

Diagnostic Uncertainty, Guilt, Mood and Disability in Back Pain

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Abstract

Objective: In the majority of patients a definitive cause for low back pain (LBP) cannot be established and many patients report feeling uncertain about their diagnosis, accompanied by guilt. The relationship between diagnostic uncertainty, guilt, mood and disability is currently unknown. This study tested three theoretical models to explore possible pathways between these factors. In Model 1, diagnostic uncertainty was hypothesised to correlate with pain-related guilt, which in turn would positively correlate with depression, anxiety and disability. Two alternative models were tested: a) a path from depression and anxiety to guilt, from guilt to diagnostic uncertainty and finally to disability; b) a model in which depression and anxiety, and independently, diagnostic uncertainty, were associated with guilt, which in turn was associated with disability. *Method:* Structural equation modelling was employed on data from 413 participants with chronic LBP. *Results:* All three models showed a reasonable-to-good fit with the data, with the two alternative models providing marginally better fit indices. Guilt, and especially social guilt, was associated with disability in all three models. Diagnostic uncertainty was associated with guilt, but only moderately. Low mood was also associated with guilt. *Conclusions:* Two newly defined factors, pain related guilt and diagnostic uncertainty appear to be linked to disability and mood in people with LBP. The causal path of these links cannot be established in this cross sectional study. However, pain-related guilt especially appears to be important, and future research should examine whether interventions directly targeting guilt improve outcomes.

Introduction

Low back pain (LBP) is a highly prevalent condition, with a devastating impact on society. According to the Global Burden of Disease (GBD) Project 2010 (Lim et al., 2012) LBP has the highest global impact as measured by the number of years lived with disability, and it is now recognized as the leading cause of disability worldwide. Identifying factors that mediate recovery in LBP is vital for improving outcomes in patients with LBP. A plethora of tested predictors in prospective cohorts (Hayden, Dunn, van der Windt, & Shaw, 2010) suggests that psychological factors play an important role in the transition from acute to chronic LBP. Among the most robust predictors are depression, catastrophic cognitions, fear of movement and activity, and beliefs about recovery (Pincus & McCracken, 2013). Despite this, psychological interventions have delivered only small improvements in trials (Williams, Eccleston, & Morley, 2012). Underdeveloped theoretical models have been blamed for small and short-term effects of psychological interventions in LBP (Pincus & McCracken, 2013). This study aimed to test one mechanism – pain-related guilt associated with diagnostic uncertainty, which may compromise recovery in LBP.

In the majority of patients with LBP clear physical causes for back pain cannot be identified by current radiological methods (Krismer & van Tulder, 2007), which means that clear diagnostic labels can only be given to a small percentage of LBP patients. There is only limited research, mostly qualitative, into patients' perception and response to diagnostic uncertainty in LBP. This research has shown that patients often feel uncertain about the diagnosis and explanations for their LBP given by practitioners (Hopayian & Notley, 2014; Serbic & Pincus, 2013). Recent research (Serbic & Pincus, 2014a) has shown that patients' perception of their diagnosis is not clearly related to the labels and diagnoses they received from their health care providers (HCP), even when they agree with these. In a study of 68 patients, Serbic & Pincus (2014a) demonstrated that over 40% of patients who believed that

there was something wrong with their backs, yet undetected, also stated that they received and agreed with their diagnosis and/or explanation. The authors propose that this might reflect patients' belief that the diagnosis is correct but does not capture the true severity of their condition; or that it represents a belief that the diagnosis is correct, but that it fails to capture something else that is going on, in addition to the diagnosis. We have used this evidence as an operational definition for the term 'diagnostic uncertainty', utilised in the current study. Therefore, this study assessed 'perceived' diagnostic uncertainty in patients with mechanical LBP (for which there is no clear physical cause) by asking if they believed that there was 'something else' going on with their back, above and beyond any diagnoses or explanations they had been given.

