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Health-related guilt in chronic primary pain: A systematic review of evidence

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Purpose. Chronic primary pain conditions are characterized by significant functional disability, emotional distress, and diagnostic uncertainty. Health-related guilt associated with coping and living with chronic pain is poorly understood. There had been no attempts to synthesize findings on health-related guilt across studies. Therefore, the aim of this study was to conduct a systemic review of evidence, to enable an understanding of the role of health-related guilt in chronic primary pain, and to provide directions for future research.

Method. A search strategy was developed based on our eligibility criteria. Four databases (PsycINFO, Scopus, PubMed, and Web of Science) were searched for relevant papers from inception to 8 July 2020. Data from 12 qualitative and six quantitative studies were synthesized narratively.

Results. The review of qualitative studies resulted in three themes, relating to the management of pain, diagnostic uncertainty/legitimizing pain, and how participants' actions or inactions affect others. These findings were integrated with evidence from quantitative studies, which showed that higher levels of guilt were associated with more pain and pain interference, functional impairment, and poorer psychological and social functioning.

Conclusions. The findings demonstrate that health-related guilt is an important psychological factor associated with more pain and poorer function in people with chronic primary pain conditions. Future research should examine health-related guilt as a potential mediating/moderating factor leading to more distress and suffering in this population and as a potential target for interventions.

Statement of contribution

What is already known on this subject?

- Health-related guilt associated with chronic pain is poorly understood.
- There have been no systematic reviews of this topic.

What does this study add?

- Health-related guilt is associated with more pain and poorer function in chronic primary pain conditions.

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- It should be examined as a potential mechanism leading to more distress and suffering in this population.
- It should be also examined as a potential target for interventions.

Background

Pain is an unpleasant sensory and emotional experience associated with actual or potential tissue damage, and it is influenced by biological, psychological, and social factors (IASP). Chronic pain remains a highly prevalent health problem; it affects an estimated 20% of people globally (Macfarlane, 2016) and is associated with disability, low mood (Pincus & McCracken, 2013), reduced social roles (Froud et al., 2014), and high clinical and societal costs (Maniadakis & Gray, 2000; Phillips, 2006). To date, interventions that have targeted patients' well-being have shown moderate success (Williams, Fisher, Hearn, & Eccleston, 2020), but there is a need to understand the psychological mechanisms that contribute to patients' distress and suffering. This review focuses on a phenomenon that has been neglected in both pain research and practice – health-related guilt.

Chronic primary pain is defined as pain lasting more than 3 months that cannot be accounted for by another condition, and is characterized by substantial functional disability and emotional distress (Nicholas et al., 2019). Chronic primary pain conditions, such as low back pain, have been identified as a leading cause of disability globally (Lim et al., 2012). Many primary chronic pain conditions are also characterized by unclear aetiology and diagnostic uncertainty, where patients invest themselves in a search of diagnosis in order to justify and legitimize their suffering. There is a growing body of evidence describing increases in anger and frustration, especially towards medical professionals, who are perceived as unaccepting or failing to acknowledge suffering, or even disbelieving the pain experience (Newton, Southall, Raphael, Ashford, & LeMarchand, 2013). Individuals with invisible chronic health conditions often face stigmatization by others (Joachim & Acorn, 2000), and stigmatizing responses by health professionals and public are common occurrence in chronic pain (DeRuddere & Kenneth, 2016). Such experiences are likely to result in guilt, which is defined as emotional distress that stems from the perception that we have done something wrong or that others may believe this (Baumeister, Stillwell, & Heatherton, 1994). In addition, the loss of independence, work, and social activities experienced by people living with chronic pain can have a major impact on patients' relationships (Froud et al., 2014) and may result in conflicting emotions including resentment, a sense of isolation, and guilt.

There is a body of theoretical and empirical evidence that describes guilt in general, but not in reference to living with pain. Guilt belongs to a family of self-conscious emotions alongside shame, pride, and embarrassment. Guilt combines both affective and cognitive aspects (Kubany & Watson, 2003), including negative self-regard and unpleasant or even painful feelings associated with the belief that we might have done something wrong or hurt somebody (Tilghman-Osborne, Cole, & Felton, 2010). A review of 23 theory-based definitions of guilt and 25 measures of guilt suggests a lack of conceptual clarity in clinical research, where many guilt measures and definitions reflect other constructs (Tilghman-Osborne et al., 2010). The most common related constructs are shame, worry/anxiety, sadness/depression, embarrassment, self-punishment, and fear. The overlap between guilt and shame is, in particular, common; they are often treated as identical concepts, and they have been used interchangeably, even in scientific literature. One of the most prominent and evidence-based distinctions between the two concepts proposes that shame implicates a negative evaluation of the 'global self', while guilt implicates a negative

evaluation of a specific behaviour (Tangney, Stuewig, & Mashek, 2007). Furthermore, shame is associated with anger and defensive responses such as blaming others, while guilt is associated with self-blame, remorse, regret, and rumination over one's actions, which can often prompt reparative behaviours such as apologizing (Tangney & Dearing, 2003).

To date, there have been no attempts to integrate evidence on health-related guilt in general, or in pain specifically. The aim of the review was to provide an understanding of the underlying evidence base for the role of guilt in coping and living with primary chronic pain, by synthesizing all quantitative and qualitative research that included an examination of guilt.

Method

A study protocol and search strategy were developed based on guidance from the Centre for Review and Dissemination (CRD, 2009) and PRISMA statement (reported in Supplemental Data; Moher, Liberati, Tetzlaff, & Altman, 2009). The review was registered with the PROSPERO (Booth et al., 2011), registration number: CRD42015029180.

Study selection criteria

We included studies if they were empirical, reported primary data, and were published in peer-reviewed journals in the English language. Where data for the same sample were reported in more than one study, the study with the largest sample was included. We included both quantitative and qualitative studies.

The population of interest was adults, aged ≥18 years with one or more primary chronic pain conditions. Chronic pain in childhood is associated with different challenges, and its management requires substantial involvement of parents and teachers. Chronic primary pain is defined as persistent 'pain for more than 3 months and is associated with significant emotional distress and/or functional disability, and the pain is not better accounted for by another condition' (Nicholas et al., 2019, p. 28). Studies that focused on secondary chronic pain (e.g., cancer pain, rheumatoid arthritis) were excluded. Any studies with mixed chronic pain diagnoses were required to explicitly mention primary chronic pain conditions as part of the group.

The primary concept of interest was guilt, related to living and coping with chronic pain. Based on the theories and definitions of guilt outlined in the introduction and to differentiate it from other concepts such as shame (Tangney et al., 2007; Tilghman-Osborne et al., 2010), we operationalized guilt in the following way: (1) guilt involves either real or imagined transgressions, which are believed to have contributed to some negative outcomes; (2) guilt should be studied in context, related to a reasonably specific event or behaviour and makes people contemplate their actions or inactions; and (3) guilt is different from constructs such as shame, and because they are often used interchangeably (Tangney et al., 2007), studies examining such concepts were included in the search/extraction and the decision on inclusion in the synthesis was made by examining the full manuscript. Finally, because guilt is a common term in suicidality, self-harm, and criminal behaviour literatures, our initial search returned a great number of such studies. We therefore refined our final search (CRD, 2009) by excluding those terms.

Factors studied in association with guilt were any physical and mental health variables, including quality of life, and psychological and social functioning variables. If a study

additionally examined guilt in relation to some other variables such as demographic variables, these were also extracted and reported in the synthesis.

