of invitation to participate
d. Participant information sheets
e. Consent forms
f. Questionnaires, surveys, demographic sheets
g. Interview schedules, interview question guides, focus group scripts
h. Debriefing sheets, resource lists

**Please note that you DO NOT need to submit pre-existing measures or handbooks which support your work, but which cannot be amended following ethical review. These should simply be referred to in your application form.**

2. Submission deadlines:

i.Projects including direct involvement of human subjects **[section 3 of the form was completed]**. The *electronic* version of your application should be submitted to Becky Case **by the committee deadline date.** Committee meeting dates and application submission dates are listed on t/he FHMREC website. Prior to the FHMREC meeting you may be

contacted by the lead reviewer for further clarification of your application. Please ensure you are available to attend the committee meeting (either in person or via telephone) on the day that your application is considered, if required to do so.

ii. The following projects will normally be dealt with via chair's action, and may be submitted at any time. **[Section 3 of the form has *not* been completed, and is not required].** Those involving:

a. existing documents/data only;
b. the evaluation of an existing project with no direct contact with human participants;
c. service evaluations.

3. **<u>You must submit this application from your Lancaster University email address, and copy your supervisor into the email in which you submit this application</u>**

# Appendices

## Appendix 4-A: Research Protocol

*The Impact of Chronic Complex Regional Pain Syndrome (CRPS) on Personal and Social Identity*

**Applicant**: Jess Smith

**Supervisors**: Dr Craig Murray (Senior Lecturer, Doctorate in Clinical Psychology, Lancaster University);
Dr Fiona Eccles (Lecturer, Doctorate in Clinical Psychology, Lancaster University);
Dr Richard Johnson (Consultant Clinical Psychologist, Salford Royal NHS Foundation Trust)

### Introduction

#### What is CRPS?

Complex Regional Pain Syndrome (CRPS) is a pain condition with an estimated incidence of 26.2 cases per 100,000 person-years (de Mos et al., 2007); in approximately 15-20% of cases, the condition lasts beyond twelve months, at which point it is considered chronic (Royal College of Physicians (RCP), 2018). Women are 3-4 times more likely to be affected than men (Castillo-Guzmán et al., 2015). The condition is present across the lifespan (Borchers & Gershwin, 2014), though average age of onset has been reported as 43 (Sandroni et al., 2003); onset generally follows a physical trauma to the affected limb, for example a sprain, fracture or surgery (Birklein et al., 2015).

Symptoms of CRPS include sensory (such as pain, hypersensitivity or burning sensation as well as reduced sensitivity to heat or cold), motor (for example tremors and reduced range of motion), trophic (changes in growth rates of hair and nails, atrophy of the skin), and autonomic (including changes in sweating, skin colour or skin temperature, as well as swelling) symptoms (Birklein et al., 2015; Drummond, 2010; RCP, 2018). Symptoms generally affect one limb, though have sometimes been reported to spread to other body parts (Drummond, 2010).

CRPS is sub-classified based on the presence of nerve damage; CRPS Type I is diagnosed in the absence of nerve damage, whilst CRPS Type II requires evidence of nerve damage. A third category, CRPS Not Otherwise Specified (CRPS-NOS) reflects patients previously diagnosed with CRPS but no longer meeting full diagnostic criteria (e.g. some symptoms have resolved but pain continues), as well as those who do not fully meet diagnostic criteria but whose symptoms cannot be better explained by another diagnosis (RCP, 2018).

There is currently no known cure for CRPS, and the National Institute for Health and Care Excellence (NICE) do not make any recommendations for its treatment, citing insufficient evidence (O'Connell et al., 2013). The National Health Service (NHS) recommends a combination of physical rehabilitation, pain relief, psychological support, and education and self-management to manage symptoms of CRPS (NHS, n.d.). The RCP also includes psychological support amongst its recommendations for long-term management of

CRPS (RCP, 2018). This interdisciplinary approach is supported in the literature, e.g. Sahli et al. (2013).

**How do chronic health/pain conditions impact identity?**

The term 'personal identity' refers to an individual's sense of self and of one's past, present and future (Ellis-Hill & Horn, 2000); 'social identity', meanwhile, is an individual's understanding of themselves as a member of social category(ies) or group(s) (Stets & Burke, 2000). Chronic illness has been found to impact upon identity, with the diagnosis of a chronic condition tending to cause alteration in both personal and social identity (Siegel & Lekas, 2002). Bury (1982) discussed the concept of chronic illness as biographical disruption, whilst Charmaz (1983) described loss of self as "a fundamental form of suffering in the chronically ill". Smith and Osborn (2007) describe chronic pain as an "assault on the self", discussing the way their participants' identities were deteriorated by their chronic pain.