Diagnostic uncertainty may impact on how patients feel and cope with their pain and they may continue searching for the causes of their back pain (Serbic & Pincus, 2013) instead of focusing on other important aspects of their pain and lives. There is some evidence that lack of knowledge about the cause of pain is associated with increased emotional distress, disability (Geisser & Roth, 1998; Reesor & Craig, 1988), pain intensity (Reesor & Craig, 1988), maladaptive pain-related cognitions such as catastrophizing (Geisser & Roth, 1998) and return to work (Lacroix et al., 1990). There is also some evidence that diagnostic uncertainty is associated with biased information processing in patients with LBP (Serbic & Pincus, 2014a), which is a hypothesised mechanism for the development and maintenance of depression. In the absence of a clear cause for their pain patients may feel that their pain is not legitimized and may feel guilty about this (Rhodes, McPhillips-Tangum, Markham, & Klenk, 1999; Serbic & Pincus, 2013). Feeling guilty about their pain may not only increase depression, but may result in increased disability-related behaviours, in an attempt to demonstrate that pain and suffering are real. Therefore, one mechanism via which diagnostic uncertainty might be linked to disability and mood is through feelings of guilt and the

primary aim of this study was to examine this hypothesized mechanism. The current study focused on people's individual understanding of guilt, manifesting in negative self-regard and painful feelings, often in reference to the perception of hurting other people (Kubany & Watson, 2003). A systematic review of research on the role of guilt (Tilghman-Osborne, Cole, & Felton, 2010) suggests that guilt is conceptually different from concepts such as anger, shame and blame and that measures of guilt should take this into consideration. Previous research (Rhodes et al., 1999; Serbic & Pincus, 2013; Serbic & Pincus, 2014b) has shown that pain-related guilt includes several aspects, including feeling guilty about being unable to provide a diagnosis and justification for pain (*verification of pain guilt*), being unable to control and manage pain better (*managing condition/pain guilt*) and failing to engage more in social situations (*social guilt*). A series of mixed methods studies (Serbic & Pincus, 2013; Serbic & Pincus, 2014b) resulted in the development of a pain-related guilt scale (PGS). By its definition *verification of pain guilt* seems to be directly linked to diagnostic uncertainty. *Managing condition/pain guilt* comprises of items which measure feeling guilty about seeing a number of different practitioners in search of help, and failing to respond to interventions. *Social guilt* includes items measuring a sense of guilt over letting friends and family down by failing to be sufficiently socially engaged and active due to pain.

This study tested three theoretical models. The *a-priori* predictions for Model 1 propose that diagnostic uncertainty is related to the three types of guilt which in turn relate to depression, anxiety and disability. The rationale here is based on the cognitive dissonance between having insufficient evidence for a physical cause of pain, and patients' own experience of pain and suffering. The conflict between these is hypothesized to result in guilt. There is preliminary evidence suggesting that LBP patients who cannot provide a diagnosis and justification for their pain feel guilty about this, as well as about being unable to control and manage their pain better and engage more in social situations (Rhodes et al., 1999; Serbic

& Pincus, 2013). The model further hypothesises that guilt will in turn be associated with increased depression, anxiety and disability. Associations between guilt and mood have been previously reported in groups with clinical mental disorders (Tilghman-Osborne et al., 2010) but the association between guilt and disability is unknown. The rationale to support this link is based on the assumption that patients may consciously or subconsciously increase their report of disability, in an attempt to reduce their own cognitive dissonance and guilt by demonstrating the legitimacy of their pain and suffering (Rhodes et al.; 1999; Salmon, 2000). They may also increase disability-related behaviours, such as avoidance of activity. Alternative models are based on the body of evidence suggesting that depression and anxiety lead to increased disability (reviewed in Pincus & McCracken, 2013). The models test how guilt and diagnostic uncertainty may be placed within this process. The first alternative model (Model 2) focuses on low mood in relation to guilt and diagnostic uncertainty. Guilt may result or be increased by low mood (Tilghman-Osborne et al., 2010), and in turn, decrease patients' ability to process and accept reassuring explanations from HCP that contradict the pessimistic and guilt-ridden perception accompanying their own pain experience, thus reinforcing the perceptions and concerns that something else, more serious, is going on with their backs. Finally, although the link between mood and guilt has theoretical underpinnings (e.g. Beck et al., 1961), diagnostic uncertainty may enhance guilt independently of mood (Model 3). In the absence of a visible cause for back pain, patients may feel that they are being perceived as imagining or exaggerating their pain or seeking attention. These perceptions are unhelpful, but may often be justified as there is evidence to suggest that a common response by orthodox medicine in situations where no clear causes for the pain can be found is to shift the responsibility back to the patient (May et al., 1999; McIntosh & Show, 2003). This may result in feelings of guilt that are not a direct outcome of negative affect. All three models propose pathways, with guilt as a mediator, to increased disability.