For qualitative studies, the inclusion criteria were guilt associated with living and coping with chronic primary pain conditions. Due to the ambiguity of overlapping concepts such as shame, worry, and frustration (Tilghman-Osborne et al., 2010), studies were only included if they explicitly mentioned guilt, as evident in participants' quotes. Studies were excluded if they failed to cite a verbatim quote mentioning guilt, even when authors interpreted guilt as a theme or category in their analysis. Studies that solely examined health care professionals' or significant others' perspectives of guilt were excluded.

For quantitative studies, the inclusion criteria were a study that had to both measure and statistically analyse guilt by measuring the association between guilt and another factor; and studies that merely measured guilt frequency and compared it with a control group were excluded. Additionally, if a study included a measure of a different concept (e.g., depression) containing a guilt item, which was not analysed separately, the study was not included. These latter exclusions were necessary to avoid ambiguity in the interpretation of findings.

Information sources and search strategy

We developed a search strategy that was in line with our operationalization of guilt and eligibility criteria. The full search strategy for one database can be found in the Supplemental Data. This search strategy was adapted for all four databases (PsycINFO, Scopus, PubMed, and Web of Science), which were searched from their beginnings to 31 August 2016 and updated on 8 July 2020. To identify additional relevant papers, manual citation searches were also conducted. It was not possible to include all individual primary chronic pain diagnoses in our original search strategy, and to ensure they were all covered, we additionally searched two databases (one medical – PubMed; and one generic – Web of Science) for each 'primary chronic pain condition' (Nicholas et al., 2019) and 'guilt'.

Screening of publications and study selection

Two Web-based software programs for managing data in systematic reviews were used: EPPI-Reviewer 4.0 (Thomas, Brunton, & Graziosi, 2010) was used for deduplication and screening of publications during the initial database search, while Rayyan (Ouzzani, Hammady, Fedorowicz, & Elmagarmid, 2016) was used when the search was updated in July 2020. Publications were independently screened by title/abstract by two reviewers (DS and TP) and a trained research assistant. At the second stage, full-text papers were screened independently by DS and TP, and any disagreements were resolved by consulting a third reviewer (ME).

Quality assessment of individual studies

Qualitative and quantitative studies were assessed using the Mixed Methods Appraisal Tool (MMAT) – version 2011 (Pluye et al., 2011). Table 3 shows the respective assessment questions. An overall quality score was calculated for each study by dividing the number of criteria met by the total number of items, with scores potentially varying from 0% (no criterion met) to 100% (all criteria met; Pluye et al., 2011). Calculating overall scores and excluding studies with low methodological quality has been criticized (Hong et al., 2018);

therefore, we used them only for guidance purposes and no studies were excluded based on their methodological quality. The MMAT demonstrated high reliability and validity in other studies (Pace et al., 2012). Three reviewers (KP, DS, and ME) and a trained research assistant assessed all studies for quality. Tasks were shared so that each study was assessed independently by two reviewers and any disagreements were decided by a third reviewer.

Data extraction

Three reviewers (DS, ME, and TP) were involved in each stage of data extraction and synthesis by sharing studies and tasks, where evidence from each study was independently extracted/coded by two reviewers and any disagreements were resolved by a third reviewer.

The following information was extracted from publications: authors, year of publication, country where the study was conducted, sample size, participants' sex, age and health condition, study setting, design, analysis, measured constructs, and findings (effect sizes were extracted from quantitative studies and participants' quotes from qualitative studies).

Data synthesis

The syntheses of qualitative and quantitative evidence were conducted separately, and they were equally weighted in terms of their importance. Evidence from qualitative studies was reported first, followed by evidence from quantitative studies, which was then integrated with the findings from qualitative studies where appropriate. Integration of evidence was achieved by looking for overlaps or associations between qualitative and quantitative evidence and reporting them.

We used a thematic synthesis approach to synthesizing evidence from qualitative studies (Thomas & Harden, 2008). The synthesis is a three-stage process: (1) extraction/coding: extracting guilt-related participants' quotes and coding them: (2) grouping: organizing related codes into 'descriptive' themes; and (3) abstraction: synthesizing descriptive themes into abstract 'analytical' themes. Firstly, participants' guilt-related quotes were extracted from each study and then coded according to their meaning. The codes were translated from one study to another, and new ones were developed when necessary. The next step was to compare and contrast the codes in order to start grouping them into descriptive themes, which still closely reflected the findings from the included studies. The final step was to synthesize these themes into more abstract analytical themes.

Next, we synthesized evidence from quantitative studies that met the inclusion criteria. Meta-analysis was not suitable, due to expected heterogeneity of data and methodologies. Therefore, we aimed to synthesize evidence narratively, following the guidance by the Centre for Review and Dissemination (CRD, 2009). This recommends a general narrative synthesis framework consisting of: (1) developing a theory; (2) developing a preliminary synthesis of findings; (3) exploring relationships within and between studies; and (4) assessing the robustness of the synthesis. These elements do not need to be conducted in a consecutive order and can overlap. An agreement was reached that results should be grouped and synthesized thematically according to the factors that were studied in association with guilt, for example, affect, cognitions, functional impairment.

Results

Database searching and coding

The PRISMA flow chart of the full study selection process is shown in Figure 1. Database searching resulted in 20,256 records. After removing duplicates, 13,706 records remained. An additional 12 articles were identified by manual citation and database searching. Screening of titles and abstracts resulted in 13,555 records being excluded. The remaining 163 articles were read full text, 16 were included in the synthesis, and 147 were excluded. A total of 16 articles (containing 18 studies) were included in the synthesis, of which five articles (containing six studies) were quantitative and 11 articles (containing 12 studies) were qualitative.

Study characteristics

Study and participant characteristics are described in Tables 1 and 2, respectively. In summary, the 12 qualitative studies from 11 articles included in the synthesis were published between 1998 and 2018. Eight were conducted in the United Kingdom, two in Australia, one in the United States, and one in New Zealand. Five studies recruited
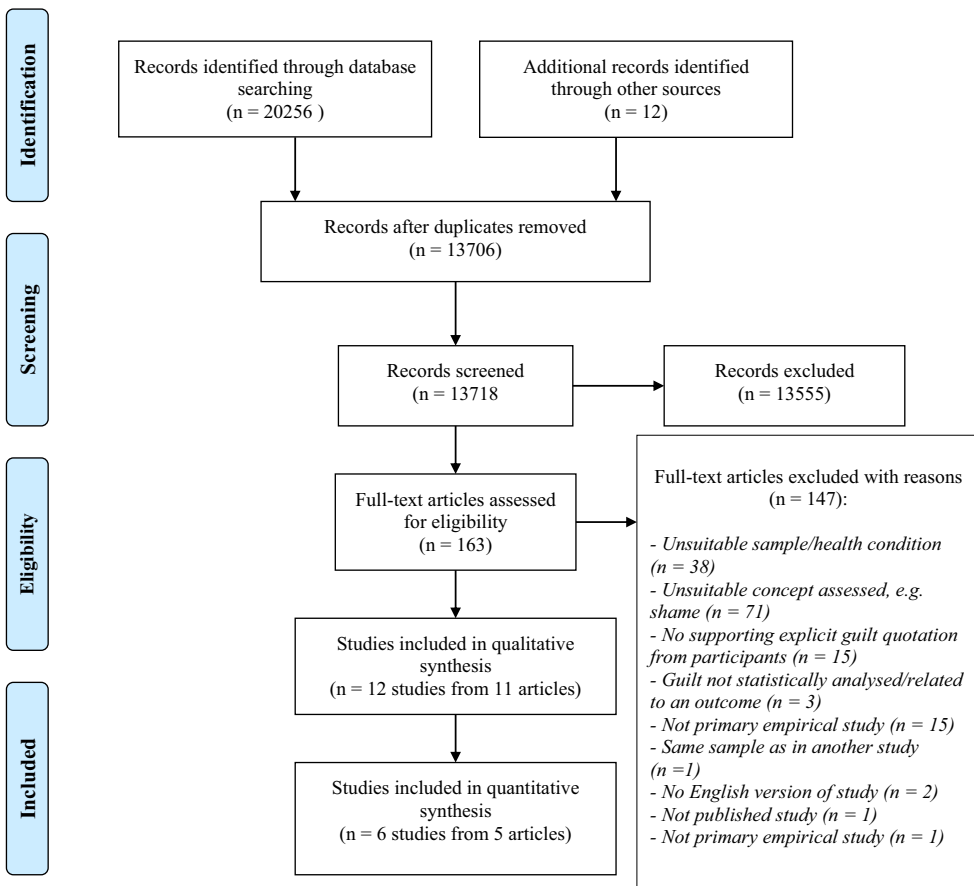
Figure 1. Flow chart of study selection process. [Colour figure can be viewed at wileyonlinelibrary.com]

