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Activity Limitation in Joint Hypermobility Syndrome

Journal of the American Academy of Orthopaedic Surgeons

Volume 18, Number 10, October 2004

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D.Clin.Psy. Thesis (Volume 1), 2007

University College London

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Overview

This thesis is divided into 3 parts:

Part 1 is a literature review that examines the role of fear and avoidance in chronic pain, focusing in particular on their role in activity limitation. It examines the existing fear-avoidance models, in particular Vlaeyen and Linton's (2000) model, it reviews the evidence for this model, and considers it in the context of models of fear and avoidance in other psychological disorders. Evidence is reviewed for other factors which contribute to activity limitation in chronic pain.

Part 2 is an empirical study which focused on activity limitation and the process of decisions about limitation of activities, in order to elucidate the applicability of Vlaeyen and Linton's (2000) fear-avoidance model to people with JHS. Using a qualitative approach, a complex decision making process was revealed in which each decision was individually considered in a cost-benefit analysis, weighing the importance of the activity against its potential aversive consequences, which is not adequately described by Vlaeyen and Linton's (2000) model.

Part 3 is a critical review of the process of carrying out the empirical research, and encompasses all aspects of that research, including the choices about the design, carrying out the interview with participants, further reflections on the participants' impact on the research process, the analysis and the results.

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Acknowledgements

I would like to thank the people who participated in this study, and acknowledge their commitment to contributing to research into Joint Hypermobility Syndrome. I would like to thank my supervisors of this research project, Amanda C de C Williams and Nancy Pistrang. I am indebted to my course tutor, Kerry Young, for her invaluable support. I would like to thank Paul, Thomas and Roseanna Schmidt for their patience, tolerance and support. Finally I would like to acknowledge the interest and support of my friends, family and colleagues.

Part 1: Literature Review

A review of fear and avoidance in chronic pain

Abstract

This review examines the role of fear and avoidance in chronic pain, focusing in particular on their role in activity limitation. It examines the existing fear-avoidance models, in particular Vlaeyen and Linton's (2000) model, it reviews the evidence for this model, and considers it in the context of models of fear and avoidance in other psychological disorders. Evidence is reviewed for other factors which contribute to activity limitation in chronic pain. In the context of this evidence, it is concluded that whilst there is much support for Vlaeyen and Linton's model, it may be useful to consider alternative models describing the process of activity limitation, and future research is proposed to support this.

Introduction

This paper will examine the role of fear and avoidance in chronic pain, focusing in particular on their role in activity limitation and their role in the progression from acute to chronic pain and disability. It will examine existing fear-avoidance models of chronic pain, reviewing the evidence supporting them, and considering them within the context of models of fear and avoidance in other psychological disorders. The evidence will then be reviewed for other factors which contribute to the limitation of activity and restriction of the lives of chronic pain sufferers, and an attempt will be made to consider how successfully the fear-avoidance models of chronic pain are able to account for these influences.

Method

The search strategy for this paper, involved using the search engine, “Google Scholar”, with “pain”, “disability” and “avoidance” as key words. The search initially included papers published from 1995 onwards, but this was later limited to focus mainly on papers published from 2000 onwards. The reason for this was that the model on which the paper focused (Vlaeyen and Linton’s model) was published in 2000.

Chronic pain

Pain is defined as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage” (International Association for the Study of Pain, 1979). It is a subjective experience and is primarily measured subjectively, not estimated by signs of damage or disease. Chronic or persistent pain is defined by timescale – pain which has lasted more than

three months – and implicitly has lasted beyond the time required for healing or resolution of lesions or pathology responsible for pain onset, and the term chronic pain is usually reserved for pain not attributable to a life-shortening or progressive disease (Williams, 2007). Chronic pain interrupts behaviour, interferes with functioning, and may affect a person's identity: their sense of who they are and what they might become (Harris, Morley, Stephen, & Barton, 2003). It can result in disability and 30% of those with neck, shoulder, or back pain report limitations in daily life (Denison, Asenlof, & Lindberg, 2004). The point prevalence of chronic pain in England has been estimated to be 11.2% (Croft, Rigby, Boswell, Schollum, & Silman, 1993). A survey of patients in general practices in Scotland, using self-report, and defining chronic pain as pain or discomfort which has persisted continuously or intermittently for more than 3 months, estimated that 46.5% of the general population have chronic pain (Elliott, Smith, Penny, Smith, & Chambers, 1999). The authors noted that previous estimates of chronic pain in the general population had ranged from 2% to 45%, which reflected differences in research methods and definitions used (Elliott et al., 1999).

Models of chronic pain

The earliest models of chronic pain are biomedical models, dating back thousands of years to Ancient Greece, which assumed a direct link between disease and physical pathology, and assumed that psychological, social and behavioural mechanisms were not important in disease, and hence failed to account for individual differences in pain perception and development of chronic pain (Asmundson, Norton, & Vlaeyen, 2004). The Gate Control Theory of Pain (Melzack & Wall, 1965) suggested that processes mediated by the central nervous system, such as

cognition and affect, could directly influence the transmission and perception of nociceptive sensory information from the periphery of the body. It was the first theory to explain individual differences in perception of pain, and how the same individual could perceive pain from a similar injury differently on separate occasions (Asmundson et al., 2004). However, it did not provide a clear explanation for the persistence of pain after damaged tissue has apparently healed. Nevertheless, it provided the framework for further research in this area (Dickenson, 2002).

The biopsychosocial approach attempts to integrate biological, psychological and social components of pain, and has contributed to explaining pain which seems to be incongruous with the extent of tissue damage, or which persists in the absence of tissue damage or organic pathology. Fear was an important element of several biopsychosocial models of pain, and several investigators observed an association between pain and significant degrees of anxiety (e.g. Rowbotham, 1946, cited in Asmundson et al., 2004).

A behavioural model was proposed by Fordyce (1976) in which reinforcement maintained avoidance behaviours associated with acute injury, causing them to become chronic and hence promoting disability. Central to this model was the idea of operant learning of avoidance behaviour. Avoidance behaviour is negatively reinforced through reduction in suffering associated with nociception. Whilst for the majority of individuals experiencing acute pain, avoidance behaviours are gradually replaced by approach behaviours facilitating a return to pre-injury activity levels, in a small number, the negative reinforcement contingencies (such as reduction of pain) can shift to other positive (such as receiving increased attention as a result of injury) and negative (such as reduced work or family responsibilities) reinforcement contingencies that, in turn, maintain avoidance behaviour. Hence avoidance is

maintained by learning that avoiding activities associated with pain reduces the likelihood of a new episode of pain (Fordyce, 1976).

Turk, Meichenbaum and Genest (1983) introduced a cognitive behavioural perspective to understanding pain, in which cognitive factors such as attribution, expectancies and self-efficacy were also regarded as important in influencing the experience of pain. This cognitive behavioural perspective was applied to the early fear-avoidance models which attempted to explain how fear of pain and avoidance behaviour contributed to the maintenance of pain in the absence of identifiable organic pathology (Lethem, Slade, Troup, & Bentley, 1983). In a review of the evidence for the role of avoidance of daily activities in maintaining chronic pain, Philips (1987) concluded that ‘the avoidance is extensive and complex and includes avoidance of stimulation, movement, activity, social interaction and leisure pursuits.’

From this developed the current fear-avoidance models of chronic pain, on which the rest of this paper will focus.

Models of fear and avoidance in chronic pain

Vlaeyen and Linton’s fear-avoidance model of pain

Vlaeyen and Linton’s (2000) fear-avoidance model of pain proposed that when a catastrophic meaning is placed on an experience of pain, this leads to pain-related fear (fear of pain, fear of (re)injury), which in turn spirals into a cycle of hypervigilance to bodily sensations and avoidance behaviours, which promotes and maintains activity limitations, disability and depression. These latter will maintain the pain experiences, thereby fuelling the vicious circle of increasing fear and avoidance. In patients who do not catastrophise, no pain-related fear occurs, permitting rapid confrontation with daily activities, leading to a fast recovery. Pain

catastrophising is assumed to be influenced by negative affectivity and threatening illness information (Vlaeyen & Linton, 2000).

Vlaeyen and Linton's (2000) model is consistent with the notion that the likelihood of developing a chronic pain-related condition will be significantly elevated if the person has both a tendency to be hypervigilant towards internal pain sensations and a tendency to interpret those sensations as dangerous or potentially threatening to their well-being (Asmundson et al., 2004). It has been suggested that the fear of pain and (re)injury may be more debilitating than pain itself, and that this refutes the early notion (for example from biomedical models) that the lowered ability to accomplish tasks of daily living in chronic pain patients is straightforwardly a consequence of pain severity (Vlaeyen, Kole-Snijders, Rotteveel, Ruesink, & Heuts, 1995). Possible mechanisms may be a misinterpretation of bodily symptoms, inaccurate predictions about pain, and hypervigilance for pain related symptoms and information. Crombez, Vervaeke, Baeyens, Lysens, and Eelen (1996) found that pain expectancies intensify escape or avoidance tendencies, but do not amplify pain intensity, thus predicting an increase in hypervigilance and disability in the absence of an increase in pain intensity.

Empirical evidence for Vlaeyen and Linton's model

Vlaeyen et al. (2000) found that fear of experiencing pain prompts avoidance of daily activities which in turn maintains fear of pain. Swinkels-Meewisse, Roelofs, Oostendorp, and Vlaeyen (2003) found that patients with acute low back pain who reported a high fear of pain were significantly more likely to experience high levels of disability and to avoid participation in a range of home, work, social, and leisure activities. Vlaeyen and colleagues also found evidence that exposure to fear-eliciting

activities resulted in reductions in pain related fear, supporting the notion that avoidance of daily activities maintains fear of pain (Vlaeyen, de Jong, Geilen, Heuts, & van Breukelen, 2001). In this study, with chronic low back pain patients reporting substantial fear of movement/(re)injury, they compared the effectiveness of a cognitive-behavioural graded exposure in vivo treatment with graded activity, in reducing pain-related fears, pain catastrophising and pain disability. (Graded exposure involved engaging in fear-provoking activities and movement previously avoided, until anxiety levels had dissipated. Graded activity involved engaging in activities which scored low on the patient's pain hierarchy, until pain prevented them from continuing.) They found that graded exposure resulted in reductions in fear of movement/(re)injury, pain catastrophising and fear of pain, and these changes were not observed in graded activity. Furthermore, the reductions in catastrophising and pain-related fear correlated with a decrease of self-reported functional disability in daily life (Vlaeyen et al., 2001).

In a study with just two participants, Linton, Overmeer, Janson, Vlaeyen, and de Jong (2002) adopted a treatment approach in which fear-avoidance was regarded as a phobia, and in vivo exposure techniques were applied, with the use of a fear thermometer and graded exposure. The results showed substantial improvements for both patients as they increased their function and decreased their fear.