Methods

Participants

It was planned to have a diverse sample of participants with LBP, and to this end a total of 541 participants were recruited: 147 participants were recruited from two pain clinics and a physiotherapy department from the London National Health Service (NHS); 170 participants were recruited online and were members of three self-help groups for back pain. The remaining 224 participants were presenting for assessment and/or treatment in a clinic of osteopathy. Inclusion criteria were that participants be over the age of 18 years and have chronic (> 3 months) musculoskeletal LBP. No limit was imposed on current pain intensity. Participants with back pain due to ankylosing spondylitis, osteoporosis, cancer and inflammatory conditions such as rheumatoid arthritis were excluded. For participants recruited in NHS these inclusion criteria were checked for each participant by their clinician; for non-NHS participants this was established by self-report. The study received ethical approval from the university research ethics committee, NHS, and participating institutions.

Materials and Procedure

Online participants were invited to take part in the study through the three self-help groups for back pain which hosted a link to the questionnaire. The questionnaire was presented using an online survey tool (SelectSurveyASP Advanced v8.6.4). This tool imposed a level of control over questionnaire access and it did not allow completion of the questionnaire from the same computer more than once. Other participants were given a paper and pencil version of the same questionnaire. The following measures were used in the questionnaire:

Diagnostic uncertainty - was measured with a single categorical question “I think there is something else happening with my back which the doctors have not found out about yet (yes/no)”. This categorization created two groups of participants: those who responded

with a ‘yes’ were in the uncertain about diagnosis group, and those who responded with a ‘no’ were in the certain about diagnosis group. This question was part of a perceived diagnostic status categorization constructed from a qualitative study (Serbic & Pincus, 2013), and used in an empirical study of recall bias (Serbic & Pincus, 2014a). We selected this measure above other measures of diagnostic uncertainty, including items asking whether patients received and agreed with their diagnosis and/or explanation, because previous research (Serbic & Pincus, 2014a) suggested that it captured concerns that were not captured in these other measures. Specifically, over 40% of patients who reported there was something else happening with their back, undetected, still reported that they received clear diagnoses and explanations and agreed with them. The item was also associated with depression and disability in LBP (Serbic & Pincus, 2014a).

Pain-related guilt - The pain-related guilt scale (PGS) was developed in a mixed-methods series of studies (Serbic & Pincus, 2013, Serbic & Pincus, 2014b) and consists of 12 items and three subscales which represent three types of guilt in LBP: *social guilt* (4 items), which relates to letting down family and friends; *managing condition/pain guilt* (5 items), which is about being unable to overcome and control pain; and *verification of pain guilt* (3 items), which relates to the absence of objective evidence and diagnosis. Initial validations of the scale through exploratory and confirmatory factor analysis showed that the subscales had good validity and reliability (Serbic & Pincus, 2014b). The scale items are headed by the phrase “Because of my back pain I have experienced feelings of guilt:...”. Responses are on a Likert-type rating scale, ranging from 1 (‘never’ feeling guilty) - 5 (‘always’ feeling guilty). The PGS was developed because no other instruments exist to measure specifically pain-related guilt in persons with LBP or in chronic pain in general. Many measurements of (general) guilt exist (e.g. Personal Feelings Questionnaire-2 (Harder & Zalma, 1990)),

however, they do not refer to specific context, such as pain experience, thus they are too general for use in the context of chronic pain.

Anxiety and Depression - The Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983) consists of 14 items which is a screening measure of anxiety and depression (7 anxiety and 7 depression items). Scores range from 0 to 21 for each scale; higher scores indicate greater likelihood of depression or anxiety. Recommended cut-offs are: 8-10: mild cases, 11-15: moderate cases and 16 or above: severe cases (Zigmond & Snaith, 1983). The HADS is a well-known and widely used screening measure of anxiety and depression in medical populations.