Table 1. Information about qualitative studies

Author/ year/country	N/sex (F)/Age	Health issue (HI)/setting	Design	Analysis
Bee et al. (2016) United Kingdom	N: 44[a] F: 75% Age: M = 58, SD = 13	HI: chronic widespread pain Setting: general practices	Semi-structured interviews	Framework analysis
Darlow et al. (2013) New Zealand	Chronic pain sample.[b] N: 11 F: 63% Age: 25–65 (M = 45.6, SD = 14.1)	HI: acute and chronic LBP[b] Setting: participants responded to advert placed in health care facilities and public spaces	Semi-structured interviews	Interpretative description framework
Holloway et al. (2007) United Kingdom	N: 18 F: 67% Age: 28–62 (Median: 53)	HI: chronic back pain Setting: pain clinics	Narrative interviews (participants invited to tell their story)	Interpretative phenomenological approach
Mason et al. (2004) United Kingdom	*Study 1:* N: 32 F: 69% Age: 28-69 (M = 49.4) *Study 2:* N: 177 F: 79.1% Age: 12–83 (M = 44.3, SD = 11.2)	*Study 1:* HI: chronic pain[c] Setting: pain clinic; local general practices *Study 2:* HI: chronic pain[c] Setting: online Web survey	*Study 1:* focus groups *Study 2:* online survey	*Study 1:* thematic analysis *Study 2:* content analysis; frequency (%) of guilt being elicited
Osborn and Smith (1998) United Kingdom	N: 9 F: 100% Age: 25–55	HI: chronic LBP Setting: outpatient back pain clinic	Semi-structured interviews	Interpretative phenomenological approach
Rhodes et al. (1999) United States	N: 54 F: 63% Age: 26–65 (M = 47)	HI: chronic back pain Setting: enrolees of a health care plan who were eligible	Semi-structured interviews	Thematic analysis
Ryan et al. (2014)	N: 5	HI: chronic LBP		

Table 1. (*Continued*)

Author/ year/country	N/sex (F)/Age	Health issue (HI)/setting	Design	Analysis
United Kingdom	F: 100% Age: working-age participants	Setting: UK University	Semi-structured interviews	Interpretative phenomenological approach
Serbic and Pincus (2013) United Kingdom	N: 20 F: 70% Age: M = 46.6	HI: chronic LBP Setting: Pain clinic & osteopathic clinic	Semi-structured interviews	Grounded theory
Slade et al. (2009) Australia	N: 18 F: 67% Age: 26–64 (M = 51.2)	HI: non-specific chronic LBP Setting: specialist care (exercise programme)	Focus groups	Grounded theory
Wainwright et al. (2015) United Kingdom	N: 30, of which 13 doctors[d] F: not reported Age: all > 18	HI: chronic pain[c] Setting: general practices; pain management services; UK pain management websites	Semi-structured interviews	Grounded theory
White and Seibold (2008) Australia	N: 5 F: 100% Age: 32–44	HI: chronic back pain Setting: community setting	Open-ended interviews/conversations	Thematic analysis

Note. N, sample size; F, female; LBP, lower back pain; M, mean; SD, standard deviation.
[a]Nested within a randomized controlled trial; [b]Study included both participants with acute and chronic pain; only data collected from participants who had chronic pain were used in the synthesis; [c]Included primary chronic pain conditions; [d]Only patients' data (quotations) were used in the synthesis.

Table 2. Information about quantitative studies

Author/year/country	N/Sex (F)/Age	Health issue (HI)/setting	Design	Analysis[a]	Measured constructs[a]
Conant (1998) United States	N: 103 F: 34% Age: M = 42, SD = 11.7	HI: chronic pain at least 1 year post-injury Setting: outpatient clinic; organizations that serve chronic pain individuals	Cross-sectional; questionnaire study	Correlations; SEM	Guilt; anger; anger suppression; pain severity and interference; punishing responses from significant others; internal health locus of control
Serbic and Pincus (2017) United Kingdom	N: 287 F: 62.2% Age: M = 49.9, SD = 15.4	HI: chronic LBP Setting: pain clinics; physiotherapy department; osteopathy clinic	Cross-sectional; questionnaire study	Regression analysis; mediation analyses	Pain-related guilt; disability; pain; acceptance
Serbic et al. (2016) United Kingdom	N: 413 F: 68.8% Age: M = 49, SD = 14.7	HI: chronic LBP Setting: pain clinics; physiotherapy department; osteopathy clinic; members of three online self-help groups for back pain	Cross-sectional; questionnaire study	SEM	Pain-related guilt; disability; pain; depression; anxiety; diagnostic uncertainty
Turner-Cobb et al. (2015) United Kingdom	*Patients:* N: 64 F: 60.9% Age: 18–73 (M = 46) *Controls:*[b] N: 63 F: 58.7% Age: 24–75 (M = 43)	*Patients:* HI: chronic back pain and sciatica Setting: pain clinic *Controls:*[b] Setting: hospital and university staff	Cross-sectional; questionnaire study	*t*-tests; regression analysis	Guilt; pain; disability; fear of negative evaluation; mental defeat

Continued

Table 2. (*Continued*)

Author/year/country	N/Sex (F)/Age	Health issue (HI)/setting	Design	Analysis[a]	Measured constructs[a]
Ziadni et al. (2018) United States	*Study phase 1:* N: 729 F: 74.1% Age: *M* = 50.8, SD = 14.4 *Study phase 2:* N: 1,452 F: 74.9% Age: *M* = 49.3, SD = 14	*Study phase 1:* HI: chronic pain (LBP, fibromyalgia, migraine) Setting: online sample *Study phase 2:* HI: chronic pain (fibromyalgia, LBP, neck pain, headache, migraine) Setting: online sample and pain clinic	*Study phase 1:* cross-sectional questionnaire study *Study phase 2:* cross-sectional questionnaire study	*Study phase 1:* Spearman's rhos *Study phase 2:* Mann–Whitney U- tests	*Study phase 1:* Worry/guilt; pain, and psychological and physical functioning; pain catastrophizing *Study phase 2:* Worry/guilt; gender

Note. N, sample size; F, female; *M*, mean; SD, standard deviation; SEM, structural equation modelling; LBP, low back pain.
[a]Only analyses and constructs used in this study synthesis are reported; [b]Control participants' data were not used in the synthesis.