Chronic illness can impact upon identity constructs through its disruption of relationships and activities considered important by the individual – for example, Adams et al. (1997) discuss the social stigma around a diagnosis of asthma and the lengths to which some of their participants went to avoid seeing themselves as asthmatic, including avoiding activities which they previously enjoyed; Wisdom et al. (2008) considered how internalisation of negative social responses can alter personal identity. Charmaz (1983) wrote about the loss of former self-image, without the opportunity to develop a valued new one – often accumulating over time as a chronic condition goes on, leading to increasing impact on personal identity. Alongside this, reduced ability to work, pursue hobbies, and maintain relationships can also impact on the social identity (Charmaz, 1983). Indeed, Charmaz states that people exist as social beings and so any impact on one's social identity is bound to impact one's personal identity also.

More specifically relating to chronic pain, Crowe et al. (2010) identified that chronic lower back pain affected participants' sense of self, discussed within their theme 'The alteration to sense of self'. Within this theme Crowe et al. spoke about how the pain had affected not only participants' lifestyles (activities, career etc.) but also their self-image. They discussed a sense of tension around how participants viewed themselves, in contrast with how they wished to be. Similarly, Toye et al. (2013) discuss the struggle to hold on to the 'real me' faced by patients with chronic pain, and how this impacts on relationships with the self and with others.

**How does CRPS differ from other chronic conditions including chronic pain?**

CRPS is relatively unique amongst chronic health and pain conditions; unlike conditions such as AIDS or cancer, it is not life-threatening, and it is much less well-known than many other chronic conditions such as diabetes or arthritis. Unlike the majority of chronic pain conditions, visible changes are present alongside the pain in CRPS, for example swelling and changes in skin colouring – visible difference has been reported to contribute to impact upon identity (Jacoby et al., 2005). Furthermore, the pain is accompanied by additional symptoms in CRPS including motor symptoms such as paralysis, involuntary movements, or neglect (van Hilten, 2010) – yet there is no clear organic cause as in other conditions with similar symptoms, such as stroke.

**Why is research required?**

Research into the experiences and beliefs of patients with CRPS (Louw et al., 2018) found that overall, patients have a poor understanding of CRPS, how it is treated, and what the future may hold for them. There was widespread confusion amongst patients, many of whom had received conflicting information regarding their condition. For example, around two-thirds of patients reported having a dual diagnosis of CRPS and Reflex Sympathetic Dystrophy (RSD; a former name for CRPS Type I), despite it being more than twenty years since the condition was re-classified (Stanton-Hicks et al., 1995). It is unclear what consequence this poor understanding of CRPS has in terms of its impact on identity for individuals with CRPS.

The role of identity has been highlighted in recovery in a variety of conditions including mental illnesses (Wisdom et al., 2008), diabetes (Luyckx et al., 2008) and CFS (Larun & Malterud, 2007). For example, Wisdom et al. (2008) found that people with diagnoses of severe mental illness experienced a 'loss of self', and difficulty distinguishing between 'old' and 'new' selves. A review by Yu et al. (2015) found that negative self-evaluation impacts on daily functioning in people suffering from chronic pain, whereas a sense of self aside from the chronic pain is associated with better daily functioning.

An understanding of how CRPS impacts on identity may therefore inform how these individuals are supported (e.g. Asbring, 2001). As Crowe et al. (2010) discussed, exploring what a condition means to a patient may contribute to improvements in quality of life, as well as contributing to a therapeutic relationship by allowing validation of experiences. Furthermore, Yu et al. (2015) discussed the role that the self plays in therapeutic approaches including mindfulness, self-compassion, and psychological flexibility; an understanding of how CRPS impacts on the self may therefore be central to effective use of these approaches.

In particular, coming to terms with a new identity may be an important part of managing CRPS. For example, Luyckx et al. (2008) found that development of a strong sense of identity contributed to coping with a chronic condition (in this case, diabetes) and more favourable outcomes, whereas failure to develop a strong sense of identity was linked with unhelpful illness-related coping strategies and poorer outcomes. Luyckx et al. therefore argued that interventions should include assisting individuals to integrate the illness into their self-definition.

### Current Study

The aim of this research project is to explore patients' experiences of CRPS (Type I or Type II) and the impact the condition has had upon their personal and social identity. There is a paucity of research on the lived experience of patients with CRPS (Butler, 2015), and the impact this may have on their personal and social identity. The proposed study therefore aims to contribute to understanding of this, with a view to informing the support clinical psychologists working with these individuals are able to offer.

The main research question is:

What is the impact of chronic Complex Regional Pain Syndrome (CRPS) on patients' personal and social identity?

## Method

### Participants

Participants will be adults aged 18 years or older, who self-report having had a diagnosis of CRPS (Type I or Type II) for at least twelve months. Participants will be

recruited via two main pathways:

1. Authors of blogs relating to CRPS will be contacted (via email or on-site contact forms) to introduce the research and invite them to participate;

If sufficient participants are not recruited via the above pathway, then the below approach will be taken:

2. A recruitment poster will be shared on Twitter, inviting interested individuals with CRPS to make contact via email.

Once potential participants have expressed interest, they will be sent the Participant Information Sheet and Consent Form via email. They will be given time to read these and encouraged to ask any questions, before deciding whether to take part.