Woby, Watson, Roach, and Urmston (2004) measured the effects of a cognitive-behavioural based intervention in 54 chronic low back pain patients. The study found that changes in the cognitive factors (catastrophising, fear-avoidance beliefs, and appraisals of control) were not significantly associated with changes in pain intensity. In contrast, reductions in fear-avoidance beliefs about work and physical activity, as well as increased perceptions of control over pain were uniquely related

to reductions in disability, even after controlling for reductions in pain intensity, age and sex. This lends support to Vlaeyen and Linton's notion that the fear is more debilitating than the pain itself.

The Vlaeyen and Linton model assumes that some patients catastrophise about pain, and this leads to pain-related fear and hence avoidance, whilst other patients do not place a catastrophic meaning on the pain and hence follow a path to recovery. However, Ciccone and Just (2001) questioned why some patients with acute pain follow one path, whilst others follow the other path. It has been pointed out that while the "no catastrophising" pathway in Vlaeyen and Linton's model leads to "recovery," it is possible that individuals on this pathway may have acknowledged that a permanent cure for their pain is unlikely and have learned to accept their pain (Goubert, Crombez, & De Bourdeaudhuij, 2004), that is, it may not be recovery in the sense of freedom from pain and disability. Thus in practice, the alternative to fear, avoidance and disability may not necessarily be no pain and no disability as implied by Vlaeyen and Linton's model.

Nevertheless, Ciccone et al. (2001) hypothesized that behavioural avoidance is due to cognitive expectation, according to the belief proposed by Philips (1987) that strenuous work is likely to cause an increase in pain. Therefore they attempted to understand the cognitive components of fear-avoidance, and looked at anticipated pain and anticipated injury. Pain and injury expectancies explained 40% to 35% of the variance in work disability compared with 12% to 10% explained by fear and avoidance (measured by the FABQ-W¹) for the acute and chronic samples, respectively. After controlling for pain duration, depression, somatization, and current pain severity, pain expectancy alone accounted for 16% of the variance in

¹ The FABQ-W is the work subscale of the Fear-Avoidance Beliefs Questionnaire, described by Waddell, Newton, and Henderson (1993) which measures avoidance beliefs related to work.

patients in the chronic group and 33% of the variance in patients in the acute group. Whilst both pain and injury expectancies were associated equally with work disability for patients in the acute group, only pain expectancy accounted for variance in the chronic group. These results suggested that fear-avoidance beliefs, in the form of cognitive expectancies, are not the result of prolonged pain exposure but rather operate in patients with acute injury who must decide whether and when they should return to work, which supports Vlaeyen and Linton's (2000) conceptualisation of the process. However, this was a cross-sectional study and in the absence of a longitudinal study, it is not possible to determine whether fear-avoidance beliefs in chronic pain patients are influenced by prolonged pain and suffering or whether these are already elevated at the acute stage and can be considered as predisposing factors.

Crombez et al. (1996) found in a study of back pain patients that a high expectation of pain co-occurred with a fear of (re)injury, and a lower level of performance on an exercise task. These results are consistent with conditioning models which state that pain expectations are associated with a fear response and an urge to avoid the pain.

Grotle, Vollestad, Veierod, and Brox (2004) also found support for the Vlaeyen and Linton model at both the chronic and acute stages, in a comparison of fear-avoidance beliefs and distress in patients at an early stage of lower back pain (LBP) with those at a chronic stage. Although the levels of fear-avoidance beliefs and distress were significantly lower in the acute compared to the chronic sample, in both acute and chronic low back pain, fear-avoidance beliefs and distress were significantly related to disability after adjusting for sociodemographic, pain, and clinical variables. The results were in line with the assumptions in Vlaeyen and

Linton's model, that fear-avoidance beliefs and distress are linked to disability and provide further evidence for the validity of this model not only for patients with chronic LBP, but also for patients with acute LBP.

Alternative perspectives

Boersma and Linton (2005) found that the relationship between fear of movement and impairment of daily activity due to pain (disability), is moderated by the stage of chronicity, with fear of movement explaining variance in daily activity impairment due to pain, where pain duration was longer than 1 year, but not below 1 year, suggesting that the time point in the development of a musculoskeletal pain problem might be an essential aspect of the importance of the relationship between psychological components and disability. They also found that pain intensity and fear of movement were not significantly related at any stage of pain duration, while disability and fear of movement appeared to become increasingly associated as the duration of pain progressed from under 1 year, to between 1 and 3 years, to over 3 years. Thus, there was an indication that fear of movement and disability could be differentially related across the stages of chronicity. Depression and disability were strongly correlated at all three stages, suggesting an involvement of negative affect in functional difficulties.

From a review of prospective studies on the determinants of chronic disability Truchon (2001) identified 3 factors which were helpful in predicting chronic disability (defined as inability to work) in lower back pain. Firstly medical factors, such as obtaining positive results for clinical tests (for example the presence of radiating pain). Secondly psychosocial factors such as the worker's appraisal of his/her capacity to perform a task, dissatisfaction with work, or problematic

relationships with co-workers. Thirdly, psychological predictors, for example reporting intense and persistent pain disproportionate to medical measures or organic pathology, or a negative cognitive appraisal of pain. Truchon concluded that this reveals the biopsychosocial character of lower back pain chronic disability.

Truchon and Fillion (2000) went on to apply a particular biopsychosocial model, the Stress Coping Model (Cohen, Kessler, & Gordon, 1995) to understanding chronic disability in lower back pain. They suggested that in the case of lower back pain, the mechanism may be as follows: an environmental demand in the form of a low back pain episode and related stressors such as limited information about aetiology, persistence of symptoms, stressors at work, treatment failures, or litigation, leads to a negative impact on the biological responses through the endocrine and immune systems. These in turn have a negative impact on the cognitive appraisal of the demands (in other words the personal resources are considered inadequate to manage this threat due to lack of control or low self-efficacy), which generates a negative emotional response to the stressor (such as anger, fear or anxiety). This impacts negatively on the behavioural response, resulting in avoidance of the threatening situation, which increases the risk of chronic disability developing (Truchon, 2001). The advantage of this model is that it incorporates the physiological impact on the psychological response, as well as the psychological and psychosocial aspects.

A review of fear and avoidance in chronic pain noted several studies reporting an increase in fear and avoidance in chronic pain patients that was not limited to fear and avoidance of physical activity, but extended to enhanced fear and avoidance of social activities and health related issues (Asmundsen, Norton, & Norton, 1999). Morley and Eccleston (2004) suggested that a range of feared objects in chronic pain is to be expected because of the overwhelming threat value of pain and its capacity to

interrupt current thinking, interfere with almost every aspect of daily life, and threaten the person's identity, both current and future concepts of self. They saw chronic pain as presenting multiple threats and threats to identity.

Fear and avoidance in other psychological disorders

Vlaeyen and Linton's (2000) model predicts that fear of experiencing pain prompts hypervigilance for sensations of pain and avoidance of daily activities, leading to disuse, depression and disability, which in turn maintains catastrophic beliefs about pain, and hence maintains fear of pain (Vlaeyen et al., 2000). This is consistent with clinical psychology literature pertaining to other disorders, in which attention towards threat is associated with increased negative thoughts and perception of threat (Clark et al., 1997) and with avoidance of, or withdrawal from, threatening situations. The behaviours and mental processes used in an attempt to reduce, avoid, escape or alleviate threat or fear have been referred to as safety behaviours (Salkovskis, 1989). Safety behaviours contribute to the persistence of disorders by preventing disconfirmation of unhelpful beliefs and increasing the risk of the feared outcome actually occurring. It can be difficult to draw a distinction between escape and avoidance on one hand and adaptive coping strategies on the other (Thwaites & Freeston, 2005).

Escape and avoidance refer to situations in which an individual does not enter, or prematurely leaves a fear-evoking situation (Harvey, Watkins, Mansell, & Shafran, 2004). If a threat is detected, a patient is likely to experience high levels of anxiety, which is aversive and could itself be interpreted as a sign of impending danger. Hence the patient leaves the situation, avoids it in future and believes it is dangerous. However such avoidance has several negative consequences: It may serve to

maintain unhelpful beliefs about the extreme danger of the situation as it removes the opportunity to disconfirm negative beliefs (Salkovskis, 1991), it denies the person the chance for positive reinforcement and could thereby contribute to the maintenance of low mood (Martell, Addis, & Jacobsen, 2001; Salkovskis, 1991), and it narrows the person's interests and reduces the number of external stimuli present in the environment, which may exacerbate self-focused attention and recurrent thinking (Harvey et al., 2004). Avoidance behaviour is intrinsically problematic insofar as it interferes with functioning (Harvey et al., 2004). According to learning theory, for example Mowrer (1960), avoidance is negatively reinforced and can become self-perpetuating. According to Mowrer's two stage model of fear and avoidance (Mowrer, 1939; Mowrer, 1960), avoidance behaviour is reinforced when it is followed by a reduction in anxiety. That is the avoidance persists because it works: it reduces anxiety. Vlaeyen and Linton's fear-avoidance model has clear parallels with this: avoidance of activity reduces fear of pain and fear of (re)injury, hence the avoidance is negatively reinforced and is maintained.

In looking for parallels in other psychological disorders regarding the question of why acute pain progresses to chronic pain in some people but not others, the cognitive model provides some suggestions. In terms of the cognitive model, an individual progresses to develop a psychological disorder, such as depression, phobia and other anxiety disorders, as a result of the interaction of precipitating factors with key developmental events which may have predisposed the individual to enduring patterns of interpreting these events, that is, their beliefs and assumptions (schemata), and hence to emotional disorder (Beck, 1995). This is a diathesis-stress approach, in which (for example in depression) individuals with depressogenic schemata (diathesis) are more likely than others to develop depressive symptoms following

negative events (stress) (Beck, 1983). This cognitive approach is adopted in Vlaeyen and Linton's model, which assumes that the individual's initial interpretation of the acute pain episode as either threatening, or not threatening, determines whether an individual is like to become disabled by the pain.

An alternative explanation for why some individuals with chronic pain avoid activity, while others do not, may be found by reference to Klinger's current concerns theory (Klinger, 1996). This theory helps to explain why some stimuli are avoided at certain times and under particular circumstances, whilst other stimuli are not. Current concerns appear to determine specific stimuli that are attended to and remembered, the specific situations that are misinterpreted, the content of thought and the specific behaviours that are used to avert danger (Harvey et al., 2004). Why do people with different psychological disorders have different current concerns? It is likely that a variety of factors determine this, including biology (for example genes), personality, learning history, traumatic experiences and culture (Klinger, 1996; Wells, 1997). It is possible that some of these same factors determine which individuals who experience an acute episode of pain or suffer an injury go on to develop chronic pain, that is the individual's current concerns pertaining to their experience of pain or injury may determine their response to that pain or injury.