Disability - Roland Disability Questionnaire (RDQ) (Roland & Morris, 1983) was used to measure back pain related disability. It is composed of 24 yes/no questions where 0 = no disability to 24 = maximum disability. This is a widely used and reliable measure of low back disability (Waddell, 2004).

Demographics and pain details - Participants were asked to give details about their age, gender, duration of their back pain (0-3 months, 3-6 months, 7-12 months, 1-2 years, 2-3 years, 4-5 years, 5+ years, 10+ years), and they were asked whether they had any other health-related problems or not. *Pain intensity* - was measured using a single question: 'How would you rate your back pain over the past week on a scale of 0 - 10, where 0 is 'no pain' and 10 is 'pain as bad as could be'? (Cleeland & Ryan, 1994).

Study Design

The study was cross sectional in design and it examined pathways within a theoretical model using structural equation modelling (SEM). SEM is used to evaluate whether theoretical models are plausible when compared to observed data, and it uses a complex form of multiple regressions to do this. **[Insert Figure 1 about here]**

The hypothesized Model 1 (see Model 1 in Figure 1) - A direct path between diagnostic uncertainty and mood was included, but not between diagnostic uncertainty and disability because preliminary analysis using point-biserial correlations between diagnostic uncertainty and mood and disability, showed that diagnostic uncertainty was correlated with depression $r_{pb}(413) = .145, p = .003$, and anxiety, $r_{pb}(413) = .170, p = .001$, but not with disability, $r_{pb}(413) = .065, p = .186$. Also, there were no significant differences between the certain and uncertain diagnosis group in their disability scores, but there were significant differences in their depression and anxiety scores (see Table 1). The preliminary analysis also showed that the two groups' pain-related guilt scores (for all three pain-related guilt subscales) were significantly different, supporting the relationship between diagnostic uncertainty and pain-related guilt (see Table 1). Indirect (mediating) effects between diagnostic uncertainty and the three outcome variables through each of the three types of pain-related guilt were calculated and reported (Klein, 2011).

Additional features of Model 1 - The residuals of the three guilt scales were permitted to correlate; this can be justified as all three are subscales of the pain-related guilt scale (PGS) (Serbic & Pincus, 2014b). The residuals for anxiety and depression were also permitted to correlate; this can be justified as both are subscales of the HADS (Zigmond & Snaith, 1983). In light of the evidence that depression and disability, and anxiety and disability are highly associated (Linton & Bergbom, 2011; Pincus & McCracken, 2013) and that the direction of these associations is not entirely clear, reciprocal pathways between these variables were included. Finally, there is substantial research evidence (Hayden et al., 2010) that pain intensity is a predictor of disability in LBP, thus the structural model also included this pathway, and it was connected indirectly to anxiety and depression via disability.

Alternative Models 2 and 3 - Two alternative structural models were also tested to examine whether Model 1 was the most viable model. The first alternative model proposed

that anxiety and depression preceded both guilt and diagnostic uncertainty (see Model 2 in Figure 1). The second alternative model proposed that the three types of pain related guilt are preceded by both anxiety and depression, and diagnostic uncertainty independently (see Model 3 in Figure 1). Additional features of Model 1 were also included in Models 2 and 3.

Planned analyses

Data preparation - Forty nine participants who reported suffering from non-musculoskeletal back pain (osteoporosis, back pain due to cancer and inflammatory conditions such as rheumatoid arthritis and ankylosing spondylitis) and acute back pain were excluded. Participants who were missing more than 10% of responses on any of the scales were also excluded from the analysis (Bennett, 2001). Because the scales used in study were subscales of the PGS and HADS they were short (3 to 7 items); this meant that if a participant missed only one item on a scale the responses already exceeded the cut-off of 10%. Participants missing data on the categorical (diagnostic uncertainty) and non-latent (disability and pain intensity) variables were also excluded. All together 79 participants were excluded due to missing data. Thus, the final sample included 413 participants, in both CFA and SEM analyses. In order to examine whether attrition would lead to bias, we compared the two groups of participants: out of 79 recruited participants with missing data 21 (19 of which were in the online sample) stopped responding after having answered only a few initial question, therefore, the remaining 58 participants with missing data were compared to the 413 included participants. There were no significant differences between the two groups on age, pain intensity and disability scores.