Given the Interpretative Phenomenological Analysis (IPA) methodology, a minimum of three and maximum of six participants will be recruited; as discussed in Smith, Larkin & Flowers (2009), small homogenous samples are considered optimum for IPA. Blog posts written by participants recruited via step one above may also be included in analysis, should it not be possible to recruit sufficient numbers of participants via the above methods; consent will be sought for this at the time of interview.

**Design**

The study will use a qualitative methodology informed by an IPA approach (e.g. Smith, Flowers & Larkin, 2009). An individual semi-structured interview will be conducted with each participant. This will be audio-recorded and then transcribed by the student researcher, prior to analysis.

**Materials**

An interview schedule will be used to guide interviews with participants; further details are given below.

**Procedure**

When participants contact the student researcher to express interest (either via direct response to an invitation email, or by making contact via email in response to an advertisement shared on Twitter), they will be sent the Participant Information Sheet and Consent Form via email. Participants will be encouraged to ask any questions before deciding whether to take part.

Interviews will not be scheduled until at least 48 hours after the Participant Information Sheet has been received by each participant, to allow sufficient time for them to be read. If they are happy to participate, a convenient time and means of interview will be agreed.

Interviews will take place remotely via video conferencing software such as Microsoft Teams, Zoom, or Skype – depending upon each participant's preference. Verbal consent will be gained by reading out each item from the consent form for the participant to verbally agree. The participant will then be asked to give overall verbal consent to take part. This process will be audio-recorded separately to the rest of the interview.

Interviews will be video and/or audio-recorded to allow for later transcription and analysis. Should a participant not wish for their interview to be recorded, it will not be possible for them to take part as it would not be possible to analyse their data in line with IPA processes. Interviews will follow a broad schedule of open questions exploring participants' experiences of living with CRPS and how this has impacted upon their identity.

A debrief sheet will be emailed to each participant at the end of their interview. This will give details of potential sources of support in the event of any distress following the interview.

Participants will be asked if they would like to receive a copy of the finished research project, and details taken (email address) to allow the student researcher to send this if desired. These details will be stored separately to all other information, to protect participants' confidentiality. Data storage is discussed in more detail in the 'Ethical Concerns' section below.

## Proposed Analysis

The interviews will be video or audio recorded and transcribed to allow for analysis. Analysis will follow an IPA methodology as described by Smith et al. (2009). IPA seeks to understand participants' experiences of the phenomenon under investigation, both individually as well as in terms of patterns of convergence and divergence.

If blog posts are to be included alongside interview transcriptions, additional consent from the authors will be sought for this. Guidance for this process will be taken from Thomas, Allison and Latour (2017), who used blogs as a means of exploring the lived experience of life after stroke.

IPA has been selected as an appropriate approach to analysis given the phenomenological nature of the research question; that is, it focuses on participants' subjective experiences of CRPS and how this has affected them personally. IPA seeks to understand participants' experiences in their own terms, rather than trying to fit them into a pre-determined model or theory (Smith & Osborn, 2015).

Once the interviews have been transcribed, the student researcher will commence analysis following the steps set out by Smith et al. (2009). The researcher will begin by reading and re-reading each transcript to familiarise themselves with the data. Initial notes and observations will be made through this process. This leads into the next step, initial noting, wherein points of interest within the data are highlighted, and detailed notes and comments are made. These comments may be descriptive in nature, or they may concern the participants' use of language, building into more conceptual comments. Next, emergent themes will be developed, before the next case is analysed. Finally, the researcher will look for themes across the participants and make interpretations from these.

## Practical Issues

It is possible that there may be some difficulty in recruiting, given the relative rarity of CRPS which means the potential pool of participants is quite small. However, given the relatively small number of participants required for this type of research, it is anticipated that the recruitment methods described above should allow recruitment of sufficient numbers of participants to suit the IPA method. As discussed above, blog posts may also be included in the analysis should recruitment prove more difficult than anticipated.

## Ethical Concerns

### Confidentiality

All data will be stored electronically on the university's secure encrypted server or in university-approved secure cloud storage. All identifying or personal data (e.g. names and email addresses) will be stored separately from interview transcripts in password-protected files. Audio-recorded verbal consent will be stored separately from all interview data.

Confidentiality will be discussed with each participant prior to their interview. This will include explaining the circumstances in which confidentiality may be broken – if the researcher has concerns that the participant, or someone else, may be at risk.

Given that participants will possibly be completing the interviews from home, they will be encouraged to find a quiet, private space where they are not likely to be interrupted and where they feel comfortable to discuss personal matters such as their health.

**Anonymity**

Complete confidentiality cannot be assured given that quotations from interviews will be used in the write-up of the research project; however, such quotations will be anonymised as far as possible. Interviews will be anonymised at the point of transcription, with potential identifiers removed. No individually identifying data will be used in the write-up of the study. However, given the relative rarity of the condition, there is a small chance that participants may be recognised from their quotations by people who know them. Participants will be made aware of this risk before deciding whether they wish to take part.

If participants' blog posts are included in analysis, they may also be identifiable from these – particularly given the public availability of these blogs. Participants will be made aware of this risk prior to agreeing to take part in the research; they will be given the option to consent to interview *only* if they are not willing for their blog posts to be included, or to inclusion of blog posts *only* if they do not wish to be interviewed.