There is a lot of evidence in support of exposure as the method of choice to reduce avoidance across the anxiety disorders (Harvey et al., 2004). Marshall (1985) found that exposure was inferior if it was terminated while the participant was in a high state of anxiety relative to when participants only left the situation when their fear levels had declined, as prolonged exposure is necessary to allow cognitive reappraisals of the feared situations. To reduce avoidance of activity in chronic pain patients, it would be necessary for the individual to reappraise their beliefs about

pain and (re)injury. However, in practice, activity may exacerbate the pain initially. Therefore, the individual may have to endure a period of increased pain in order to experience the benefits of increased fitness and muscle strength and hence reduced pain in the longer term. This immediate increase in pain may cause anxiety and increased fear of pain, confirming the belief that activity and use of the body is harmful or dangerous or will increase pain. Indeed, the habituation model of anxiety (Lader & Wing, 1966) implies that decreases in anxiety will only occur after prolonged exposure and relatively brief exposure periods may actually serve to 'sensitise' patients to their feared stimuli and prove detrimental (for example Marshall, 1985), particularly as avoidance prevents prolonged exposure. Even if the pain that follows the activity is not as bad as anticipated, this does not lead to a generalised adaptation of predictions about pain. For example Crombez et al. (2002) found that chronic pain patients' overpredictions about pain as a result of a particular movement were readily corrected by exposure. However, this correction did not extend to different movements, for which overpredictions continued to be made. This further illustrates the difficulty in attempting to apply models of fear and exposure to fear and avoidance in chronic pain.

This may highlight a difference between fear and avoidance in chronic pain compared with other anxiety disorders. In anxiety disorders, such as phobia, panic and social anxiety, cognitive behavioural interventions assume that exposure will provide an opportunity for disconfirmation of negative beliefs, and this will occur because the feared outcome will not take place. However, if a spider phobic were exposed to a spider, which proceeded to crawl rapidly up his/her arm, the model would predict that the negative belief would be confirmed, leading to an increase in both anxiety and avoidance. In chronic pain, Vlaeyen and Linton's model assumes

that the path to recovery involves no catastrophising, no fear and confrontation of daily activities. However, if the individual who fears pain, engages in graded exposure to activity and consequently experiences an increase in pain, this individual could be expected to experience an increase in fear and avoidance of activity. In the case of phobia, panic and social anxiety, it is assumed that the feared outcome is very unlikely to occur, however, in the case of chronic pain, it is possible that activity will result in pain, particularly after prolonged disuse. The increase in pain is likely to have an immediate effect on the individual's well-being, even if they escape from or avoid activity. However, (as mentioned above) patients with chronic pain fear not only movement and (re)injury, but report multiple fears, such as the fear of disability, the fear of altered identity, fear of physical illness and fear of social activities (Morley et al., 2004).

Mowrer revised his two stage model of fear and avoidance (Mowrer, 1939; Mowrer, 1960) to distinguish between between danger signals and safety signals. A safety signal was defined as a behaviour or strategy that enhances a patient's sense of safety and enables the patient to participate in activities that, without the safety signal, would be avoided. A danger signal provided an indication of threat or harm. Mowrer argued that conditioned stimuli associated with painful experiences could "take on" danger signals and that conditioned stimuli associated with pleasant experiences could "take on" safety signals, and both signals had motivating qualities. Hence for pain, if proximity to assistance enhances a person's sense of safety, it could "take on" a safety signal, which could lead to the person limiting the extent to which they venture out away from proximity to assistance (for example distance from their home). Likewise, in pain, if exercising or activity is a painful experience, that activity could "take on" a danger signal, and hence be avoided, which would be

a logical action. In simple phobia too, if a person perceives something as threatening it will be regarded as dangerous and hence avoided.

Similarly, evidence was found in a study of 147 patients with panic disorder that they were taking logical action to avert their feared outcomes and the safety seeking behaviour they adopted was meaningfully related to the threats they perceived (Salkovskis, Clark, & Gelder, 1996). In this study, safety seeking behaviour was referred to as safety behaviour. In a similar vein, it could be argued that if patients believe that activity is likely to lead to injury or pain, they will be more likely to avoid that activity, or take action to do it differently, which may have a deleterious effect on their pain and physical condition in the long run, but at the moment of making the decision, it is rational, based on their beliefs, and the responses they are receiving from their body.

The assumptions underlying work avoidance in chronic pain and avoidance behaviour in phobic anxiety are the same (Ciccone et al., 2001), that is outcome expectancies are modified when feared consequences fail to occur during exposure. Ciccone et al. (2001) suggested that treatment programmes enabling the chronic pain patient to perform increasing amounts of strenuous work activity are an equivalent to in vivo exposure for phobic anxiety. Fear-avoidance models predict that those patients who habitually overpredict pain or injury and hence become excessively inactive should benefit most. However, not all pain expectancies are subject to empirical falsification and some patients may be motivated by factors other than the avoidance of work related pain, for example those who avoid work in the pursuit of increased social reward (Ciccone et al., 2001). Hence Ciccone et al. (2001) concluded that the fear-avoidance model provides only a partial explanation for work disability in chronic pain (Ciccone et al., 2001).

Hence in the following section, evidence for other factors influencing pain outcomes is reviewed.

Factors influencing pain outcomes

Psychological factors related to the adjustment to persistent pain can be grouped into those associated with decreased pain, decreased psychological distress, and decreased physical disability such as self-efficacy, pain coping strategies, readiness to change and acceptance; and those associated with increased pain, increased psychological distress, and increased physical disability, such as pain catastrophising, pain-related anxiety and fear, and helplessness (Keefe, Rumble, Scipio, Giordano, & Perri, 2004). Aldrich, Eccleston, and Crombez (2000) represented chronic pain as chronic vigilance to threat that may lead to a perseveration of attempts at solving the problem of achieving escape from pain (Aldrich et al., 2000). People make repeated attempts to reduce the widespread and negative impact of chronic pain on their lives by seeking lasting escape, largely by the avoidance of pain-inducing activity, or by the use of analgesic agents (Davies, Crombie, Macrae, & Rogers, 1992).

In order to understand how well the existing models of fear and avoidance describe the progression from acute to chronic pain, and to activity limitation and disability, it is necessary to consider the evidence for the various factors influencing this process.

Role of fear in predicting avoidance and disability

The specific psychosocial factors most associated with the transition from acute to chronic lower back pain remain uncertain (Fritz, George, & Delitto, 2001).

However, it is possible that fear-avoidance beliefs may develop at an early stage of lower back pain and may facilitate early identification of patients at risk for chronic disability (Fritz et al., 2001). In a study of patients with low back pain that looked at the relationship between fear-avoidance beliefs and current and future measures of disability and work loss, Fritz et al. (2001) found that fear-avoidance beliefs did not explain a significant amount of the variability in initial disability levels after controlling for pain intensity and physical impairment. However, fear-avoidance beliefs did significantly predict disability and work status 4 weeks later even after controlling for initial levels of pain intensity, physical impairment and disability.

Based on these results, Fritz et al. (2001) argue that whilst it has been proposed that avoidance behaviours in response to pain may be adaptive in the acute phase of an injury, helping an individual to avoid situations that might increase tissue damage and nociceptive input, and only become maladaptive in the chronic stage, the results of their study contradict this. That is, higher levels of initial fear-avoidance beliefs did not offer any protective benefits, but were related to more persistent disability and difficulty returning to work (Fritz et al., 2001). However, the chronic stage in this study was merely 4 weeks after the initial measures were taken, and may not be a true representation of the chronic phase, as there is still scope for change to take place. In contrast to this, Walsh and Radcliffe (2002) suggested that pain beliefs that are unhelpful in chronic low back pain may be helpful in the acute phase where they may lead to an appropriate search for a medical cure.

Crombez, Vlaeyen, Heuts and Lysens (1999) posed the question "...what exactly do patients with pain-related fear, fear?" This question was taken up by Ciccone et al., (2001) who investigated why some patients with acute injury or illness are able to return to work, whilst others with similar symptoms are not, and become disabled. In

an attempt to elucidate Crombez et al.'s question, Ciccone and colleagues looked specifically at measures of pain expectancy and injury expectancy, and found that pain expectancy accounted for a larger part of the variance in work disability in both chronic and acute pain patients (Ciccone et al., 2001). They suggested that when pain symptoms persist despite medical intervention, patients may come to adopt a hopeless-helpless attitude which distorts their expectation of pain, and in so doing, their level of fear and avoidance. Alternatively, fear and avoidance may represent a stable personality trait that exists well before the onset of acute injury (Ciccone et al., 2001).

Fear of movement (measured by the Tampa Scale for Kinesiophobia (TSK)) and a rating of baseline neck pain disability within a week of trauma, can be used to predict chronic disability after 6 months, in patients suffering from whiplash injury (Nederhand, IJzerman, Hermens, Turk, & Zilvold, 2004). In a study with 33 low back pain patients, Vlaeyen et al. (1995) found that physical pathology was not predictive of disability, whereas pain-related fear was. Peters, Vlaeyen, and Weber (2005) examined the contribution of physical pathology, pain-related fear and catastrophising cognitions to pain intensity and disability in 100 patients with non-specific chronic low back pain. The strongest predictors of disability were found to be pain intensity (predicted 17% of the variance) and fear of movement (measured by the TSK, predicted 4% of the variance). Fear of movement and getting (re)injured may have mainly behavioural consequences, namely avoidance of movement to prevent the occurrence of harmful consequences to the body, finally leading to a state of disuse and increased disability. On the other hand, pain related fear (measured by the Pain Anxiety Symptoms Scale (PASS)) as well as age and physiological pathology predicted pain intensity, which may indicate that catastrophising about

pain and fear of pain lead to a preoccupation with pain and a heightened awareness of pain signals, thereby directly increasing pain perception (Peters et al., 2005).

Catastrophising

Catastrophising has been found to be a potent predictor of pain intensity, disability, and psychological distress, even when controlling for physical impairment (Severijns, Vlaeyen, van den Hout, & Weber, 2001). Severijns, van den Hout, Vlaeyen, and Picavet (2002) demonstrated that there was no relation between physical impairment and catastrophising, and found that pain catastrophising was significantly related to a number of negative outcomes including greater limitations in social activities, and lower energy level.

In a community study of 230 individuals with spinal cord injury, Turner, Jensen, Warm, and Cardenas (2002) found that greater catastrophising was associated with greater pain intensity, greater psychological distress, greater pain interference with activities and greater pain-related disability. Even after controlling for pain intensity, catastrophising was associated significantly with both psychological distress and pain interference with activities. Less use of coping self-statements and of ignoring pain was associated with greater psychological distress. Although cause-effect relations cannot be determined in this study, these findings are consistent with the view that catastrophising may contribute to increased psychological and functional disability in individuals with chronic pain. Catastrophising has been associated with poor functional outcome (Walsh et al., 2002).