Table 3. Quality assessment ratings for qualitative, quantitative and mixed-methods studies

Study	Quality assessment questions								% overall rating
Qualitative	1.1. Are the sources of qualitative data (archives, documents, informants, observations) relevant to address the research question (objective)?	1.2. Is the process for analysing qualitative data relevant to address the research question (objective)?	1.3. Is appropriate consideration given to how findings relate to the context, for example, the setting, in which the data were collected?	1.4. Is appropriate consideration given to how findings relate to researchers' influence, for example, through their interactions with participants?					
Bee et al. (2016)	✓	✓	✓						75
Darlow et al. (2013)	✓	✓	✓	✓					100
Holloway et al. (2007)	✓	✓	✓	✓					100
Osborn and Smith (1998)	✓	✓		✓					75
Rhodes et al. (1999)	✓	✓							50
Ryan et al. (2014)	✓	✓	✓	✓					100
Serbic and Pincus (2013)	✓	✓	✓	✓					100
Slade et al. (2009)	✓	✓	✓	✓					100
Wainwright et al. (2015)	✓	✓	✓	✓					100
White and Seibold (2008)	✓	✓							50
Quantitative non-randomized					3.1. Are participants (organizations) recruited in a way that minimizes selection bias?	3.2. Are measurements appropriate (clear origin, or validity known, or standard instrument; and absence of contamination between groups when appropriate) regarding the exposure/intervention and outcomes?	3.3. In the groups being compared (exposed vs. non-exposed; with intervention vs. without; cases vs. controls), are the participants comparable, or do researchers take into account (control for) the difference between these groups?	3.4. Are there complete outcome data (80% or above), and, when applicable, an acceptable response rate (60% or above), or an acceptable follow-up rate for cohort studies (depending on the duration of follow-up)?	
Turner-Cobb et al. (2015)					✓	✓	✓	✓	100

Table 3. (*Continued*)

Quantitative descriptive	4.1. Is the sampling strategy relevant to address the quantitative research question (quantitative aspect of the mixed-methods question)?	4.2. Is the sample representative of the population under study?	4.3. Are measurements appropriate (clear origin, or validity known, or standard instrument)?	4.4. Is there an acceptable response rate (60% or above)?
Conant (1998)	✓	✓	✓	✓	100
Serbic et al. (2016)[a]	✓	✓	✓		75
Serbic and Pincus (2017)[a]	✓	✓	✓		75
Ziadni et al. (2018)	✓	✓	✓		75

Mixed methods	5.1. Is the mixed-methods research design relevant to address the qualitative and quantitative research(or objectives), or the qualitative and quantitative aspects of the mixed-methods question (or objective)?	5.2. Is the integration of qualitative and quantitative data (or results) relevant to address the research question (objective)?	5.3. Is appropriate consideration given to the limitations associated with this integration, for example, the divergence of qualitative and quantitative data (or results)?	
Mason et al. (2004)[b]	✓	✓	✓	100

Note. A tick (✓) indicates that the criterion was was met, and a cross ( ) indicates that the criterion was either not met or it was unclear whether the criterion was met. [a]In these studies, one of the criteria (MMAT 4.4.) was not applicable; however, to be in line with how other studies were rated, we rated them as not meeting the criterion; [b]This study employed content analysis; however, because the findings were summarized and analysed quantitatively (%), we described and assessed it as a mixed-methods study.

participants from either inpatient or outpatient clinics, five studies recruited participants either in community or via patients' networks, one study recruited participants from both clinic and community, and one study recruited participants from specialist care (an exercise programme). Three studies used interpretative phenomenological analysis, three studies used grounded theory, three studies used thematic analysis, one study used framework analysis, one study used interpretative description framework, and one study used content analysis. Seven studies used semi-structured interviews as a data collection method, two studies used open-ended narrative interviews, two studies used focused groups, and one study conducted content analysis of online survey responses.

The six quantitative studies from five articles included in the synthesis were published between 1998 and 2018. Three studies were conducted in the United Kingdom and three in the United States. Three studies recruited participants from either inpatient or outpatient clinics, two studies additionally recruited participants from both pain clinics and online pain self-help groups, and one study recruited participants online. All six studies were cross-sectional questionnaire studies and employed either independent *t*-test, Mann–Whitney *U*-test, correlational, regression, mediation, structural equation modelling (SEM), or a combination of these analyses.

Participant characteristics

The 12 qualitative studies included 410 participants. Eight studies included participants with chronic low back pain diagnoses, three included participants with more than one chronic pain diagnoses, and one study included participants with chronic widespread pain diagnoses.

The five quantitative studies included 2,316 participants with one or more chronic pain condition. Two studies included participants with a chronic low back pain diagnosis, one study included participants with chronic low back pain/sciatica diagnoses, one included participants with more than one chronic pain diagnoses, and one study included participants with chronic pain with at least 1 year post-injury with no clear pathological cause.

Quality assessment

Table 3 shows quality assessment information for all studies. Six qualitative studies met 100% of criteria, two studies met 75% of criteria, and two studies met 50% of criteria. Two quantitative studies met 100% of criteria, and three studies met 75% of criteria. One mixed-methods study met 100% of criteria.

Synthesis of evidence

Synthesis of qualitative evidence

Analytical themes, descriptive themes, and supporting quotes are presented in Table 4. Three analytical themes resulted from seven descriptive themes, relating to guilt about: (1) how participants manage their condition; (2) legitimacy of their condition; and (3) how their actions/inactions affect others.

Guilt related to managing condition consists of three descriptive themes, where participants expressed: (1) feeling guilty about non-adhering and not being able to follow treatment (Darlow et al., 2013; Slade, Molloy, & Keating, 2009); (2) the presence or

Table 4. Synthesis of qualitative evidence

Analytical themes	Descriptive themes	Supporting quotes
Guilt about not managing condition better	Guilt related to non-adherence/not being able to follow treatment	. . . not doing the exercises that I'm supposed to do. . . and guilt rides on my shoulder like a gremlin. . . (Darlow et al., 2013) . . . the people who were running the program would get angry with me when I couldn't do the exercise correctly and so it didn't actually help I felt guilty for a long time. (Slade et al., 2009)
	The presence/absence of practitioner-related guilt	You feel like you're letting the doctor down because you're not in control. . . You sense the doctor is frustrated because he's got a patient that really hurts and there's times when I've had to come in on an emergency basis so it screws up their scheduling. . . There's a whole lot of dynamics. . . As the patient, you're the focal point and you feel guilty internally and externally. (Rhodes et al., 1999) . . .(the personal trainer) he didn't make me feel guilty for being in the situation that I was in he was encouraging, every little step was an achievement, but he wasn't a dictator. (Slade et al., 2009)
	Treatment (Cognitive Behavioural Therapy) helps to relieve guilt	If I say to my husband, 'I'm not going out, I really don't want to', I don't feel guilty now. Before I would never admit that, so from that perspective, things have changed, because I will say what I think and I will say no, I'm not doing it or I can't do it. So the way I cope with my pain has changed. (Bee et al., 2016)
Guilt about condition not being legitimate	Feeling like a criminal/fraud	When I had a broken arm, it was wonderful, they all rushed towards me to help me in the supermarket and I didn't feel a bit guilty because it was in a plaster and in a sling and it was OK and I didn't mind. But now you do, to all intents and purposes, look perfectly alright and you do feel a bit of a fool and some of the other people just look at you and you just feel guilty about it all. Do they think I'm a fraud perhaps? I don't know, if something showed it might be easier but it is depressing. . . you're afraid of what people think. (Holloway et al., 2007) . . .didn't like my GPs before – I just didn't like the face that they pulled, like, 'Oh, again'. They would do the note but I would leave their practice with a very guilty feeling and I was feeling like a criminal sometimes . . . [the pain] was real . . . I looked healthy, but it was true. . . (Wainwright et al., 2015)
	Guilt about not having concrete evidence/diagnosis	I've beaten myself up on a regular basis, why I can't. . .why it's not better, why am I still getting episodes of pain, why hasn't it gone. . .I feel guilty that I can't tell anybody something concrete, that I cannot give a specific reason. I would have loved the doctor just to have gone 'that's what's wrong with you', and be happy, because then I've got something more concrete to say to everybody'. (Serbic & Pincus, 2013) I do feel guilty, I know that my back really does hurt and I'm not making it up and I feel sort of angry that I can't do it and I think well I wish I could just prove to them that my back really is bad. (Osborn & Smith, 1998) [The pain] made me feel kind of guilty, like I shouldn't be feeling this. You know, there's not real proof with back pain, anybody can say my back hurts. (Rhodes et al., 1999)