**Distress Management**

It is possible that talking about their condition and its impact may be distressing for some participants. If this arises, it will be addressed by the student researcher (Jess Smith, trainee clinical psychologist) during the interview; the participant will be offered the choice to pause or discontinue the interview and the researcher will address any issues raised. The researcher will offer to call someone for the participant, if appropriate; this person may be a friend or family member, for example, and will be agreed between the participant and researcher at the beginning of the interview. In addition, all participants will be provided with resources in their information sheet and debrief, in case of any distress arising after the interview has finished.

**Data Storage**

All interviews will be video- or audio-recorded to allow for later transcription and analysis. Once each interview is completed, audio recordings will be transferred from the portable audio recorder to Lancaster University's secure network drive or secure cloud storage as soon as possible, since the portable device cannot be encrypted. The file will then be deleted from the audio recorder. Video recordings will be uploaded directly to the secure network drive or secure cloud storage. Given the remote nature of the interviews, it is anticipated that this will be done immediately following completion of the interview, however for any time period between interviewing and uploading the recording, the portable device will be stored securely and kept with the student researcher. All other portable devices used for data storage (e.g. laptops, memory sticks) will be encrypted.

All electronic documents will be password-protected and stored securely on Lancaster University's encrypted network drive or university-approved secure cloud storage.

Once the research is completed, video and audio recordings of interviews will be deleted, and the anonymised transcriptions of interviews will be transferred electronically to

the Doctorate in Clinical Psychology Research Co-Ordinator using a secure method supported by the University. These transcripts will be stored for 10 years before being deleted; the Research Co-Ordinator will be instructed with a date of when to delete the transcripts.

Files containing participants' personal or identifying information will be kept in a password-protected file, separate from the anonymised transcripts. An ID number will be used to match participants' identifying information to their transcripts. Video or audio recordings of consent will be held by the Doctorate in Clinical Psychology programme for ten years following completion of the research; these will be transferred electronically using a secure method, and stored separately to other data. All other personal information will be deleted within six months of the research being completed.

**Safety of Researchers**

No direct risk of harm to the researcher is anticipated, given the remote nature of interviews. However, the nature of topics discussed within the interviews may lead to distress for the interviewer; this will be managed within supervision with the research supervisors.

**Informed Consent**

Potential participants will be given information about the nature and purpose of the research. They will be given opportunity to ask any questions, before deciding whether to take part. A minimum of 48 hours will be allowed to consider the information before each participant decides. It will also be made clear (on the information sheet and verbally) that participants can withdraw their data up to two weeks following their interview.

**Timescale**

| | |
|---|---|
| May 2020 | Preparation of Ethics materials; submission to University |
| March-May 2020 | Write-up of Introduction and Methods sections |
| July 2020 | Recruitment, data collection and transcription |
| July-September 2020 | Data analysis (alongside collection) |
| September 2020 | Write-up of Results and Discussion sections |
| September 2020 | Draft submission |
| October 2020 | Amendments based on draft |
| October 2020 | Final submission |

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## Appendix 4-B: Ethics Committee Approval Letter

Lancaster University

Applicant: Jess Smith
Supervisor: Craig Murray and Fiona Eccles
Department: DHR DClinPhys
FHMREC Reference: FHMREC19106

25 June 2020

**Re: FHMREC19106**
**The Impact of Complex Regional Pain Syndrome (CRPS) on Personal and Social Identity**

Dear Jess Smith,

Thank you for submitting your research ethics application for the above project for review by the Faculty of Health and Medicine Research Ethics Committee (FHMREC). The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for the amendment to this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