Beliefs about pain and activity limitation

Goubert et al. (2004) found that pain-free individuals in the general population tend to hold a biomedical view of pain and have unrealistically high expectations about medical diagnosis and cure. They also held more misconceptions about back pain than individuals with mild back pain. Individuals with high pain levels had more misconceptions about back pain than both pain-free individuals, and than those individuals with mild pain but no disability. Examples of misconceptions about back pain include the belief that back pain is related to bodily injury, that an incorrect movement can lead to serious problems, and that back pain means one should reduce physical activity (Goubert et al., 2004). These findings support the idea that misconceptions about back pain play a prominent role in the development of chronic pain problems, and are not confined to a small group of extremely disabled back pain sufferers, but are widely held in the general population. Indeed, Linton, Vlaeyen, and Ostelo (2002) surveyed general practitioners and physical therapists regarding their fear-avoidance beliefs, and found that more than two thirds of the practitioners reported that they would recommend that a patient avoid painful movements. Hence the ability to resume daily activities after an acute episode of back pain may depend on the ability to correct these misconceptions.

A population based intervention involving provision of explicit advice about back pain was found to positively alter beliefs in the general population and positively influence knowledge and attitudes amongst doctors, and resulted in a 15% reduction in the number of claims for back problems over the duration of the campaign (Buchbinder, Jolley, & Wyatt, 2001).

Walsh et al. (2002) suggested that the direct influence of beliefs on behaviour and disability could be seen in the finding that patients who perceive themselves as more

disabled display submaximal effort and achieve lower levels of function. They also found that beliefs that one is by necessity disabled by pain, that pain signifies harm, or that one has little personal control over pain not only are associated with disability in cross-sectional studies, but also that changes in these beliefs are associated with changes in reported disability.

Distress and depression

Emotional distress generally corresponds to the presence of symptoms of anxiety, depression, and anger. In a review of prospective studies on the biopsychosocial factors predictive of nonreturn to work due to low back pain, Truchon et al. (2000) found that emotional distress measured in the first 12 weeks after onset of a lower back pain episode was not able to predict chronic disability. However, distress, and more specifically depression, could play a role in chronic disability due to a phenomenon of circularity by which persistent pain produces distress, which in turn produces inactivity and disability, which have an impact on the pain felt, distress, chronic disability, and so on (Truchon et al., 2000).

Depression but not baseline personality traits, nor the diagnosis of a personality disorder, was found to be an important predictor of disability in chronic pain patients, with a follow up of at least 2.5 years (Ericsson et al., 2002). Currie and colleagues examined the relationship between chronic back pain and major depression using a large epidemiological data set, and found that the combination of depression and chronic back pain was associated with greater socioeconomic disadvantage and disability than having either condition alone (Currie & Wang, 2004). It is possible that the presence of depression may contribute further to the activity limitation resulting from chronic pain, both conditions perhaps fuelling each other.

Self-efficacy

Self-efficacy, one's confidence in performing a particular behaviour and in overcoming barriers to that behaviour, is believed to be an important mediator of disability related to pain (Denison et al., 2004). In a test of a path analytic model with self-efficacy as a mediator of disability, low self-efficacy was found to be an important variable contributing to the disability of chronic pain patients, and it could help explain the circumstances under which disability develops more in some chronic pain patients than in others (Arnstein, 2000). When prediction of disability by both self-efficacy and fear-avoidance was examined simultaneously, self-efficacy was found to be the more powerful predictor (Ayre & Tyson, 2001). In a prospective study of two similar samples of primary health care patients with subacute, chronic or recurring musculoskeletal pain, Denison et al. (2004) found that self-efficacy explained a considerably larger proportion of the variance in disability scores than the fear-avoidance variables in both samples, whilst pain intensity explained a small, but significant proportion of the variance in disability scores in one sample only. These findings confirmed the authors' hypothesis that self-efficacy is a better predictor of disability than fear-avoidance variables and pain intensity in a primary health care sample of patients with subacute, chronic or recurring musculoskeletal pain, and that pain-related beliefs, such as self-efficacy and fear avoidance, in turn, are more important determinants of disability than pain intensity and pain duration in these patients. In fact, self-efficacy showed higher correlations with disability than both pain catastrophising and kinesiophobia, and self-efficacy was significantly negatively correlated with both these variables (Denison et al., 2004).

Barry, Guo, Kerns, Duong, and Reid (2003) examined the relationship between functional self-efficacy and pain-related disability in a sample of veterans with chronic pain, aged 65 years or older. Functional self-efficacy was measured using a ten-item questionnaire, and categorised as either low, moderate or high self-efficacy. Pain-related disability was defined as having one or more days of restricted activity due to pain in the previous month. They found that disability was significantly more likely for those with low or moderate self-efficacy scores, than for those with high self-efficacy scores. Whilst this provides further support for the inverse relationship between self-efficacy and pain-related disability, to more clearly establish the causal role of self-efficacy it would be necessary to carry out prospective studies in which self-efficacy is measured before the onset of pain, to ensure that self-efficacy scores are not confounded by pain-related factors which become effective only after the onset of pain.

Locus of control is said to be internal when the individual feels that he/she can have an impact on his/her health problems and health outcomes. In a study of the predictors of readiness to self-manage pain, amongst chronic pain patients, a low internal locus of control was found to predict a low intention to self-manage pain. However, the same study found that self-efficacy did not uniquely predict an intention to self-manage pain (Hadjistavropoulos & Shymkiw, 2007).

Pain coping and social resources

Individuals with pain use a variety of cognitive and behavioural coping strategies (Mercado, Carroll, Cassidy, & Cote, 2005). Besides the cognitive-behavioural factors set out in Vlaeyen and Linton's model, social resources, including social networks and perceived support from others, are assumed to have an impact on long-

term chronic pain outcomes, which may inhibit avoidance of physical and social activities and have a beneficial impact on functional disability and pain (Keefe, Smith, Gibson, Studts, & Caldwell, 2002). There is also increasing evidence that perceived social support and the size of social networks, affect future functional limitations and pain in chronic pain patients. For example, lower levels of perceived support have been shown to be prospectively related to more interference in daily activities in rheumatoid arthritis patients after one year (Smith & Wallston, 1992) and increased pain after one year (Waltz, Kriegel, & van't Pad Bosch, 1998), while less extended social networks predicted functional disability after one year.

Evers, Kraaimaat, Geenen, Jacobs, and Bijlsma (2003) looked at both pain coping and social resources in relation to the long-term outcome of functional disability and pain in early rheumatoid arthritis. The use of pain coping strategies, such as catastrophic cognitions about pain and resting and retreating, at the time of diagnosis was significantly related to an increase in functional disability after three years, but not after five years. Perceived support and social networks were significantly related to less increase in functional disability at the three and the five-year follow-ups, irrespective of the personality characteristics of neuroticism and extraversion, clinical status and use of medication. Higher levels of functional disability and lower levels of perceived support at the time of diagnosis were related to an increase in pain at the three and five-year follow ups. The favourable effects of social support on long-term outcomes might result from less withdrawal from social activities, the stimulating effects of participation in social activities, inhibiting avoidance behaviour and offering assistance in coping. In addition, altered autonomic and muscular reactivity or immunological function may be responsible for the favourable effects of social support. Although social support appears initially to have only marginal effects, its

influence increases on long term functional disability and pain outcomes, suggesting that the role of social resources may have been largely underestimated in chronic pain research (Evers et al., 2003).

Pain intensity

In a study examining the contribution of physical pathology (from medical charts), and self-reported pain-related fear and catastrophising cognitions, to pain intensity and disability in 100 patients with chronic non-specific low back pain, it was found that the strongest predictors of disability (measured as perceived difficulty with simple physical activities) were found to be pain intensity (predicted 17% of the variance) and fear of movement (measured by the TSK, predicted 4% of the variance). Neither pain catastrophising nor pain-related fear significantly contributed to total explained variance (Peters et al., 2005).

In a survey of 118,533 household residents in Canada, Currie and Wang (2004) found that pain severity was a stronger predictor of disability than either major depression or the number of chronic health problems reported. Individuals with severe pain were also more likely to report days of total disability (staying in bed all or most of the day) even after controlling for demographic factors such as age. A possible explanation for this finding was that individuals with severe pain had more serious medical conditions (such as cancer). Individuals who reported their pain intensity as severe also reported a higher rate of major depression (Currie et al., 2004). Considering this finding with Ericsson et al.'s finding that depression was an important predictor of disability in chronic pain patients (Ericsson et al., 2002), suggests that depression and pain severity could interact in their prediction of disability.

A prior history of lower back pain, marked by episodes sufficiently severe to have resulted in sick leave or consultation with a physician appears crucial in predicting chronic disability (Truchon et al., 2000).

The effect of compensation on pain-related disability

McDermid et al., (2002) collected baseline measures of age, sex, education level, injury compensation, and injury severity in 120 patients with distal radius fractures. Six months later self-reported measures of pain and disability were obtained and regression analyses revealed that the most influential predictor of pain and disability at 6 months was injury compensation. Patients with injury compensation reported more than twice the pain and disability as those who were not on either Worker's Compensation or involved in legal action. Furthermore, the impact of injury compensation on pain and disability exceeded injury severity as an explanatory variable (MacDermid, Donner, Richards, & Roth, 2002).

Similarly, a study by Carragee et al. (2005) found that prior worker's compensation or personal injury claims predicted future disability for lower back pain and health care usage (Carragee, Alamin, Miller, & Carragee, 2005). In addition, a current claim for lower back pain problems and past disputed claims strongly predicted longer disability, fewer remissions and greater health-care utilization (Carragee et al., 2005).

Secondary gain has been equated with conscious malingering. However the clinical assumption that follows from this, that once financial claims are successfully resolved, the alleged illness improves, has not been confirmed by research (Gatchel, Adams, Polatin, & Kishino, 2002).

This evidence suggests that compensation interacts with other variables, perhaps amplifying their effect. For example, involvement in a compensation claim, may increase anxiety or depression or both, or may increase disuse, all of which could influence the extent to which the individual is disabled by their pain. However, it could also be an indication that individuals with greater disability are more likely to make claims for compensation.

Education and occupation

The relationship between education and health has been observed in many countries and time periods, and for a wide variety of health measures. However, work on the mechanisms underlying the link between health and education has not been conclusive. Not all relevant theories have been tested, and when they have, studies will often conflict with each other (Cutler & Lleras-Muney, 2006). It has been suggested that education has a cumulative effect on health outcomes on several levels including socio-economic, such as work and income, behavioural, such as health behaviours like exercising and psychological, such as perception of control over one's life and health (Mirowsky & Ross, 2005).

A significant relationship has been found between lower educational level and an increase in pain five years after onset, but not three years after onset (Evers et al., 2003). In patients with chronic pain as a result of distal radius fractures, MacDermid et al., (2002) found that education level was a significant predictor of pain and disability after 6 months. They suggested that as patients with lower levels of education tend to be employed in jobs that require more manual labour, the effect of education may be masking an underlying effect of occupation on outcomes. In addition, more educated workers may be able to reassign elements of their job that

are physically stressing or relocate to new jobs more easily (MacDermid et al., 2002).