Continued

Table 4. (*Continued*)

Analytical themes	Descriptive themes	Supporting quotes
Guilt about how actions/inactions affect others	Guilt about not being able to work	*The guilt is big when it comes to money, and not being able to work, that's really bad, that's horrendous.* (Serbic & Pincus, 2013) *...the pressure is always on to return, because while you are not there others will be doing your job, which is a major guilt factor.* (Ryan et al., 2014) *...and the pressure on my colleagues and you know you can see the stress in their faces. They have recruited a temp...that's relieved my guilt actually.* (Serbic & Pincus, 2013)
	Guilt about not being able to carry out family/social roles	*Guilt, always saying sorry to everybody as if it's my fault'; "You feel like you're letting people down, like when you should be able...to be a good friend or be a good employee, then you feel guilty 'cause you can't and it sucks.* (Serbic & Pincus, 2013) *I think your partners suffer, which makes you feel very guilty, and because you feel guilty, you then get depressed and it's a vicious circle, it really, really is.* (Mason et al., 2004) *...that makes me feel guilty as well because they [friends] are making allowances.* (Serbic & Pincus, 2013) *...as your children grow up with you in pain, you are likely to feel guilty because they have to face issues and shoulder burdens that other kids don't. . . .* (White & Seibold, 2008) *She [mother-in-law] wanted her house decorating the other week. I said I can't even do my own and I feel guilty.* (Osborn & Smith, 1998)

absence of practitioner-related guilt (Rhodes, McPhillips-Tangum, Markham, & Klenk, 1999; Slade et al., 2009); and (3) treatment, such as cognitive behavioural therapy (CBT), helps to relieve guilt (Bee, McBeth, MacFarlane, & Lovell, 2016). For example, participants reported feeling guilty about not doing the recommended exercise: *. . .not doing the exercises that I'm supposed to do. . .and guilt rides on my shoulder like a gremlin. . .* (Darlow et al., 2013); and letting their doctor down because they were unable to control their condition: *As the patient, you're the focal point and you feel guilty internally and externally* (Rhodes et al., 1999).

Guilt relating to legitimacy of condition comprised two descriptive themes where participants reported guilt related to: (1) the lack of concrete evidence/diagnosis for their condition (Osborn & Smith, 1998; Serbic & Pincus, 2013), for example: *I feel guilty that I can't tell anybody something concrete, that I cannot give a specific reason* (Serbic & Pincus, 2013); and (2) feeling like a fraud or criminal when others did not believe their symptoms (Holloway, Sofaer-Bennett, & Walker, 2007; Wainwright, Wainwright, Keogh, & Eccleston, 2015).

Finally, guilt relating to how participants' actions/inactions affected others encompassed two descriptive themes, where participants expressed feeling guilty about: (1) not being able to work (Ryan, Lauchlan, Rooney, Hollins Martins, & Gray, 2014; Serbic & Pincus, 2013); and (2) not being able to carry out family and social roles (Mason, Skevington, & Osborn, 2004; Osborn & Smith, 1998; Serbic & Pincus, 2013; White & Seibold, 2008). For example, participants reported letting others down and not living up to their own and others' expectations of themselves. They discussed these feelings with reference to their relationships, family/parental responsibilities, and friends, for example: *. . .as your children grow up with you in pain, you are likely to feel guilty because they have to face issues and shoulder burdens that other kids don't. . .* (White & Seibold, 2008). These findings are further supported by Mason et al. (2004) second study, where they content analysed responses from 177 participants; guilt and burdening others facet was elicited by 17.5% of respondents.

Synthesis of quantitative evidence

Table 5 shows how evidence from quantitative studies was synthesized. Results were grouped according to the factors that were associated with guilt. This resulted in five broad categories, which included associations with: (1) pain and pain interference/ behaviours; (2) functional impairment; (3) psychological functioning (affect and cognitions/coping strategies); (4) social functioning; and (5) demographics. These associations are described in more detail below. If a relationship was examined twice within a study, for example, as a univariate relationship (e.g., Pearson's correlation or *t*-test) and as a multivariate relationship (e.g., multiple regression or SEM), both results were reported. Where quantitative results could be integrated with qualitative results, this is noted and examples are provided. This was achieved by looking for overlaps or associations between qualitative and quantitative evidence and reporting them. Quality assessment scores were similar and good overall (75–100%; see Table 3).

The included studies used either a dispositional measure of guilt such as guilt proneness (Conant, 1998; Turner-Cobb, Michalaki, & Osborn, 2015) or a situational measure. Two situational measures of guilt were used: (1) guilt/worry related to the impact of one's pain on significant relationships, and limitations on one's ability to provide care to others (Ziadni, You, Wilson, & Darnall, 2018); and (2) pain-related guilt, which consists of three subscales: social guilt (relating to how pain interferes with social life/

Table 5. Synthesis of quantitative evidence

Associations with guilt	Guilt type	Univariate effect size $r/r_s/\beta/d/\eta_p^2/k^2$ Significant values are in bold	Multivariate effect size $r/r_s/\beta/d/\eta_p^2/k^2$ Significant values are in bold	Guilt measure	Associated variable measure	Study
Pain						
Pain perceptions (severity)	Guilt proneness	.07 (r)		PFQ2	WHYMPI	Conant (1998)
Pain perceptions (interference)	Guilt proneness	**.25 (r)**		PFQ2	WHYMPI	Conant (1998)
Pain perceptions (severity and interference)	Guilt proneness	$(\beta)^a$		PFQ2	WHYMPI	Conant (1998)
Pain intensity	Social guilt	**.44 (r)**		PGS	NRS	Serbic et al. (2016)
	Managing condition/pain guilt	**.46 (r)**		PGS	NRS	Serbic et al. (2016)
	Verification of pain guilt	**.40 (r)**		PGS	NRS	Serbic et al. (2016)
	Guilt proneness	$(\beta)^a$	**.27 (k²)ᵇ**	TOSCA-3	NRS	Turner-Cobb et al. (2015)
	Social guilt	**.44 (r)**		PGS	NRS	Serbic and Pincus (2017)
	Managing condition/pain guilt	**.47 (r)**	**.16 (k²)ᵇ**	PGS	NRS	Serbic and Pincus (2017)
	Verification of pain guilt	**.38 (r)**		PGS	NRS	Serbic and Pincus (2017)
	Overall pain-related guiltᶜ	**.48 (r)**	**.12 (k²)ᵇ**	PGS	NRS	Serbic and Pincus (2017)
	Guilt/worry related to pain/relationships	**.32 (r_s)**		CARE	BPI	Ziadni et al. (2018)
Pain interference – general	Guilt/worry related to pain/relationships	**.55 (r_s)**		CARE	BPI	Ziadni et al. (2018)
Pain interference – activity	Guilt/worry related to pain/relationships	**.44 (r_s)**		CARE	BPI	Ziadni et al. (2018)
Pain interference – walking	Guilt/worry related to pain/relationships	**.34 (r_s)**		CARE	BPI	Ziadni et al. (2018)