Structural equation modelling - The main statistical analysis was structural equation modelling (SEM). A two-step modelling approach was employed (Kline, 2011) whereby the structural regression model was first specified as a measurement model before the structural components were examined. The first step was to perform a CFA on the latent variables in

order to examine the validity of the measurement model and its adequacy for use in the structural model. The following latent variables were examined using CFA: *social guilt*, *managing condition/pain guilt*, *verification of pain guilt*, depression and anxiety. Based on the findings of a previous study (Serbic & Pincus, 2014b) these latent variables were allowed to correlate within the measurement model. These latent variables were then entered into the structural models (explained in the study design section) and examined with a SEM analysis. Both CFA and SEM were performed using AMOS 21, (Arbuckle, 2012) and the maximum likelihood estimation method was used. Both analyses were evaluated using a number of established goodness-of-fit indices. Initially, the chi-square statistic (χ^2) was evaluated as the initial indicator of model fit. Because the χ^2 has a tendency to indicate significant ill-fit in larger samples, model fit was assessed by establishing whether the observed chi square value was less than two times the model degrees of freedom (χ^2 /df) (Tabachnick & Fidell, 2013). The following goodness of fit indices were used: the Goodness of Fit Index (GFI > 0.95 close fit; GFI > 0.90 good fit); Adjusted goodness-of-fit index, which adjusts for degrees of freedom (AGFI > 0.90 good fit); Comparative fit index (CFI close to 0.95 close fit; CFI > 0.90 adequate fit) (Byrne, 2010; Hu & Bentler, 1999; Kline, 2011); SRMR- Standardized root mean square residual (SRMR < 0.08 good fit), Tucker Lewis index (TLI close to 0.95 good fit), and Root Mean Square Error Approximation (RMSEA < 0.06 good) (Hu & Bentler, 1999). When a model failed to meet these criteria, modification indices were inspected to indicate potential miss-specified parameters and they were used only when it was theoretically justified (Harrington, 2009). As the three models were not nested they were compared with AIC (Akaike Information Criterion,), and ECVI (Expected cross-validation index, single sample cross-validation index) measures (Byrne, 2010). The lower the AIC and ECVI measure, the better the fit.

Results

Description of sample

The sample characteristics are reported in Table 1, which also shows descriptive statistics for all variables used in the analysis. Participants who were uncertain about their diagnosis had significantly higher levels of pain, anxiety, depression and all three types of guilt. They also had pain for longer, although in both groups > 85% of participants had pain duration > 12 months. Additional analyses were conducted to compare the online participants vs. participants who were recruited within the NHS and BCOM clinic (who were seeking treatment). The online participants had higher levels of disability, $t(241.81) = 4.27, p < .001$, depression, $t(411) = .4.87, p < .001$, and anxiety, $t(411) = 3.89, p < .001$ than the participants who were seeking treatment. There were more participants who were uncertain about their diagnosis in the online sample, although this difference was not highly significant, $\chi^2(1) = 4.47, p = .035$. [Insert Table 1 about here].

Reliability and confirmatory factor analysis of the measurement model

Cronbach's alpha values were either good or excellent for the latent variables/scales: .93 for *social guilt*, .91 for *managing condition/pain guilt*, .87 for *verification of pain guilt*, .84 for anxiety and .84 for depression. No items had to be removed to improve these values. The CFA, after some minor alternations, demonstrated a good underlying structure of the measurement model. The CFA results are presented in the Supplementary Table 1.