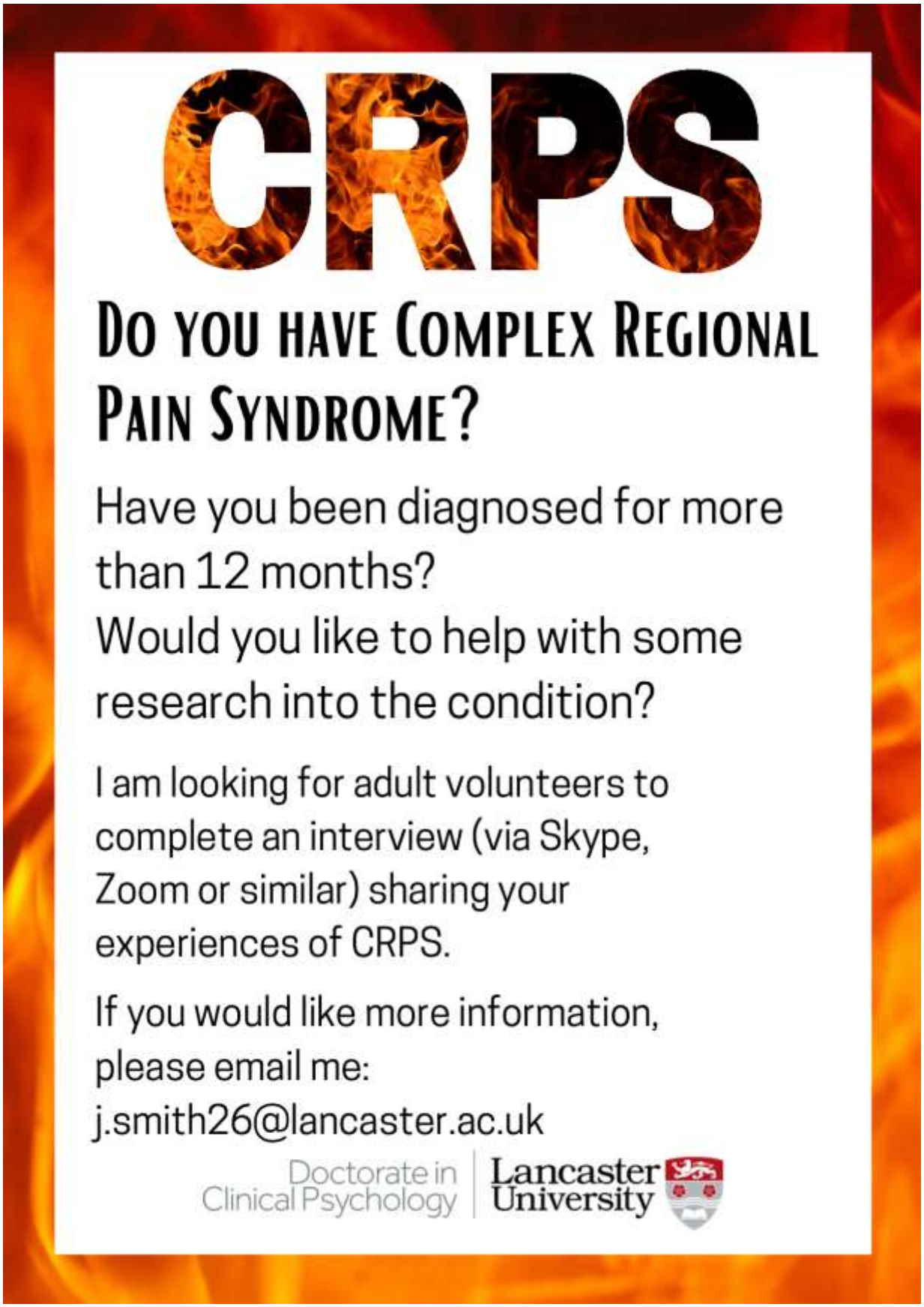
Email:- fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

E. Suri-Payer

Dr. Elisabeth Suri-Payer,
Interim Research Ethics Officer, Secretary to FHMREC.

**Appendix 4-C: Recruitment poster**



## Appendix 4-D: Recruitment adverts

**Recruitment Advertisements – Version 0.1, 13/05/2020**

***Email to be sent to blog authors***

Dear [name],

Thank you for your response to my previous email, and for expressing interest in taking part in my research project relating to Complex Regional Pain Syndrome. I am now able to provide further details about this project.

I am looking for people with CRPS to complete an interview about how CRPS affects them. The interview would be completed via Microsoft Team, Zoom, Skype or similar, and would last up to 90 minutes. The aim of the research is to improve understanding of what it is like to live with CRPS, in hopes that this can improve the support that individuals with this condition receive.

If you are still interested in taking part, please find attached a Participant Information Sheet and a Consent Form which I ask you to read through in your own time. Once you have read these, I would be grateful if you could let me know whether you are happy to take part. We can then arrange a convenient time for an interview via your preferred video calling software. You do not need to complete the consent form (this is provided for your information only), as we will go through this at the beginning of the interview.

If you have any questions or would like any further information to help you decide, please do let me know.

Many thanks for your time, and I hope to hear from you soon.

Best wishes,

Jess Smith

Trainee Clinical Psychologist

Lancaster University

***Tweet from research account***

Do you have Complex Regional Pain Syndrome (CRPS)? I'm looking for people to take part in some research! Please get in touch for more details if interested. [Attach advertising poster as an image]

***Tweet to relevant Twitter accounts***

Hi! I'm carrying out some research into CRPS and wondered if you'd mind sharing my ad? Please let me know if you'd like any further details. Thank you so much! [Attach advertising poster as an image]

@CRPSUK

@BNightsCRPS

@RSDSA

@TeamCRPS1

**Appendix 4-E: Participant Information Sheet**

**Participant Information Sheet – Version 0.3, 17/06/2020**

***The Impact of Chronic Complex Regional Pain Syndrome (CRPS) on Identity***

For further information about how Lancaster University processes personal data for research purposes and your data rights, please visit our webpage: https://www.lancaster.ac.uk/research/data-protection

My name is Jess Smith and I am conducting this research as a student in the Doctorate in Clinical Psychology programme at Lancaster University, Lancaster, United Kingdom.

**What is the study about?**
This project aims to improve understanding of how chronic Complex Regional Pain Syndrome (CRPS) impacts on people's identity.