However, the relationship between job demands and symptoms or injury rates is inconsistent. In general there is little evidence that physical load in modern work causes permanent damage. The development of chronic pain and disability depends more on individual and work-related psychosocial issues than on physical or clinical features (Waddell & Burton, 2001). In a review of low back pain at work, Waddell et al., (2001) found that people with physically or psychologically demanding jobs may have more difficulty working when they have lower back pain, and so lose more time from work, but that may be the effect rather than the cause of their lower back pain.

Education may also affect outcome through an effect on compliance with rehabilitation: those with more education may be more compliant with rehabilitation or home programmes (MacDermid et al., 2002). However, it is recognized that there may be other explanations for the relationship between education levels and pain and disability.

Conclusions and directions for future research

There appear to be many factors predictive of disability in chronic pain, including the individual's cognitive appraisal of pain (fear of pain, catastrophising, beliefs about pain, distress and depression), their approach to coping (self-efficacy and pain coping), psychosocial and environmental factors (social resources, compensation and secondary gain, education, history of pain, attitude to work), as well as the physiological response. Whilst there is a lot of evidence supporting Vlaeyen and Linton's model, this model does not appear to provide a comprehensive description

of why pain persists in the absence of identifiable organic pathology, or why some individuals and not others develop chronic pain, limit their activity and become disabled, as it cannot account for all the factors which influence pain outcomes. These include factors such as self-efficacy, which in some studies have been shown to be more strongly associated with disability than fear-avoidance (Denison et al., 2004). Furthermore, Vlaeyen and Linton's model predicts that in vivo graded exposure will lead to a reduction in avoidance. However, not all pain expectancies are subject to empirical falsification and some patients are motivated by factors other than avoidance of work related pain (Ciccone et al., 2001). In the context of the wide range of factors influencing pain outcomes, activity limitation and disability and the fact that these may interact with each other, and be present to differing degrees in individuals suffering chronic pain, it would seem that Vlaeyen and Linton's model may oversimplify the process involved in activity limitation and disability. Therefore, the use of models such as Klinger's Current Concerns model (Klinger, 1996) may allow a broader spectrum of variables to be taken into consideration in the formulation of an individual's progression towards activity limitation and disability.

Whilst (self-reported) avoidance is associated with greater disability (Vlaeyen & Linton, 2000), these findings are always at a rather broad level and do not give any indication as to how specific a decision it is to avoid an activity; how consistent within and across activities; and to what extent avoidance is associated with high estimates of risk of immediate pain or damage rather than with moderate estimates of a major risk, since the former is more easily disconfirmed. Therefore future studies should investigate the process of decisions around avoidance of activities and activity limitation.

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Part 2: Empirical Paper

Activity Limitation in Joint Hypermobility Syndrome

Abstract

Joint Hypermobility Syndrome (JHS) is characterized by joint hypermobility, cutaneous fragility, hyperextensibility and a predisposition to everyday trauma. People with JHS experience chronic pain and tend to live restricted lives. This study focused on activity limitation and the process of decisions about limitation of activities, in order to elucidate the applicability of Vlaeyen and Linton's (2000) fear-avoidance model to people with JHS. A qualitative method was used to examine the process of making decisions about activity limitation of 11 women with JHS attending a pain management clinic. Semi-structured interviews were conducted, and analysed using Interpretative Phenomenological Analysis, which revealed 5 themes in the domain "Impact on activities" and 10 themes in the domain "Process of decision making about activity limitation". The women's accounts revealed a complex process in which each decision was individually considered in a cost-benefit analysis, weighing the importance of the activity against its potential aversive consequences, which is not adequately described by Vlaeyen and Linton's (2000) model. Suggestions were made for applying these findings in clinical settings with patients with JHS, and the need for further research was highlighted.

Introduction

Joint Hypermobility Syndrome (JHS) is a disorder of the musculoskeletal system, causing chronic pain, and not widely recognised by consultant rheumatologists (Grahame & Bird, 2001).

Pain is defined as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage” (International Association for the Study of Pain, 1979). It is a subjective experience and is primarily measured subjectively, not estimated by signs of damage or disease. Chronic or persistent pain is defined by timescale – pain which has lasted more than three months – and implicitly as beyond the time required for healing or resolution of lesions or pathology responsible for pain onset, and the term chronic pain is usually reserved for pain not attributable to a life-shortening or progressive disease (Williams, 2007). Chronic pain interrupts behaviour, interferes with functioning, and may affect a person’s identity: their sense of who they are and what they might become (Harris, Morley, Stephen, & Barton, 2003).

Joint Hypermobility Syndrome

JHS is thought to be a subtype of Ehlers-Danlos Syndrome (EDS), a heterogeneous group of inherited disorders characterized by joint hypermobility, cutaneous fragility, and hyperextensibility. These disorders affect connective tissue proteins such as collagen, which give the body its intrinsic toughness. When these proteins are differently formed, the result is joint laxity with hypermobility and a vulnerability to the effects of injury, for example dislocation. Pain can dominate the lives of people with JHS, in particular, chronic pain in joints, muscles and ligaments, which arises from an inherent predisposition to the effects of everyday trauma, but

other factors such as associated osteoarthritis or fibromyalgia are also important (Grahame, 2000). JHS patients may experience pain for no visibly obvious reasons, which contributes to the characteristic difficulty in diagnosis because patients may present looking well, and because presentation varies from patient to patient (Grahame, 2000). They report feeling that they have no control over their lives, which is exacerbated because there is not always a clear cause and effect relationship between an activity or movement and the onset of pain. Hence all their activities can become linked with pain (Gurley-Green, 2001).

Fear and avoidance in chronic pain

Psychological factors related to the adjustment to persistent pain can be grouped into those associated with decreased pain, decreased psychological distress, and decreased physical disability such as self-efficacy, pain coping strategies, readiness to change and acceptance; and those associated with increased pain, increased psychological distress, and increased physical disability, such as pain catastrophizing, pain-related anxiety and fear, and helplessness (Keefe, Rumble, Scipio, Giordano, & Perri, 2004). It has been suggested that the fear of pain and (re)injury may be more debilitating than pain itself, and that this refutes the early notion that the lowered ability to accomplish tasks of daily living in chronic pain patients is straightforwardly a consequence of pain severity (Vlaeyen, Kole-Snijders, Rotteveel, Ruesink, & Heuts, 1995). Possible mechanisms may be a misinterpretation of bodily symptoms, inaccurate predictions about pain, and hypervigilance for pain related symptoms and information.

Vlaeyen and Linton's (2000) fear-avoidance model is based on the notion that fear of experiencing pain prompts avoidance of daily activities which in turn maintains

fear of pain (Vlaeyen & Linton, 2000). This is consistent with clinical psychology literature pertaining to other disorders, in which attention towards threat is associated with increased negative thoughts and perception of threat (Clark et al., 1997) and with avoidance of, or withdrawal from, threatening situations.

The behaviours and mental processes used in an attempt to reduce, avoid, escape or alleviate threat or fear have been referred to as safety behaviours (Salkovskis, 1989). Safety behaviours contribute to the persistence of disorders by preventing disconfirmation of unhelpful beliefs and increasing the risk of the feared outcome actually occurring. It can be difficult to draw a distinction between escape and avoidance on one hand and adaptive coping strategies on the other (Thwaites & Freeston, 2005). The same behaviour could function, for any given person, both as an adaptive coping strategy and as a safety behaviour, but to differing degrees and in different contexts (Thwaites et al., 2005). For example, in the context of chronic pain, activity limitation could function as an adaptive coping strategy when it allows rest and recovery, but also as a safety behaviour which prevents disconfirmation of unhelpful beliefs about an activity. It has been found that pain expectancies intensify escape or avoidance tendencies, but do not amplify pain intensity, thus predicting an increase in hypervigilance and disability in the absence of an increase in pain intensity (Crombez, Vervaeke, Baeyens, Lysens, & Eelen, 1996).

Vlaeyen and Linton's (2000) fear-avoidance model of pain and the concepts of fear and avoidance have been very influential in the field of chronic pain, illustrated by the fact that, since 2000, their paper has been cited over 390 times², and over 1,400 papers have been published using these concepts³. This model proposed that when a catastrophic meaning is placed on an experience of pain, this leads to pain-

² Quoted in Google Scholar at the time of writing.

related fear (fear of pain, fear of (re)injury), which in turn spirals into a cycle of hypervigilance and fear-avoidance, which promotes and maintains activity limitations, disability and pain.

Chronic pain has been re-presented as chronic vigilance to threat that may lead to a perseveration of attempts at solving the problem of achieving escape from pain (Aldrich, Eccleston, & Crombez, 2000). People make repeated attempts to reduce the widespread and negative impact of chronic pain on their lives by seeking lasting escape, largely by the avoidance of pain-inducing activity, or by the use of analgesic agents (Davies, Crombie, Macrae, & Rogers, 1992). Vlaeyen and Linton's (2000) model is consistent with the notion that the likelihood of developing a chronic pain-related condition will be significantly elevated if the person has both a tendency to be hypervigilant towards internal pain sensations and a tendency to interpret those sensations as dangerous or potentially threatening to their well-being (Asmundson, Norton, & Vlaeyen, 2004). Severijns, van den Hout, Vlaeyen, and Picavet (2002) found that pain catastrophizing was significantly related to a number of negative outcomes including greater limitations in social activities, and lower energy levels. Swinkels-Meewisse, Roelofs, Oostendorp, and Vlaeyen (2003) found that patients with acute low back pain who reported a high fear of pain were significantly more likely to experience high levels of disability and to avoid participation in a range of home, work, social, and leisure activities.

While (self-reported) avoidance is associated with greater disability (Vlaeyen, de Jong, Geilen, Heuts, & van Breukelen, 2001), these findings are always at a rather broad level and do not give any indication as to how specific a decision it is to avoid an activity; how consistent within and across activities; and to what extent avoidance

³ Number of papers obtained by searching in Google Scholar for papers published from 2000, using "pain" and "fear-avoidance" as key words.

is associated with high estimates of risk of immediate pain or damage rather than with moderate estimates of a major risk, since the former is more easily disconfirmed.

Boersma and Linton (2005) found that the relationship between fear of movement and impairment of daily activity due to pain is moderated by the stage of chronicity, with fear of movement explaining variance where pain duration was between 1 and 4 years, but not below 1 year, suggesting that the time point in the development of a musculoskeletal pain problem might be an essential aspect of the importance of the relationship between psychological components and disability.

Linton, Vlaeyen, and Ostelo (2002) surveyed general practitioners and physical therapists regarding their fear-avoidance beliefs, and found that more than two thirds of the practitioners reported that they would recommend that a patient avoid painful movements. This suggests that patients are likely to get advice to avoid activity from authoritative sources, however, the influence of this on their decisions about activity limitation has been largely neglected by research.