Structural models

Model 1- The data fulfilled criteria for univariate (Kline, 2011) and multivariate normality (Bollen, 1989; Raykov & Marcoulides, 2008). Table 2 shows zero order correlations between all variables within the model and the model fit indices. Model fit was adequate to good. We also repeated the SEM analysis excluding the online sample, because they: a) reported higher diagnostic uncertainty, and b) we could not verify their true clinical

status from treating clinicians, as we did for the other samples. The fit indices remained very similar to the full sample fit indices. **[Insert Table 2]**

All standardized path coefficients are reported in Table 3. Diagnostic uncertainty was not directly correlated with depression, but the relationship was significant through *social guilt*. Diagnostic uncertainty was not directly correlated with anxiety, but it was through both *managing condition/pain and verification of pain guilt* (although the latter path was only marginally significant). Standardized path coefficients between diagnostic uncertainty and the three PGS subscales were all positive and significant. Being uncertain about diagnosis positively correlated with all three types of pain-related guilt. These correlations were moderate but significant. Participants who experienced *social guilt* (about letting down family and friends) were more likely to have more anxiety, depression and disability. The correlation between *social guilt* and disability was particularly strong (.834). Participants who had guilt about absence of objective evidence and diagnosis were more likely to have less anxiety (although this zero-order correlation was positive). *Managing condition/pain guilt* was significantly correlated with anxiety; participants who had a guilt about being unable to overcome and control pain were more likely to be more anxious. **[Insert table 3 about here]**

Alternative Models 2 and 3 - Fit indices for Model 2 and 3 were slightly better than for the hypothesised Model 1 and their AIC and ECVI were marginally lower (see Table 2). Fit indices for Model 2 were slightly better than for Model 3 and its AIC and ECVI were marginally lower. Direct and indirect effects for both alternative models are reported in Table 3. The table shows that in both alternative models anxiety was positively correlated to *managing condition/pain guilt*. Depression was positively correlated to all three types of guilt, and it was positively correlated to disability through *social and managing condition/pain guilt*. *Social guilt* was positively correlated with disability while *managing*

condition/pain guilt was negatively correlated with disability. Correlations between pain and disability, and disability and depression/anxiety were all significant.

Discussion

Main findings and fit with past research

The study explored three models of pathways via which two newly formulated and defined concepts, pain-related guilt and diagnostic uncertainty might be associated with disability in LBP. The pathways included: 1) pathways from diagnostic uncertainty to guilt, thus diagnostic uncertainty is associated with depression, anxiety and disability through specific pathways, depending on the focus of guilt; 2) pathways from anxiety and depression to guilt which are in turn associated with diagnostic uncertainty and finally with disability; and 3) independent pathways from depression and anxiety, and diagnostic uncertainty to guilt, followed by disability. All three models had a good fit with the data, but the best model was Model 2, emphasising the probable role of mood in association to all other factors. Model 2 and Model 3 had marginally better fit with the data than the first hypothesised model, but the differences between all three models' fit indices were very similar, suggesting that all three models are viable. This may suggest a cyclical relationship between the studied variables, which cannot be confirmed with cross-sectional data. The important questions for future research arising from the current findings focus on the need to reduce disability in LBP. Evidence from studies that attempt to reduce negative mood and cognitions in LBP populations with a primary outcome of reduced disability at follow up, are only partially successful (Pincus & McCracken, 2013), indicating that there is a need to identify and intervene on additional factors. Currently, such interventions only refer to diagnostic uncertainty in that they include elements of education about LBP, and there is no explicit goal or method of intervening on pain-related guilt. As all three models support a link between guilt and disability, new directions for research include addressing two key questions: a) can

interventions be designed to specifically address pain-related guilt; and b) will reductions in pain-related guilt improve other outcomes in these patients? In addition, there appears to be some mileage in exploring how to deliver effective reassurance through explanations that are acceptable to patients, without delegitimizing their suffering (Pincus et al., 2013).

Despite the limitations associated with the lack of time-line inherent in cross sectional studies, the findings highlight the roles played by both guilt and diagnostic uncertainty.