**Why have I been approached?**
You have been approached because the study requires information from people who have been diagnosed with CRPS for at least a year.

**Do I have to take part?**
No, you do not have to take part. You are welcome to ask questions before you decide whether you would like to participate. My contact details are below.

In addition, if you decide to take part, you can change your mind and withdraw your information up to two weeks after your interview has been completed.

**What will I be asked to do if I take part?**
If you decide you would like to take part, you will be invited to complete an interview with myself. This will last up to an hour and a half. I will ask you some questions about living with CRPS and how it has affected you. I will video and/or audio record the interview, and then type it up into a written transcript.

The interview will be conducted via video calling software such as Microsoft Teams, Skype, Zoom, or similar. Please be aware that the internet cannot be guaranteed to be a secure means of communication.

If I have contacted you via your blog about CRPS, I may also ask if you are happy for me to include some of your blog posts in my analysis. You should be aware that this would mean you may be identifiable, as your blog posts are publicly available. If you would prefer to participate in an interview *only*, that is fine. You may also choose for your blogs posts only to be included, without completing an interview.

**Will my data be identifiable?**
The information you provide will be anonymised as far as possible. I will use quotations from interviews when writing up my research. Any names and other identifying information will be removed. CRPS is a relatively rare condition, so there is a chance that you could still be identifiable by people who know you from quotations used. In addition, if you allow me to include your blog posts in my analysis, you may be identifiable from this.

The data collected for this study will be stored securely and only the researchers conducting this study will have access to this data:

- Video/audio recordings of interviews will be deleted once the project has been examined.
- The files on the computer will be encrypted (that is, no-one other than the researchers will be able to access them) and the computer itself password protected.
- The transcription of your interview will be made anonymous by removing any identifying information including your name. Anonymised direct quotations from your interview may be used in the reports or publications from the study, so your name will not be attached to them.
- All your personal data will be confidential and will be kept separately from your interview responses.
- Transcriptions of the interviews will be held by the Doctorate in Clinical Psychology programme for ten years before being deleted. All personally identifiable data will be held separately, and will be deleted within six months of the research being completed.
- Video/audio recordings of consent will be held by the Doctorate in Clinical Psychology programme for ten years. They will be held separately from your other data.

**What will happen to the results?**
The results will be reported in a thesis, and may be submitted for publication in an academic or professional journal. This means that the results can help professionals working with people with CRPS and can guide future research.
I will ask if you would like to be emailed a copy of my completed research project.

**Are there any risks?**
There are no risks anticipated with participating in this study. However, if you experience any distress following participation you are encouraged to inform the researcher and utilise the resources provided at the end of this sheet.

**Are there any benefits to taking part?**
There are no direct benefits in taking part. However, I hope you would find it interesting to take part, and that the research will contribute to improvements in care for people with CRPS in the future.

**Who has reviewed the project?**
This study has been reviewed and approved by Lancaster University Faculty of Health & Medicine's Research Ethics Committee.

**Resources in the event of distress**
At the beginning of the interview, the researcher will agree a safety plan with you in case of any distress. This will include asking if there is someone such as a friend or family member we can call, should you become distressed during the interview.

Should you feel distressed either as a result of taking part, or in the future, the following charities offer support and resources around living with CRPS.
CRPS-UK: https://crps-uk.org
Burning Nights CRPS: http://burningnightscrps.org/
RSDSA: https://rsds.org/
You may also contact your GP or family practitioner.

In an emergency you should contact your local emergency services, or attend your nearest Accident & Emergency Department.

**Where can I obtain further information about the study if I need it?**
If you have any questions about the study, please contact the main researcher:
Jess Smith – j.smith26@lancaster.ac.uk
You can also contact one of my supervisors:
Dr Fiona Eccles – f.eccles@lancaster.ac.uk, +44 (0)1524 592807
Dr Craig Murray – c.murray@lancaster.ac.uk, +44 (0)1524 592730

**Complaints**
If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher or their supervisors, you can contact:

Dr Ian C. Smith, Senior Clinical Tutor & Research Director
**Tel**: +44 (0)1524 592282
**Email**: i.smith@lancaster.ac.uk
Doctorate in Clinical Psychology
Furness College
Lancaster University
Lancaster, LA1 4YG

If you wish to speak to someone outside of the Clinical Psychology Doctorate Programme, you may also contact:

Dr Laura Machin
**Tel**: +44 (0)1524 5934973
**Email**: l.machin@lancaster.ac.uk
Faculty of Health and Medicine
(Lancaster Medical School)
Lancaster University
Lancaster, LA1 4YG

Thank you for taking the time to read this information sheet.

**Appendix 4-F: Consent Form**

**Participant Consent Form – Version 0.3, 17/06/2020**

***The Impact of Chronic Complex Regional Pain Syndrome (CRPS) on Identity***

I am asking if you would like to take part in a research project investigating the impact of Complex Regional Pain Syndrome (CRPS).

Before you consent to participating in the study, please read the **Participant Information Sheet**. You will be asked to verbally agree to each of the statements below, before commencing the interview. This will be audio- or video-recorded; this recording will be stored separately from the rest of your interview.