Fear and avoidance in JHS

Relatively little research has been carried out to examine these processes in JHS patients, or whether this fear-avoidance model applies to this group. Whilst it could be expected to apply because JHS involves persistent pain, unlike many other forms of persistent pain, JHS often starts before adulthood and is a very specific diagnosis. Berglund, Nordstrom, and Lutzen (2000) conducted a qualitative study to explore how individuals with different symptoms of EDS described their symptoms and perceived their daily life. They found that "living a restricted life", seemed to explain the way in which fears, pain, stigmatisation and experiences of non-affirmation in

health-care limited the possibility of self-actualisation in daily living and social life. Anecdotal evidence based on clinical observations of JHS patients in pain management clinics highlights some of their beliefs which may contribute to their decisions about activity limitation. These include their lack of trust of the medical profession and the treatment they have been offered, their sense of actual damage and injury, the hereditary nature of the disorder, the global, all-encompassing nature of JHS, and their lack of control of JHS.

Aims of the present study

This study aimed to explore further what “living a restricted life” means to JHS patients, and focused in particular on what JHS patients avoid and why. The aim was to understand the process of decisions about avoidance to elucidate the applicability of Vlaeyen and Linton’s (2000) fear-avoidance model to JHS patients. A better understanding of this process will contribute to a better targeting of clinical interventions, will guide clinicians’ formulations when working with JHS patients, and may provide material for future empirical studies of avoidance in JHS. Only a detailed exploration of this sort can distinguish likely coping strategies from safety behaviours.

Given the lack of existing research from which research questions could be hypothesised and tested a qualitative approach was adopted. Qualitative research methods are particularly suited to research areas where there is little existing knowledge. Furthermore, data collection in these methods is not constrained by pre-existing hypotheses, and they allow the nature of individuals’ experiences to be examined in detail (Barker, Pistrang, & Elliot, 2002). The aim of qualitative research is to understand how people make sense of the world, how they experience events

and how they attribute meanings to events (Willig, 2001). Since the aim of this study was to understand the complex processes involved in decision making, a phenomenological approach was adopted, as these approaches attempt to understand people's perceptions of their experiences, the meanings they attach to their experiences and their underlying assumptions (Barker et al., 2002).

The study aimed to examine two research questions: (1) What impact does JHS have on the nature and level of the person's activities? and (2) How do people with JHS make decisions about activity limitation?

Method

Participants

Eligible participants were patients with JHS who had experienced pain lasting at least 6 months, were attending a pain management clinic, were not suffering with a serious psychiatric impairment or substance misuse/abuse problems or another general health condition which restricted activity, and were fluent in English.

Letters (see Appendix A) and Information Sheets (see Appendix B) about the research were sent to all patients with JHS currently on a waiting list to attend a pain management programme and who were attending the pain management clinics of two consultant rheumatologists in a central London hospital. A total of 33 patients, 1 man and 32 women, were sent letters. Sixteen (48%) indicated an interest in participating. Of these 16, 4 were unable to attend appointments, and 1 was not fluent in English and was excluded from the study.

Eleven women (33% of those invited) took part in the study. Their ages ranged from 22 to 55 years, with a mean age of 34 years. The ethnic background of the sample was 10 white and 1 Black British woman. The age of symptom onset ranged

from early childhood to 45 years, with a mean age of onset of 16 years. All but one were using medication for pain relief. Four participants had previously had some psychological intervention for their pain. Table 1. sets out the characteristics of individual participants.

Procedure

Ethical approval was obtained from the local research ethics committee (see Appendix C). The study was also approved by the University College London Hospitals Research and Development Directorate (see Appendix D).

A semi-structured interview took place in a private room, at either University College London, or at the hospital. The Information Sheet was reviewed with the participant, and any questions and concerns were discussed, before the participant was asked to sign the Consent Form (see Appendix E). Demographic information was collected using a brief questionnaire (see Appendix F), before beginning the semi-structured interview (see below). Interviews lasted approximately one and a half hours and were audio-recorded. At the end of each interview, participants were asked whether there was anything else about their experiences which they wished to discuss. After the interview, participants were asked to complete the Brief Pain Inventory (short form) (BPI) (see below).

Table 1
Participant characteristics

Ref	Age range	Age of symptom onset (years)	Age at diagnosis of JHS (years)	Highest level of education	In paid employment	Marital status	Children	BPI ¹ pain severity	BPI ¹ pain interference
P1	30-39	30	31	Post-graduate	No	Co-habiting	No	6.0	7.6
P2	20-29	14	18	College/University	Yes	Single	No	5.3	1.6
P3	20-29	7	22	School	Yes	Single	No	8.0	9.1
P4	30-39	14	34	School	No	Single	Yes	6.5	5.7
P5	20-29	9 months	20	College/University	No	Co-habiting	Yes	3.8	2.3
P6	40-49	6	39	School	No	Married	Yes	7.8	9.7
P7	30-39	12	28	School	Yes	Single	No	5.8	4.9
P8	50-59	45	53	College/University	No	Married	Yes	6.8	3.0
P9	50-59	30	49	Post-graduate	No	Single	No	2.8	5.1
P10	30-39	12	28	College/University	Yes	Co-habiting	Yes	7.5	6.9
P11	20-29	7	11	College/University	No	Single	No	6.3	9.0

¹Brief Pain Inventory (see below)

Semi-structured interview

A semi-structured interview (see Appendix G) was used which aimed to elicit the participants' perspectives on the extent of activity limitation in their lives and the process of decision-making about activity limitation.

The interview was semi-structured to allow participants to discuss their experiences from their own perspectives. The use of a semi-structured interview allows initial questions to be modified in the light of the participants' responses, and allows the researcher to probe interesting and important areas as they arise. The interviews aimed to elicit participants' perceptions of their experience of JHS and pain, the impact of JHS on their lives, and how they went about making decisions about activity limitations. During the design of the interview, the questions were discussed with a psychologist working on the pain management programme for patients with JHS. The interview schedule was used as a guide, and allowed the interviewer the freedom to focus on experiences which the participant chose to discuss. Smith and Osborn's (2003) guidelines on interviewing techniques were used, in particular, their advice that the interviewer's role is to facilitate and guide, not to dictate exactly how the interview proceeds.

The Brief Pain Inventory (short form) (BPI)

The BPI (Cleeland & Ryan, 1994) was used to assess participants' severity of pain and the impact of pain on their daily functions. This self-report questionnaire assesses pain severity and interference using a numeric rating scale (0–10). Cronbach alpha reliability of BPI data collected from non-cancer pain patients was found to be greater than 0.70, and comparable to that reported in the literature for cancer patients (Keller et al., 2004). The factor structure of the BPI was replicated in

non-cancer patients and the relationship of the BPI to generic measures of pain was strong. Support was found for the validity of the BPI as a measure of pain in patients without cancer (Keller et al., 2004). Participants' scores for pain severity on the BPI ranged from 2.8 to 8.0 (out of 10) with a mean of 6.0. Scores for pain interference on the BPI ranged from 1.6 to 9.7 (out of 10) with a mean of 5.9 (see Table 1.).

Analysis

The method of Interpretative Phenomenological Analysis (IPA) (Smith et al., 2003) was used to analyse the interview data. It aims to explore in detail how participants make sense of their personal and social world, focusing in particular on the meanings particular experiences, events and states hold for participants. It is phenomenological in that it is concerned with an individual's personal perception or account of an object or event, as opposed to attempting to produce an objective statement of the object or event itself. IPA explores the research participants' experience from their own perspective but recognises that this will necessarily be influenced by the researcher's own view of the world as well as the nature of the interaction between researcher and participant (Willig, 2001). Hence it recognises that phenomenological analysis produced by the researcher is always an interpretation of the participant's experience.

All interviews were transcribed verbatim. The method of analysis followed that set out by Smith et al. (2003). Interviews were examined on a case by case basis, looking in detail at the transcript of one interview before moving on to examine the next. In order to become as familiar as possible with the participant's account, each transcript was read several times, and statements which were interesting or

significant in relation to the research questions were annotated. These initial notes were transformed into a summary of the main ideas.

Similar ideas were then grouped into clusters, according to theoretical similarities, attempting to identify connections between them, and with particular reference to the research questions to which they pertained. These themes were checked against the transcript to ensure that the researcher's interpretation related to what the person actually said. This process was repeated for each transcript, the themes from earlier transcripts being used to inform the analysis of subsequent transcripts, whilst also allowing new themes to be identified. A final list of themes was constructed and organised into 2 domains which corresponded to the research questions. Thereafter, all transcripts were re-read, and a brief narrative synopsis which focused on the research questions was produced for each transcript. The list of themes was checked against each synopsis to ensure that they adequately captured the essential quality of what was found in each transcript. The stages of the analysis are illustrated in Appendix H.

A number of credibility checks were carried out (Elliott, Fischer, & Rennie, 1999). The analysis was discussed with the supervisors of this research at various stages, focusing on the nature of the themes identified and the best ways of representing them. One of the supervisors checked the results against transcripts to ensure that they accurately represented the data. Testimonial validity was assessed by checking the results with the original participants. At the time of writing, 4 participants had provided feedback. All 4 participants agreed with the findings, did not suggest any changes, and 2 of them expressed surprise at how similar the experience of the other participants was to their own.

When I began this research, I had no prior experience of working with patients suffering from chronic pain, which meant that my expectations about the outcome were influenced more by my reading of the literature than by personal experience. Before beginning the research, I observed one session of a pain management programme at the hospital at which the research was conducted, met some of the patients (none of whom took part in this study), and discussed the research with a psychologist working on the pain management programme. My theoretical orientation was a cognitive behavioural approach, which meant that I was particularly interested in participants' cognitions and emotions and their effects on behaviour.

Results

Analysis of the qualitative interview data yielded themes relating to 2 broad domains. The first domain focused on the impact on the nature and level of activities and formed the general context for the second domain. The second domain focused on the process of decision making about activity limitation. These themes and their clustering into domains is set out in Table 2.

The domains and themes are described below, and illustrated using quotations from participants' transcripts (the number following the quotation is the participant's identification number).

Table 2
Themes from the qualitative analysis

Domains	Themes
1. Impact on activities	<ul style="list-style-type: none">1.1. Limitation and restriction1.2. Changing how I do things1.3. The difficulties of travel1.4. Curtailment of social life1.5. Narrowing of roles within the family
2. Process of decision making about activity limitation	<ul style="list-style-type: none">2.1. Is it worth it?2.2. What I want2.3. Pacing and adapting2.4. Balancing the pain2.5. Thinking about the activity2.6. There's nothing to think about2.7. Struggling with unpredictability2.8. How I'm feeling2.9. Fear and anxiety2.10. Staying in control

Domain 1: Impact on activities

Participants described experiencing a wide range of limitations on physical activities, movements and positions. These limitations resulted either in giving up activities completely usually in the case of enjoyable pursuits such as sports, dancing, hiking, whilst in the case of necessary or essential activities, they resulted in

adopting a more gradual approach to accomplishing the activity. Travel was frequently noted as presenting difficulties, and hence being limited, and resulting in a knock-on effect of restrictions in the person's life, for example, in holidays, socialising, and working. Their relationships and social life were curtailed and their roles within their families became narrowed.