Pain-related guilt - Pain-related guilt in all three models was significantly correlated with mood and disability. The findings highlight some specific relationships between the different types of guilt, disability and mood. *Social guilt*, in particular, has strong associations with disability. While depression appears to be closely linked with all types of guilt, anxiety appears to be associated most closely with guilt about failure to manage one's pain. The association between *social guilt* and disability is particularly promising. While the causal path between these two variables is unknown, the possibility of a 'vicious cycle' in which disability increases *social guilt*, and the response to *social guilt* is further withdrawal from social engagement, in turn increasing isolation, disability and depression, warrants further investigation. Past research (Serbic & Pincus, 2013) showed that persons with LBP reported distancing themselves from other people to avoid feeling guilty about their pain-related behaviours. This explanation is also in line with theoretical explanations of guilt which describe it as a maladaptive state, motivating avoidance (Tilghman-Osborne et al., 2010).

Of interest is the negative relationship between guilt about failure to manage pain and disability (evident in Model 2 and 3), and between *verification of pain guilt* and anxiety, (evident in Model 1). The zero-order correlations between these pairs of variables were positive. This finding is puzzling, and is difficult to interpret. It might be an artefact of the interaction between the three types of guilt in the model. On the other hand, this could be explained through a positive behavioural response to guilt, in which patients who feel guilty

about their failure to respond to interventions increase their levels of activity and are more motivated to recover, resulting in lower rates of disability. Alternatively, high rates of guilt about failure to manage one's pain might affect responses to the disability questionnaire items, and result in lower scores. Future research should address this issue and examine if this pattern of results occurs in new samples.

The results support the findings from other studies (Serbic & Pincus, 2013, 2014b; Snelgrove, Edwards, & Lioffi, 2013) which show that pain-related guilt is a common experience among patients with LBP. High levels of pain-related guilt were reported by over 40% of participants with LBP in one study (Serbic & Pincus, 2014b). Several qualitative studies have suggested that an important focus of pain-related guilt is social. Thus, patients have reported feelings of guilt about letting their family down and about family members undertaking their responsibilities (Serbic & Pincus, 2013; Snelgrove et al., 2013) and feeling guilty in their marital interactions (Newton-John & Williams, 2006). In the context of uncertainty and absence of objective tests to verify their pain, patients report feeling guilty for 'letting the doctor down' (Rhodes et al., 1999). The results are also in line with a study exploring patients cognitions about the impact of their pain on their lives (Harris, Morley, & Barton, 2003), which found that the loss of social roles was particularly prominent, and closely associated with depression in patients with chronic pain.

Diagnostic uncertainty - The relationship of diagnostic uncertainty to other factors appears more modest, although significant. The findings do however suggest the diagnostic uncertainty is associated with guilt. We propose that even modest associations should be considered informative in studies of LBP, because of the lack of evidence about mechanisms leading to long term disability. For example, systematic reviews of prospective cohorts in LBP have concluded that no single predictor is conclusively and strongly linked to outcomes,

and that combining all known predictors explains only around 50% of the variance in outcomes (Hayden et al., 2010).

Mood - Models 2 and 3 suggest that mood plays a pivotal path in mechanisms leading to increased disability. Overall, both models show that depression is associated with all three types of guilt. Model 2, which was marginally a better model, suggests that depression drives pain-related guilt, and that certain types of pain-related guilt mediate between depression, disability and diagnostic uncertainty. Anxiety was positively correlated with guilt over one's inability to manage the condition and recover. Past research (Serbic & Pincus, 2013; Verbeek et al., 2004) suggested that this may be related to an increased search for a cure, and consequently increased health care utilisation. This may also suggest that these patients have unrealistic expectations about the treatment and management of their back pain (Serbic & Pincus, 2013; Verbeek et al., 2004). The findings add to a large body of evidence suggesting that eliciting and addressing depression and anxiety should be a priority in managing LBP, especially in light of evidence suggesting that current practice fails to do so adequately, especially in primary care (van der Windt, Hay, Jellema, & Main, 2008).

Strengths and limitations

In order to improve the outcomes of interventions in LBP it is necessary to understand better the specific mechanisms that lead to poor outcomes (McCracken & Morley, 2014; Pincus & McCracken, 2013). Therefore, strength of the current study is that it identifies factors, previously unexplored, and sets to examine how they might fit within known associations, between mood and disability. The sample was varied and representative of both participants who were treated and those that were not seeking treatment for their back pain. It was also representative of both private and NHS patients.