If you have any questions or queries, please speak to the student researcher, Jess Smith.

1. I confirm that I have read the information sheet and fully understand what is expected of me within this study.
2. I confirm that I have had the opportunity to ask any questions, and they have been answered satisfactorily.
3. I understand that my interview will be video and/or audio recorded and then made into an anonymised written transcript.
4. I understand that video and/or audio recordings will be kept until the research project has been examined.
5. I understand that my participation is voluntary and that I am free to withdraw my data without giving any reason, up to two weeks after completing my interview. I understand that withdrawing will not affect my medical care or legal rights.
6. I understand that once my data have been anonymised and incorporated into themes it might not be possible to withdraw.
7. I understand that the information from my interview will be pooled with other participants' responses, anonymised, and may be published.
8. I consent to information and quotations from my interview being used in reports. I understand that this may mean some people could identify me from the quotations used.
9. I understand that the researcher will discuss data with their supervisors as needed.
10. I understand that any information I give will remain anonymous unless there is a risk of harm to myself or others, in which case the principal investigator may need to share this information.
11. I understand that the internet cannot be guaranteed to be a secure means of communication, and therefore my interview cannot be guaranteed to be secure.
12. [I understand that if I consent to posts from my blog to be included in analysis, this may mean that I can be identified, as my blog posts are publicly available.]*

13. I understand that Lancaster University will keep written transcriptions of the interview for 10 years after the study has finished.
14. I consent to take part in an interview.
15. [I consent for posts from my blog to be included in analysis.]*
16. [I consent to both an interview and for posts from my blog to be used in analysis.]*

*Points 12, 15 and 16 apply **only** to participants recruited via their online blogs relating to CRPS. Blog posts will only be included in analysis if we are not able to recruit enough participants for interview.

## Appendix 4-G: Interview Schedule

Interview Schedule – Version 0.1, 13/05/2020

***The Impact of Chronic Complex Regional Pain Syndrome (CRPS) on Identity***

### Introduction

Formal introduction of myself and the project.

Answer any questions the participant may have.

**Privacy & Anonymity**

Before we begin the main interview, I need to make sure that you understand the private nature of the interview. The things you talk about will be private – I may use quotations when I write up my research, but these will be anonymised and any identifying information will be removed.

There are some exceptions to this privacy. If I am worried about you, or someone else, I may need to share my worries in order to keep everyone safe. I will always try to speak to you about this first. I may then speak to one of my supervisors about my concerns. If I think there is an urgent risk, I may contact the emergency services.

I hope it is unlikely that you will become distressed during the interview, but I'd like to have a safety plan just in case. Is it okay to work that out together now? [Anything you'd like me to do if you become upset, anyone I should call? Does anyone know you are doing this interview? Is there someone at home with you? Etc.]

[START RECORDING OF CONSENT PROCESS]

**Consent**

Before we start, I want to make sure that you understand what is going to happen and why, and that you are happy to participate.

Check that participant has received and read the Information Sheet. Answer any questions arising from this.

[Read out each statement from the consent form and ask the participant to verbally agree to each, and to give overall verbal consent to participate.]

[STOP RECORDING AND START NEW RECORDING FOR INTERVIEW]

### Interview

**Background**

So, as you are aware, my research is looking at what it's like to have Complex Regional Pain Syndrome (CRPS) and how it affects people. We'll start by getting a little bit of background about you and your condition.

- Age now, if you don't mind
- How would you describe your ethnicity?
- Gender identity
- Current situation – work (type of role, full or part-time?), partner, children/grandchildren etc.
- Official diagnosis (CRPS? RSD? Type I/II?) and (roughly) when did you receive this?
- Journey to diagnosis – e.g. when/ how did symptoms start? Who did you see (healthcare professionals?) Did you have any tests, scans, treatments? Did you have any other diagnoses first? How long did it take to get the CRPS diagnosis? What

impact did this journey have on you, if any? How did you find this journey – what went well? What could have been better and how?

- How was the diagnosis given? (In person? By who? Much information/ support/ resources given?)
- If you don't mind answering - do you receive any benefits or welfare support? Have you in the past? How do you feel about this? (*Thinking about the impact of receiving benefits on sense of self, especially given stigmatisation of benefits recipients in society.*)
- What were things like before the CRPS? Previous health and activity levels, roles (e.g. jobs, roles in the home, hobbies and interests, leisure activities)
- How did these change once your symptoms developed – what impact did the symptoms have? What was your experience of this process (i.e. sudden / gradual changes)

**Understanding of CRPS**

- Can you tell me what you understand about CRPS? What are your symptoms? Which bother you the most? Do they affect how you feel about yourself? How? How do you manage your symptoms? Have these strategies arisen from professional advice or things that you have developed yourself?
- How does the condition affect you day-to-day? Are there good days and bad days? What do these look like (i.e. compare good vs bad for mood / activity). Are there any other factors that influence good / bad days? – I.e. fatigue / insomnia / other psychosocial stressors.
- What is your prognosis? How do you expect the condition to change/ develop/ improve? What have you been told about this?
- How have you learnt about CRPS? Information from healthcare professionals/ own research etc.? How do you feel about this? How has this information influenced / changed your understanding and the way you live with CRPS?
- Is there anything that you still have questions about or don't understand about the condition?
- How do you feel you have responded to/coped with your diagnosis? What do you think this says about you?
- How have healthcare professionals responded to your diagnosis? Have you felt supported/ understood? How has this affected you?