Continued

Table 5. (*Continued*)

Associations with guilt	Guilt type	Univariate effect size $r/r_s/\beta/d/\eta^2_p$ η^2_p/k^2 Significant values are in bold	Multivariate effect size $r/r_s/\beta/d/\eta^2_p/k^2$ Significant values are in bold	Guilt measure	Associated variable measure	Study
Pain interference – work	Guilt/worry related to pain/relationships	.46 (r_s)		CARE	BPI	Ziadni et al. (2018)
Pain interference – sleep	Guilt/worry related to pain/relationships	.39 (r_s)		CARE	BPI	Ziadni et al. (2018)
Pain interference – relationship with others	Guilt/worry related to pain/relationships	.51 (r_s)		CARE	BPI	Ziadni et al. (2018)
Pain behaviours	Guilt/worry related to pain/relationships	.51 (r_s)		CARE	PROMIS	Ziadni et al. (2018)
Functional impairment						
Disability	Social guilt	.63 (r)	**.77 (β)**[b]	PGS	RMDQ	Serbic et al. (2016)
	Managing condition/pain guilt	.46 (r)	**−.46 (β)**[b]	PGS	RMDQ	Serbic et al. (2016)
	Verification of pain guilt	.29 (r)	−.19(β)[b]	PGS	RMDQ	Serbic et al. (2016)
	Guilt proneness	.09 (r)		TOSCA-3	SFMPQ	Turner-Cobb et al. (2015)
	Social guilt	.63 (r)	**.30 (k^2)**[b]	PGS	RMDQ	Serbic and Pincus (2017)
	Managing condition/pain guilt	.49 (r)	**.19 (k^2)**[b]	PGS	RMDQ	Serbic and Pincus (2017)
	Verification of pain guilt	.36 (r)	**.15 (k^2)**[b]	PGS	RMDQ	Serbic and Pincus (2017)
	Overall pain-related guilt[c]	.56 (r)		PGS	RMDQ	Serbic and Pincus (2017)
Physical function[d]	Guilt/worry related to pain/relationships	−.44 (r_s)		CARE	PROMIS	Ziadni et al. (2018)
Fatigue	Guilt/worry related to pain/relationships	.46 (r_s)		CARE	PROMIS	Ziadni et al. (2018)

Continued

Table 5. (*Continued*)

Associations with guilt	Guilt type	Univariate effect size r/rs/β/d/ηp²/k² Significant values are in bold	Multivariate effect size r/rs/β/d/ηp²/k² Significant values are in bold	Guilt measure	Associated variable measure	Study
Psychological functioning						
Affect						
Anxiety	Guilt/worry related to pain/relationships	**.47 (rs)**		CARE	PROMIS	Ziadni et al. (2018)
	Social guilt	**.50 (r)**	03 (β)[b]	PGS	HADS	Serbic et al. (2016)
	Managing condition/pain guilt	**.54 (r)**	**.23 (β)**[b]	PGS	HADS	Serbic et al. (2016)
	Verification of pain guilt	**.38 (r)**	.07 (β)[b]	PGS	HADS	Serbic et al. (2016)
	Guilt/worry related to pain/relationships	**.51 (rs)**		CARE	PROMIS	Ziadni et al. (2018)
Depression	Social guilt	**.61 (r)**	**.59 (β)**[b]	PGS	HADS	Serbic et al. (2016)
	Managing condition/pain guilt	**.51 (r)**	**.53 (β)**[b]	PGS	HADS	Serbic et al. (2016)
	Verification of pain guilt	**.38 (r)**	**.57 (β)**[b]	PGS	HADS	Serbic et al. (2016)
Anger	Guilt/worry related to pain/relationships	**.42 (rs)**		CARE	PROMIS	Ziadni et al. (2018)
Anger suppression	Guilt proneness	**.35 (r)**	**.22 (β)**[b]	PFQ2	STAXI	Conant (1998)
	Guilt proneness	**.42 (r)**	**.36 (β)**[b]	PFQ2	STAXI	Conant (1998)
Cognitions/coping strategy						
Diagnostic uncertainty (uncertain reported more guilt)	Social guilt	**.30 (d)**	−.11 (β)[b]	PGS	DU question	Serbic et al. (2016)
	Managing condition/pain guilt	**.46 (d)**	−.26 (β)[b]	PGS	DU question	Serbic et al. (2016)
	Verification of pain guilt	**.77 (d)**	**.61 (β)2**[b]	PGS	DU question	Serbic et al. (2016)
	Guilt proneness	−.06 (r)		PFQ2	MHLC-I	Conant (1998)

Table 5. (*Continued*)

Associations with guilt	Guilt type	Univariate effect size $r/r_s/\beta/d/\eta_p^2/\eta_p^2/k^2$ Significant values are in bold	Multivariate effect size $r/r_s/\beta/d/\eta_p^2/k^2$ Significant values are in bold	Guilt measure	Associated variable measure	Study
Internal health locus of control						
Pain acceptance (pain willingness subscale)	Social guilt	**−.53 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Managing condition/pain guilt	**−.40 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Verification of pain guilt	**−.26 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Overall pain-related guilt[c]	**−.45 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
Pain acceptance (activity engagement subscale)	Social guilt	**−.54 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Managing condition/pain guilt	**−.40 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Verification of pain guilt	**−.35 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Overall pain-related guilt[c]	**−.40 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
Pain acceptance (total)	Social guilt	**−.68 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Managing condition/pain guilt	**−.51 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Verification of pain guilt	**−.39 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
	Overall pain-related guilt[c]	**−.59 (r)**		PGS	CPAQ-8	Serbic and Pincus (2017)
Pain catastrophizing (rumination)	Guilt/worry related to pain/relationships	**.41 (r_s)**		CARE	PCS	Ziadni et al. (2018)
Pain catastrophizing (magnification)	Guilt/worry related to pain/relationships	**.41 (r_s)**		CARE	PCS	Ziadni et al. (2018)
Pain catastrophizing (helplessness)	Guilt/worry related to pain/relationships	**.51 (r_s)**		CARE	PCS	Ziadni et al. (2018)
Pain catastrophizing (total)	Guilt/worry related to pain/relationships	**.50 (r_s)**		CARE	PCS	Ziadni et al. (2018)

Table 5. (Continued)