There are also several limitations. Although causal path modelling is often presented as a method to assess causality between a set of variables, causality cannot be established in

the absence of a timeline (Kline, 2011). Our study was cross sectional and therefore causation cannot be inferred from the findings.

Diagnostic uncertainty was only moderately correlated with the three types of pain-related guilt. While the findings may be due to limitations in our measure of diagnostic uncertainty, they also might suggest that our measures of guilt are not comprehensive, and might be missing specific focuses, for example feeling guilty during periods of absence from work due to the impact of this on work colleagues (Wynne-Jones et al., 2011). Anger, frustration and blame may also be important concepts, but they were not the focus of the current investigation. Research on pain-related guilt is extremely limited, and is almost exclusively reported in qualitative studies that did not specifically set out to study guilt. Furthermore, research evidence suggests that guilt is culturally distinct (Tilghman-Osborne et al., 2010); therefore our findings may not be entirely applicable in non-western cultures. The samples recruited for the current study included an online sample (people subscribing to self-help groups) who might have been more self-motivated to take part in the study and express their pain related concerns; this group had higher levels of disability, depression and anxiety than the participants who were seeking treatment.

Conclusions and directions for future research

To our knowledge, this study represents the first investigation to systematically examine the relationships between diagnostic uncertainty, pain-related guilt and disability and mood in persons with chronic LBP. The findings suggest that diagnostic uncertainty is moderately associated with pain-related guilt but further research is needed to fully understand the strength and meaning of this association. Pain-related guilt, and especially social aspect of guilt, are important factors closely associated with disability, and mood. Future research should focus on further clarifying these relationships using longitudinal designs. Like the majority of studies in LBP patients, this study measured reported disability,

but failed to measure changes in behaviour in response to diagnostic uncertainty and increased guilt. Future prospective studies should measure not only reported mood and disability, but also explicitly measure changes in behaviour. Promising improvements in technology, such as unobtrusive accelerometers and other wearables could improve the measurement of function, which could be distinguished from reported disability.

Such studies may shed light on the most effective ways to introduce interventions to reduce the associations between diagnostic uncertainty, mood, guilt, (especially social guilt), and subsequent unhelpful behaviours, if such associations are evident in future studies. In this context we note that clear causal paths are often unclear in the evidence from pain populations. For example, the causal path between depression and disability, much debated, remains unclear and likely to be cyclical (Linton & Bergbom, 2011). More important, perhaps, is how interventions to reduce one factor impact long term on the other factors.

In addition, future research should integrate the new factors that are the focus of the current study into broader models that include evident cognitive constructs that are likely to be related to either diagnostic uncertainty or pain related guilt. Past research showed that many LBP patients who believed there was something else, undiscovered going on with their back, still said they were given a diagnostic label for their pain (Serbic & Pincus, 2014a). This might suggest that diagnostic labels do not always reduce diagnostic certainty in LBP patients, and that diagnostic uncertainty might stem from worry and beliefs about the pain, which may in some patients lead to catastrophic thoughts (Quartana, Campbell, & Edwards, 2009). This is important to study because catastrophizing has been identified as a key mechanism leading to poorer outcomes in LBP patients (Pincus & McCracken, 2013). Catastrophic pain perceptions may also potentially increase pain-related guilt or be increased by it, and indirectly place pressure on the emotion regulation system (Linton & Bergbom, 2011). Future research could also examine whether patients with less effective emotion

regulation systems might be more prone to experience diagnostic uncertainty and pain-related guilt. For instance, there is some research showing that perceived control over pain might be linked to diagnostic uncertainty, but further research is necessary to examine this relationship (Geisser & Ruth, 1998). Other relevant factors include acceptance and avoidance, which have been described as two extremes of the same concept (de Boer et al., 2014); patients who engage in avoidance behaviours are usually less acceptant of their pain and pain experiences. Acceptance of pain has been associated with less pain, pain-related anxiety, avoidance, depression and disability (McCracken, 1998). Therefore, future research could examine whether changing diagnosis-related perceptions may lead to a greater acceptance of pain and pain experiences, and whether interventions that aim to increase acceptance result in reduced guilt and avoidance.

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Conflict of interest statement

The authors report no conflict of interest.

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