Theme 1.1: Limitation and restriction

Participants described feeling limited in what they could do, and restricted in their activities:

“I feel like I'm in a compacter, you know where the walls come in and crush. That's how I feel, I feel like it's come in and I can only do so much now, it's limited me so much.” [P4]

They frequently reported giving up physical activities which they previously enjoyed, and experiencing these as losses:

“I don't want to think negatively, I try to think positively but it's very limiting I cannot do everything I want to do, I cannot do the sport I want to do, I cannot do all the things I want to do in one day, I have to rest quite a lot, I cannot see my friends when I want, where I want, there's a lot of things I cannot do, and I have to deal with this, but not thinking too much about it.” [P1]

The awareness of the extent of their limitations led to a sense of frustration with themselves, which was often experienced as depressing and sad. Participants' accounts also reflected a loss of independence:

“I have to rely on people to take my children to school which I don't like doing. I have to rely on people to bring them home. I have to rely on

people in case they need collecting in case of anything. I have to rely on people to get me a bit of shopping.” [P4]

Many examples of physical activities were given in which participants were no longer able to participate including swimming, going to the gym, horse-riding, cycling, long distance running, skiing, ice skating, and housework. Difficulty adopting certain movements also restricted their activities, for example lifting the arms above the head restricted hanging up washing, drying hair, and reaching shelves and cupboards. Sitting in one position for a long time, or standing were frequently described as contributing to stiffness and pain and hence limiting activities involving those. Finding a comfortable sleeping position was a common difficulty. Day to day activities were also restricted by difficulties with certain movements and positions, and participants reported difficulties with lifting heavy objects, climbing steps, standing, writing, and using a computer. In many cases, these limitations had led to the individual limiting or stopping their work. As one participant put it:

“Well it’s basically an invisible sort of disease that’s taken over my life, it’s changed me, it’s like unreal you know, from day to day, um and it’s working so fast, generating all through my body, so fast, it’s restricted me from most things. My daily activities, my role as a mother, as a wife, my role as a person. The pain the tiredness the fatigue.” [P6]

Theme 1.2: Changing how I do things

In response to these restrictions, participants broke tasks down into smaller parts, which could be accomplished in a shorter space of time, would involve a shorter use of one group of muscles, or would make the task easier such as doing small amounts of shopping on a regular basis instead of doing it all in one go. Housework was often

described as being spread over the whole week instead of completed in one day as they would have preferred or as they may have done before the onset of chronic pain.

This was summarised by one participant:

“Probably not changes to what I would avoid or what I would do, but it made changes on how I would do them.” [P5]

Many participants either retrained in order to change their job to one which was more physically manageable, or reduced their working hours, or stopped working. Fatigue, a greater need for rest, and becoming tired more quickly as a result of activity also resulted in a need to limit the time for which an activity was done, or in doing things more slowly.

Theme 1.3: The difficulties of travel

The difficulties encountered in travelling were an important part of participants' lives:

“Uh, seeing my friends because I cannot see them as much as I want because living in London, it's a big city, and every time you want to see someone, you have to travel, and so just the travel already makes me tired” [P1]

Public transport presented difficulties in that they may not be guaranteed a seat, they were unlikely to be offered assistance if they fell, and if they did have a seat they would find it difficult sitting in one position for any length of time, particularly in air travel and driving. Where participants did drive, this would always be for a limited period of time to avoid sitting in one position for too long, and they invariably stated that an automatic car was a necessity. In planning a trip, they

would have to plan where they could stop for breaks both to enable them to shift position and to reduce fatigue.

Theme 1.4: Curtailment of social life

Difficulties with physical activities such as standing, walking and a need to be able to sit down when tired or in pain, and travelling, contributed to a curtailment of their social lives. They avoided activities in which they could not participate fully for example dancing. They reported finding it difficult to plan social engagements in advance due to the unpredictability of their pain. Their difficulties with travel led to a limitation of holidays which involved travelling abroad. Their embarrassment about using aids such as sticks or wheelchairs also led to avoidance of social activities. The emotional impact of depression, isolation and pain interfered with their social lives:

“You can’t go and sit in a cinema cos you’re always fidgeting, going out for a meal, fair enough for a little while, cos you have to move around and I’ve missed out on a lot, a lot of get togethers, I won’t go, with friends, with family.” [P6]

Theme 1.5: Narrowing of roles within the family

Participants who were parents described difficulty fulfilling their roles and taking care of their children in the way they would wish to:

“I can’t do a lot of things that other mums can do you know, like, god like, just going out for a day it’s just such a, it’s a hassle. It’s more hassle than it’s worth, cos I’m just aching by the time I get anywhere,

and then I'm moody and I take it out on her, and it's just, she doesn't really need that, you know" [P10]

They were unable to participate in all the activities their children wished them to, and sometimes roles were reversed in which children helped take care of their parents. Relationships with partners were also affected and the physical side of their relationships were restricted. In some cases relationships had broken down as a result of their condition.

Domain 2: Process of decision making about activity limitation

Participants' accounts suggested that when faced with the possibility of engaging in an activity, they adopted an approach of a cost-benefit analysis, in which they weighed up the importance of the activity against their estimation of the likelihood of aversive consequences occurring as a result of doing it. The central consideration was the intensity and duration of pain which it would cause, as well as the consequences of that pain, in particular, the steps which would need to be taken to alleviate the pain, which may involve bed-rest for an uncertain number of days, the activities which would be missed as a result of this, and the effect on people within their network of friends and family, who rely on them to be available.

Balanced against this was the importance which the participant attached to that activity. Activities which were considered necessary, urgent or to which the participant attached high personal value would be given greater weight when balanced against the likely consequences. For some participants this appeared to be an implicit process, which they might describe as "just knowing" because of the way their body would feel, whilst for others it appeared to be an explicit process, in which

the nature of the activity, its physical demands, risks for injury and the likely consequences were carefully considered.

However, in spite of all these factors which they may consider, at times, an overriding desire to do something which is of high personal value to them, would lead them to bypass the weighing of costs and benefits, and engage in the activity, with little regard for the consequences.

Theme 2.1: Is it worth it?

As described above, participants appeared to engage in a cost benefit analysis, weighing up the importance of the activity against the potential aversive consequences. In many cases participants were able to articulate explicitly their weighing of the importance of the activities:

“I have things, you know, different levels of importance. Me missing a meal isn’t important, but the dogs have to be sorted out, but the ironing doesn’t have to be done, it’s not important, and the world’s not going to end if I don’t do the ironing.” [P7]

Consideration of the consequences encompassed risk of pain, risk to their health, the impact on their ability to engage in other activities not just immediately but in the ensuing days, and the impact on other people. In many cases this was an explicit process:

“Something that is potentially high risk of dislocation then it’s just not worth doing it, because then you got to take someone’s time getting you to the hospital, so they’ve got to stop doing what they want to be doing, you got to waste someone’s time the next day looking after me and the baby. It’s just not worth it, so you just don’t do it.” [P5]

Participants also recalled previous experiences of engaging in an activity which may have had aversive emotional consequences:

“It was frightening, so I thought never ever again am I going to sit down in the bath.” [P8].

Participants’ accounts suggested that at times, this process led to conflicting considerations, for example when engaging in the activity could lead to pain and a need for rest, whilst not engaging in it could lead to feelings of guilt at letting others down.

Theme 2.2: What I want

Despite this pervasive theme of weighing activities against their consequences, participants frequently described how, at times, the importance to them of the activity would almost completely outweigh any consideration of its consequences, leading them to a rather liberating decision to engage in the activity and address the consequences later:

“I mean some things are worth it. If it’s something I really want to do, then I just do it. You think, I’m going to feel that tomorrow, but I want it.” [P5]

This seemed to suggest an implicit temporary denial of their condition and of the potential consequences, but with an outcome which they did not seem to regret.

“I know it’s going to affect me later but I have to forget about it in order to carry on with what I’m doing.” [P3]

Besides simply ignoring the consequences, participants also appeared to be prepared to find a way of working around their circumstances in order to achieve important goals:

“When I know that I want something very very much, I know the strategy of going about it.” [P9]

A frequently expressed desire was to be normal, and to be treated by others as such:

“The main thing I have about it is that I don’t want to be treated any differently from anyone else.” [P2]

Besides being treated normally, they also wanted to be perceived by others as normal:

“When I go out when I’m seen by other people, I’m trying to do things like the others so I try, I want people to see me like normal.” [P1]

Participants’ accounts also suggested that they felt that their choices about activities were influenced by their level of determination, their willingness to rise to challenges, and their resilience. They sometimes explicitly stated that this affected their decision making:

“I’d say there’s nothing that I would let it stop me, because that’s just the kind of person I am.” [P2]

“So I get around it. I will not give up, I will not give in. It’s two words that I can’t do.” [P8]

However, this seemingly positive attitude of reaching for goals irrespective of consequences, did not appear to be achievable for all participants, some of whom felt constrained by their physical limitations:

“I wanted to get out, I wanted to get out and I wanted to do things, and I couldn’t because my body would just not allow me because of the pain.” [P6]

Theme 2.3: Pacing and adapting

In order to accomplish their daily goals, participants frequently paced their activities to avoid excessive fatigue, permitting themselves to do things more slowly, or in a different way:

“So I can’t, I won’t be able to do something throughout, I have to sort of break it up into pieces and do it bit by bit by bit.” [P3]

At times, their accounts suggested not only practical adaptations, but a change in their underlying assumptions pertaining to how they lived their lives:

“So it’s got to the stage where it’s yes, it’s dusty, it’s going to get dusty and it can stay dusty. But that was never me.” [P4]

This process of pacing extended to preparing themselves for planned activities to maximize their chances of being physically able to engage in them:

“If I’ve got something planned, I’d sort of try to take it easy so that I can go.” [P5]

Some of their accounts seemed to suggest that as pain was a normal part of their existence, adapting to it was not allowing it to limit their activities, but rather a positive choice:

“I have to phrase it as things that I avoid in order to explain to other people, but in fact it’s how I choose it.” [P9]

Theme 2.4: Balancing the pain

Pain was the central feature in participants’ accounts of making decisions about activity. They invariably spoke of having an awareness of a level of pain (in terms of both intensity and duration) beyond which they should not allow their bodies to go,

as this would disturb their equilibrium, a fine balance between an acceptable level of daily activity and a tolerable, manageable level of pain.