Associations with guilt	Guilt type	Univariate effect size $r/r_s/\beta/d/\eta_p^2\,\eta^2/k^2$ Significant values are in bold	Multivariate effect size $r/r_s/\beta/d/\eta_p^2/k^2$ Significant values are in bold	Guilt measure	Associated variable measure	Study
Social functioning						
Isolation	Guilt/worry related to pain/relationships	.44 (r_s)		CARE	PROMIS	Ziadni et al. (2018)
(Perceived) punishing responses from others	Guilt proneness	.24 (r)	.23 (β)[b]	PFQ2	WHYMPI	Conant (1998)
Demographics						
Sex	Social guilt	.02 (d)		PGS	Male/female	Serbic and Pincus (2017)
	Managing condition/pain guilt	.07 (d)		PGS	Male/female	Serbic and Pincus (2017)
	Verification of pain guilt	.04 (d)		PGS	Male/female	Serbic and Pincus (2017)
	Guilt/worry related to pain/relationships	Women reported more guilt, $p < .001$; no effect size reported		CARE	Male/female	Ziadni et al. (2018)

Note. r, Pearson's correlation; r_s, Spearman's correlation; β, standardized regression (r, r_s, β: .1 = small effect, .3 = medium effect, .5 = large effect); *d*, Cohen's *d* (.2 = small effect, .5 = medium effect, .8 = large effect); η_p^2, partial eta-squared (.01 = small effect, .09 = medium effect, .25 = large effect); k^2, kappa-squared (.01 = small effect, .09 = medium effect, .25 = large effect); CARE, measure of interpersonal factors and their influence on self-care behaviours for adults with chronic pain (Ziadni et al., 2018); DU, diagnostic uncertainty question (Serbic & Pincus, 2013); HADS, Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983); TOSCA-3, Test of Self-Conscious Affect (Tangney & Dearing, 2002); RMDQ, Roland Morris Disability Questionnaire (Roland & Morris, 1983); PFQ2 Guilt, Personal Feelings Questionnaire-2 Guilt Scale (Harder & Zalma, 1990); PGS, pain-related Guilt Scale (Serbic & Pincus, 2014); WHYMPI, West Haven–Yale Multidimensional Pain Inventory (Kerns et al., 1985); STAXI, State–Trait Anger Expression Inventory (Spielberger, 1998); BPI, Brief Pain Inventory Short-Form (Keller et al., 2004); NRS, numerical rating scale; CPAQ-8, Chronic Pain Acceptance Questionnaire (Fish et al., 2010); PCS, Pain Catastrophizing Scale (Sullivan et al., 1995); PROMIS, Patient Reported Outcome Measurement Information System (Cella et al., 2007); MHLC-I, Multidimensional Health Locus of Control-Internal Scale (Wallston et al., 1978).
[a]Authors did not report statistical values; [b]k^2 –mediation analysis, which included several other variables, all of which are reported in Results section; [c]Overall pain-related guilt – a mean of these subscales: verification of pain guilt, social guilt, managing condition/pain guilt; [d]Higher scores reflect better physical functioning.

relationships), managing condition/pain guilt (relating to inability to manage pain), and verification of pain guilt (relating to the absence of clear evidence/diagnosis; Serbic & Pincus, 2017; Serbic, Pincus, Fife-Schaw, & Dawson, 2016).

Associations with pain intensity, interference, and behaviours

The relationship between pain interference and guilt proneness was significant (small effect size) in one study (Conant, 1998). This is supported by another study (Ziadni et al., 2018), in which guilt/worry related to pain/relationships was positively correlated with pain behaviours and pain interference, as well as with its five facets: activity, walking, work, sleep, and relationships (medium to large effect sizes). These findings were reflected in several participants' quotes relating to guilt stemming from how their actions or inactions affect others, for example: *She* [mother-in-law] *wanted her house decorating the other week. I said I can't even do my own and I feel guilty* (Osborn & Smith, 1998). Furthermore, pain intensity was positively associated with both guilt/worry related to pain/relationships and pain-related guilt in three studies (medium effect sizes; Serbic & Pincus, 2017; Serbic et al., 2016; Ziadni et al., 2018). However, the relationship between pain intensity and guilt proneness was not significant (Conant, 1998; Turner-Cobb et al., 2015).

Associations with functional impairment

A similar pattern of results was found when guilt was examined in relation to functional impairment, where a non-significant relationship was found between disability and guilt proneness (Turner-Cobb et al., 2015). However, guilt/worry related to pain/relationships and pain-related guilt were significantly correlated with poorer physical functioning and increased fatigue and disability (medium to large effect sizes, Serbic & Pincus, 2017; Serbic et al., 2016; Ziadni et al., 2018). When these relationships were examined as part of multivariate analyses by Serbic et al. (2016) (including depression, anxiety, pain, and diagnostic uncertainty in the model), they remained significant apart from the relationship between verification of pain guilt and disability. The largest effect sizes were for management of pain/condition guilt and social guilt, and this is also reflected in participants' quotes: '*You feel like you're letting people down, like when you should be able. . .to be a good friend or be a good employee, then you feel guilty 'cause you can't and it sucks*' (Serbic & Pincus, 2013).

Associations with psychological function – cognitions/coping strategies

In line with the above findings, pain-related guilt and its subscales were associated with reduced acceptance of pain and its subscales (Serbic & Pincus, 2017); effect sizes ranged from small to large, with social guilt reaching large effect sizes in all these analyses. Acceptance was also a mediator between pain intensity/disability and pain-related guilt subscales (medium to large effect sizes). The following quote, related to CBT treatment, reflects the role of acceptance: '*If I say to my husband, "I'm not going out, I really don't want to," I don't feel guilty now. Before I would never admit that, so from that perspective, things have changed, because I will say what I think and I will say no, I'm not doing it or I can't do it. So the way I cope with my pain has changed*' (Bee et al., 2016).

Guilt/worry related to pain/relationships was associated with increased pain catastrophizing and its subscales of rumination, magnification, and helplessness (medium to large effect sizes; Ziadni et al., 2018). In line with previously reported findings, when guilt proneness was assessed in relation to internal health locus of control (Conant, 1998), the relationship was not significant.

Guilt was examined in relation to diagnostic uncertainty in one study only (Serbic et al., 2016). Participants who were uncertain about their diagnosis reported more pain-related guilt (medium to large effect sizes for the three subscales) while the multivariate analysis (including depression, anxiety, disability, and pain) showed that diagnostic uncertainty was significantly and positively associated with verification of pain guilt (medium effect size). This relationship was also evident in participants' quotes, for example: '. . .*didn't like my GPs before – I just didn't like the face that they pulled, like, "Oh, again." They would do the note but I would leave their practice with a very guilty feeling and I was feeling like a criminal sometimes . . . [the pain] was real . . . I looked healthy, but it was true. . .*' (Wainwright et al., 2015).

Associations with psychological function – affect

Following the pattern of above results, guilt/worry related to pain/relationships (Ziadni et al., 2018) and pain-related guilt subscales (Serbic et al., 2016) were also associated with increased anxiety and depression (medium to large effect sizes). Although when these relationships were examined within a multivariate analysis (where disability, diagnostic uncertainty, and pain were also included), the relationship between anxiety and social guilt and verification of pain guilt were no longer significant (Serbic et al., 2016). This is also reflected in participants' quotes: *think your partners suffer, which makes you feel very guilty, and because you feel guilty, you then get depressed and it's a vicious circle, it really, really is* (Mason et al., 2004).

Anger was positively correlated with guilt (medium effect size; Ziadni et al., 2018) and guilt proneness (Conant, 1998; in both univariate and multivariate analyses) with small to medium effect sizes. The relationship with anger is also evident in participants' quotes: *I do feel guilty, I know that my back really does hurt and I'm not making it up and I feel sort of angry that I can't do it and I think well I wish I could just prove to them that my back really is bad* (Osborn & Smith, 1998).