**Impact of/ response to CRPS**

- How would you have described yourself as a person prior to having CRPS? (Mindset, qualities, interests, activity levels.)
- How has this changed since you have developed or been diagnosed with CRPS? (Mindset, qualities, interests, activity levels.) What has changed since you developed CRPS? Are there things you have had to give up or do differently? (e.g. work, tasks around the home, hobbies and interests)
- How have these changes impacted on your view of yourself, if at all?

- How does this make you feel? When you feel like this, what runs through your mind/ why do you think you feel like this? (Questions around impact of CRPS on mood)
- Do you think you have changed as a result of CRPS? How? Do you view this as positive or negative? Why? In what ways have you *not* changed? What has stayed the same? Are there times when CRPS challenges this? How do you manage this?
- How do you feel about your affected limb(s)? Does CRPS affect how you look at/touch/think about the affected limb(s)? In what way? How does this impact on you?
- Do you worry about contact with others or objects e.g. people, clothing or bedclothes touching the affected area? How does this affect you? E.g. avoiding activities, social interactions.
- How do you see yourself in the future? Do you have future plans? Have these changed due to CRPS or other factors?
- How has CRPS affected your relationships? (Partner, family, friends, children?). Do you feel other people understand your condition and the impact it has? How / How not? What would be helpful for them to understand better? What are the barriers to their understanding? Has it affected how you are in relationships?
- Do you think CRPS has affected the way other people see you or respond to you? Examples of change? What do you make of this? How does this make you feel? How does this affect you? How do you manage these changes? i.e. communicate / ignore / feel upset etc.
- Has anything positive arisen out of CRPS – e.g. changes in mind-set / attitudes / approach to life etc.

[Thank participant for their time. Enquire as to whether they would like a copy of the completed research paper. Confirm email address if yes.]

**Appendix 4-H: Participant Debrief**

**Participant Debrief – Version 0.2, 17/06/2020**

***The Impact of Chronic Complex Regional Pain Syndrome (CRPS) on Identity***

Thank you for taking part in this research project, which I am completing as part of my Doctorate in Clinical Psychology. I am very grateful for your time. I hope that you have found participation interesting and rewarding.

**What was the purpose of the research?**

Complex Regional Pain Syndrome (CRPS) is a complex condition, and there has been limited research into how it affects people. My research aims to improve this situation, looking specifically at how CRPS affects people's identities.

I hope that this research will help healthcare professionals to support people with CRPS more effectively in the future.

**What happens next?**

I will transcribe your interview in an anonymised format, removing identifying information such as your name. I will pool this information with other participants' interviews and look for themes in how CRPS affects people's identities.

I will write the research up and submit it to the University for marking. I may also submit the research for publication in relevant academic or professional journals, so that it can help healthcare staff working with people with CRPS.

Please let me know if you would like to receive a copy of the finished research project via email. I will take your contact details so I can send this to you. These details will be stored separately to your other information including your interview recording and transcript.

**What if I change my mind about taking part?**

If you decide that you no longer want your information to be used in my research, please let me know as soon as possible, and within two weeks of your interview. After this point, your information will have been anonymised and pooled with other participants, so it may no longer be possible to identify your data to withdraw it.

**What if I feel distressed after taking part?**

There is a chance that talking about your condition may cause feelings of distress in the days to come. If this happens, please use the below resources to seek support:

- CRPS UK: https://crps-uk.org
- Burning Nights CRPS: http://burningnightscrps.org/
- RSDSA: https://rsds.org

- Your GP or family doctor
- In case of an emergency, your local Accident and Emergency department

**What if I have concerns about the research?**

In the first instance, please contact me to discuss your concerns:
**Email**: j.smith26@lancaster.ac.uk
**Mail**: Jess Smith, Doctorate in Clinical Psychology, Furness College, Lancaster University, Lancaster, LA1 4YG

Alternatively, you can contact the Doctorate in Clinical Psychology's Research Director, Dr Ian Smith:
**Email**: i.smith@lancaster.ac.uk
**Phone**: +44 (0)1524 592282
**Mail**: Doctorate in Clinical Psychology, Furness College, Lancaster University, Lancaster, LA1 4YG

If you wish to speak to someone outside of the Doctorate in Clinical Psychology programme, you can contact the Chair of the Faculty of Health & Medicine's Research Ethics Committee (FHMREC), Dr Laura Machin:
**Email**: l.machin@lancaster.ac.uk
**Phone**: +44 (0)1524 594973
**Mail**: Faculty of Health and Medicine (Lancaster Medical School), Lancaster University, Lancaster, LA1 4YG