Associations with social functioning

One study (Ziadni et al., 2018) found a positive significant relationship between isolation and guilt (medium effect size). Another study (Conant, 1998) found a positive significant relationship between guilt proneness and perceived punishing responses from others (small effect sizes). This relationship is also reflected in participants' quotes: '. . .*the people who were running the program would get angry with me [when I couldn't do the exercise correctly] and so it didn't actually help I felt guilty for a long time'* (Slade et al., 2009).

Associations with demographics

Females reported more guilt/worry related to pain/relationships (Ziadni et al., 2018). However, there were no significant differences between males and females on any types of pain-related guilt in another study (Serbic & Pincus, 2017).

Discussion

The aim of this review was to systematically appraise and integrate research evidence from studies that examined experiences of health-related guilt in people with chronic primary pain conditions. The key findings of the review of qualitative evidence were around the focus of guilt, which included three themes relating to the management of pain, diagnostic uncertainty/legitimizing pain, and how participants' actions or inactions affect others. These findings were then integrated with evidence from quantitative studies, which focused on factors associated with guilt, indicating that higher levels of guilt were associated with more pain (intensity, interference, and pain behaviours), functional impairment, and poorer psychological adjustment (affect, cognitions) and social functioning. No causal path can be inferred, but the evidence indicates towards a path in which the combination of persistent pain and impaired function results in a series of self/other evaluations, which lead to guilt, and that the presence of guilt exacerbates distress and further impairs relationships.

The process we propose for the emergence of guilt is congruent with the cognitive model of guilt-based PTSD (Lee, Scragg, & Turner, 2001). The theory, as applied to pain, suggests that if the meaning of the pain or the meaning of the consequences of the pain is associated with a violation or departure from personal standards of behaviour, and/or a feeling of responsibility for causing harm to others, guilt arises. This suggests that at the core of health-related guilt is the dissonance between the perceived ought self and the one lived-self. This explains why acceptance of pain may be a useful intervention to reduce guilt and, as a consequence, reduce distress.

An important feature of guilt centres around diagnostic uncertainty, and the imagined or correct perception that others disbelieve the pain experience and its consequences. This was particularly evident in patients' relationship with health practitioners (Rhodes et al., 1999; Slade et al., 2009; Wainwright et al., 2015). This suggests that reassuring patients and validating their pain experiences might be the first step towards helping them to avoid or reduce guilt. However, patients are also challenged at societal level and often face stigma. Potential solutions might involve enhancing public knowledge of chronic pain through education and development of interventions that target resilience of patients with chronic pain (DeRuddere & Kenneth, 2016).

Research suggests that generally guilt may be lessened through behaviours, which include apologizing and seeking forgiveness (Tangney et al., 2007), and that it can also have a positive role, motivating remorse, and reparative behaviours (Tilghman-Osborne et al., 2010). However, the relevance of such strategies in health-related guilt is highly questionable and they can even be inappropriate, as in the case of apologizing to health care providers for not improving. Instead, the relationships between guilt and various cognitions and coping strategies might provide a better insight on strategies to reduce guilt. For example, pain acceptance was associated with lower levels of guilt (Serbic & Pincus, 2017) and pain catastrophizing (Ziadni et al., 2018) was associated with increased levels of guilt, indicating that targeting underlying pain cognitions might be a beneficial approach.

Implications for future research and practice

The findings suggest that health-related guilt is perceived by people living with chronic pain as a consequence of the expectations they have from themselves, and those they perceive others to have from them, and overall, the impact of such guilt is negative,

leading to further distress and withdrawal. However, because of the methodologies used in the studies reviewed here, the relationships between pain and compromised function, guilt, and pain-related behaviours are yet to be clarified. Guilt might be a direct mediator between pain and distress, or may be part of vicious cycle in which guilt serves to reduce engagement, and leads to further impairment and reduced function. Future studies should use prospective designs and mixed methodologies combining qualitative reports of personal experiences, and quantitative measure of behaviours and clinical outcomes.

An important priority for future research is to establish how to reduce guilt in people living with chronic primary pain conditions. While guilt is a perfectly normal and often functional emotion, this review suggests that it can also be unhelpful. Recognizing and addressing this should be included in communication in routine and expert appointments, and, for people struggling to adjust, might include specific interventions within multidisciplinary or pain management psychological treatments. Our findings suggest that Acceptance and Commitment Therapy (ACT) and CBT might be potential treatments to target guilt, because they aim to change beliefs about self and others.

Furthermore, clearer definitions of guilt are needed when conducting investigations into health-related guilt. This requires a better understanding of guilt as an emotion, but it also requires an understanding of guilt from other perspectives such as philosophical and sociological. Combining these with psychological perspectives may help provide a better understanding of this complex concept (Tilghman-Osborne et al., 2010). Furthermore, researchers need to carefully consider whether to use a dispositional or a situational measure of guilt. Research shows that there is heterogeneity in both theoretical definitions and measurement of guilt, and the existing measures of guilt share only about 60% of the same concepts (Tilghman-Osborne et al., 2010). Our review of the literature showed that a substantial amount of research, in particular qualitative studies, did not clearly define guilt, and subsequently, findings were potentially compromised by other concepts that were often interpreted as guilt. Such studies were not included in our synthesis.

Limitations

There are some limitations to our review. The majority of relationships between guilt and associated factors were examined by a small number of quantitative studies; therefore, results should be interpreted with caution. In addition, the majority of qualitative studies did not specifically focus on guilt; hence, participants' references to guilt in those studies are often brief. The aim of this review was to develop a theoretical model through the synthesized data. However, no causal paths can be inferred based on our limited and mostly qualitative data. Furthermore, there were insufficient data to examine any chronic pain conditions in isolation, as the majority of the studies focused on chronic low back pain or a mix of chronic pain diagnoses.

As mentioned above, due to the lack of clarity about conceptualization of guilt, we included only qualitative studies in which participants directly expressed feelings of guilt. We believe that our approach brings clarity to our findings. However, it also assumes that people are always able to identify and discriminate their own feelings. The validity of the synthesis process was maximized by conducting the qualitative and quantitative syntheses separately and ensuring that each step of the synthesis was conducted independently by two reviewers, any disagreements were resolved by a third reviewer. Finally, the studies included in our review were mostly conducted in a small number of

Western countries. There could be cultural variations in guilt (Tilghman-Osborne et al., 2010), and future research should explore this.

Conclusions

This review indicates that health-related guilt is an important yet neglected issue in chronic primary pain conditions. It is associated with more pain and poorer function; thus, future research should examine it as a potential mediating/moderating factor leading to more distress and suffering. A better understanding of how it can be prevented, reduced, and managed is also needed. Our findings suggest that as a starting point, acceptance-based coping strategies and treatments and practitioner–patient communication should be key research targets.

Acknowledgements

Conflict of interest

Author contribution

Danijela Serbic (Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Project administration; Resources; Software; Supervision; Validation; Visualization; Writing – original draft; Writing – review & editing) Michael Evangeli (Formal analysis; Investigation; Methodology; Validation; Visualization; Writing – original draft; Writing – review & editing) Katrin Probyn (Data curation; Formal analysis; Investigation; Methodology; Software; Validation; Writing – review & editing) Tamar Pincus (Conceptualization; Formal analysis; Investigation; Methodology; Project administration; Resources; Supervision; Validation; Visualization; Writing – original draft; Writing – review & editing).

Data availability statement

This is a systematic review, and no primary data were collected. The review was registered with the PROSPERO, registration number: CRD42015029180.

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Full search strategy for one database (PubMed)