“If I can keep my pain at a level where, you know, it’s manageable, I can get on with a certain amount of pain, that’s fine.” [P4]

This level seemed to be regarded as a tipping point, and pushing their bodies beyond it was considered likely to precipitate a spiral of pain, fatigue, disablement and negative emotional consequences:

“It’s like you get a level and you know this level if you do too much the pain is going to get really worse, and there’s a level where you know you can do things and it’s not going to be worse and you can carry on, there’s like a level you know is far enough and it’s going to get worse.” [P1]

In achieving this balance, it seemed that participants were regulating their level of activity, exercise and rest, in order to manage their pain. However, this sometimes posed conflicts:

“I feel better and I don’t feel better, it’s difficult to explain, it’s like you release the pain when you do cardio, but at the same time I feel that I create the pain.” [P1]

The previous theme “What I want” suggested that participants did feel able at times to push themselves beyond this tipping point. However, several participants conveyed reluctance about doing so, and on comparison of the transcripts, it seemed that these participants expressed a greater sense of loss than the others, in terms of what they were able to do, for example, they perceived their present condition to be deteriorating rapidly, or they feared returning to a previous state of distress and disablement:

“Sometimes when you hurt a lot you are scared that it won't go away. When I first did my back in it was horrible. I was so scared I was going to be like that, that was going to be it. Then you start thinking about all the things you can't do and you get really anxious.” [P11]

Theme 2.5: Thinking about the activity

It was frequently apparent that participants gave careful consideration to the nature of the activities. This sometimes entailed use of mental imagery of engaging in the activity, or explicit consideration of the demands of the activity:

“You look where it is, you look how you've got to get there and what you're going to be doing when you do get there. You split everything down into sections, how, where and what.” [P5]

The physical demands of the activity were considered, for example,

“I've got to think all the time, how far, how far am I going to walk, how long are we going to be out for.” [P4]

The physical environment in which the activity would be undertaken and the risks which it posed for injury were also often explicitly considered:

“Walking the dogs I have to be careful where I walk them, what I do, whether the ground's level, is it a route that I know, just because I have to be really aware of my surroundings.” [P7]

Theme 2.6: There's nothing to think about

In contrast to this careful consideration of the activity and the risks posed by it, participants' accounts suggested that on some occasions, they felt that they were simply responding to pain, not engaging in a cognitive process:

“There’s nothing to decide. Yeah, if it’s [the pain is] unbearable, I could do it, but why would I do something that would make me feel bad? Because the pain is telling me, no you’re not going to do it.” [P1]

Pain was sometimes regarded as a message from their bodies:

“The pain is the way of my body trying to tell me that I’m doing something wrong or I’m hurting something so I have to lessen it or stop. I suppose that’s what a pain signal is, it’s something that lets you know something’s wrong when you have to stop doing what it is.” [P3]

In these circumstances, their accounts seemed to suggest that their bodies were perceived as separate entities, and in control of their choices about activity:

“I avoid doing totally anything at the moment, cos anything will bring it on and it doesn’t allow me. It doesn’t *allow* me.” [P6]

Theme 2.7: Struggling with unpredictability

Whilst participants spoke in detail and with certainty about the nature of the activity and its importance to them, the consideration of the potential aversive consequences appeared to pose difficulties in the decision making process, due to their awareness of the unpredictability of pain and their body’s response to a given activity:

“I mean I could do something and there’ll be no repercussion that time and I could do something again and there will be a repercussion. There doesn’t seem to be any pattern.” [P7]

This unpredictability affected their decisions about future activities, in particular limiting their ability to plan in advance:

“I don’t know how I’m going to be in 2 or 3 days so I cannot, for example to see my friends, I cannot say, let’s organise in 3 days, because I don’t know how I’m going to be in 3 days. I might be in pain.” [P1]

It seemed that when participants were uncertain about the consequences of an activity, they relied upon their previous experience of that activity:

“I think it’s based on prior experience of knowing, knowing what will be required of my body.” [P9]

However, it seemed that this unpredictability made it difficult for some participants to establish cause and effect relationships between certain activities and their levels of pain, adding to the complexity of this decision making process:

“I know I have to do it, I’m going to do it, but I always wonder if I’m going to be able, and I know I’m going to be able, but in, but how much pain am I going to get out of it, I’m going to go through, and how many days problems I’m going to have after.” [P1]

Theme 2.8: How I’m feeling

When faced with deciding whether to engage in an activity, participants’ perception of their pain at that time appeared to be an important influence on their decision. They frequently referred to having “good and bad days” and it seemed that their level of activity was related to how they were feeling at that time. They seemed to suggest that when their experience of pain was at either extreme of the spectrum, the decision making process became easier. For example, when pain levels were high:

“Well it’s easy when I’m flaring because there’s no choice, because the pain’s so bad, I just can’t do it.” [P4]

Similarly, when pain levels were low:

“I would feel there is nothing wrong with me on a good day. I can get up and do what I like. Just carry on, carry on as if there’s no tomorrow.”

[P8]

Although participants did not explicitly describe the process by which their current state affected their decision making, their accounts suggested that a high level of current pain indicated that they were closer to their tipping point (discussed above), and hence unless the activity was of considerable importance it would be unlikely to outweigh the costs of engaging in it. Their accounts also suggested that a high level of pain resulted in a higher level of fatigue and less energy, which would be likely to result in less inclination to engage in activity.

Theme 2.9: Fear and anxiety

Fear and anxiety were mentioned by several participants as affecting their decisions about activity. However, their accounts suggested that fear and anxiety related to specific activities, and were not pervasive driving forces underlying their decisions:

“I get really anxious before doing it because I know I will get a lot of pain, so I don’t go hiking any more.” [P1]

Four participants spoke of fearing pain: two of these participants also described their activities as very restricted, experiencing high levels of constant pain, and having experienced rapid deterioration in their physical conditions. One of them described herself as experiencing only “bad days, extra bad or double extra bad days.” [P6] It seemed that her fear of pain derived from the meaning she attached to the pain, in that it had resulted in her current state of disablement and distress.

The other two participants had recovered from a similar state, and their fear of pain appeared to derive from their perception that pain beyond their tipping points could result in a regression to that state:

“Yeah I’m always scared when I go back into big heavy pain because it always reminds me how I was before and I always get scared that I’ll get like that, back like that.” [P1]

Other participants’ anxiety seemed to relate to their perception of their capacity to cope in particular situations:

“I get a bit anxious. What if I pop [dislocate], or what if I fall. If people do help you, you’ve then got to try and explain to them, why you’re on the deck, why you’re on the floor in the first place.” [P5]

One participant explicitly described her insight into the effect of fear on her decision making:

“If I did have the fear now, one I wouldn’t be doing, if I had the fear, there would be no point in doing anything because if I fear the outcome then I wouldn’t do it.” [P3]

Theme 2.10: Staying in control

It appeared that part of participants’ process of considering the nature of the activity and addressing their uncertainty and concerns about the unpredictability of pain was ensuring that they felt that they were able to exercise control over their pain. This was apparent when they were considering specific activities, in their description of their distance from help, their ability to escape from the situation if necessary, and their ability to manage the pain:

“Yeah I mean if it’s something say round the corner or something short, where I know I can leave and go home and relieve the pain or something, then I do [it].” [P3]

This need for control also appeared to relate to the ability to exercise control over their level of pain in a more global sense, as opposed to in consideration of specific activities:

“I like to be able to be in control of what I do. It’s important to me. I don’t want to knock myself out and spend two days in bed and have the children come in and see me and go away thinking that mum’s really ill.”

[P8]

Discussion

Eleven participants with JHS were interviewed to explore how it limited their activity, and their process of decision making about activity limitation. Participants experienced a wide range of limitations on physical activities, movements and positions, which resulted either in giving up activities completely usually in the case of enjoyable pursuits or in finding ways of pacing or adapting themselves to achieve the same end, in the case of necessary or important activities. Participants’ descriptions of how they made decisions about activity limitation revealed a complex process, in which each decision was individually considered in a cost-benefit analysis, weighing the importance of the activity against its potential aversive consequences, in particular pain, which they felt was the most important factor which influenced their decisions. They appeared to be attempting to maintain a fine balance between an acceptable level of daily activity and a tolerable, manageable level of

pain. This is consistent with the description of pain dominating the lives of people with JHS (Grahame, 2000).

This decision making process was influenced by factors which would not be consistent across activities, or from day to day, such as the importance of the activity, their perception of their current level of pain and their estimate of their ability to cope, at the time of being faced with the decision. The risks and physical demands of the activity, and the likelihood of aversive consequences were weighed up, but on occasions this rational process gave way to participants deciding to place greater weight on what they wanted to do, in spite of potential aversive consequences. The unpredictability of pain, and the difficulty in relating physical consequences to specific activities, which concurs with previous literature (Gurley-Green, 2001), meant that under these circumstances, both the likelihood of the consequences occurring and the intensity and duration of the pain were uncertain, making the decision more difficult. Participants appeared to rely on their knowledge of the way their body had reacted on previous occasions as a guide to the likely consequences.

The findings of this study suggest that Vlaeyen et al.'s (1995) notion that the fear of pain and (re)injury may be more debilitating than pain itself, does not hold for JHS patients with chronic pain. In this group, fear of pain and injury was not reported across the sample, and even where participants did mention fear of pain or its consequences, this did not appear to be the driving force behind their day to day decisions about activity. When fear was mentioned, it related to the uncertainty caused by the unpredictability of pain and the difficulty controlling it, or the prospect of a deterioration in their condition giving rise to further losses. The cost-benefit analysis approach suggested that when the activity was perceived as very important, then either the potential aversive consequences were given much less weight in the

decision, and they engaged in the activity irrespective of the consequences, or ways would be found to accomplish the goal by adaptation or pacing. Furthermore, it was frequently reported that the current severity of pain was an important influence on the decision about a particular activity. This is also not consistent with Vlaeyen and Linton's (2000) model which suggests that the cognitive appraisal of pain drives behaviour, and does not allow for other factors such as current pain severity, and the appraisal of the importance of the activity to influence that cognitive appraisal.

The cost-benefit analysis approach was consistent with Crombez et al.'s (1996) finding that pain expectancies intensify escape and avoidance tendencies, in that participants considered the likelihood of pain as a consequence of the activity. However, it did not seem that there was a simple linear relationship between a high pain expectancy and an increased level of avoidance; rather the pain expectancy was weighed against the importance of the activity, and the benefits from engaging in the activity, in the context of the level of pain being experienced at the time of making the decision, as well as the consideration of any opportunities for adaptation or pacing.

Underpinning Vlaeyen and Linton's (2000) fear-avoidance model is the notion that placing a catastrophic meaning on an experience of pain is pivotal in determining whether pain-related fear and hence avoidance occurs. Whereas "catastrophic" implies an exaggerated expectation of imminent disaster, the participants in this study appeared to base their estimates of consequences largely on their previous experiences of that activity. It did appear that where they had experienced rapid deterioration or a state of distress and disablement, they were likely to be more cautious in attempting to maintain their equilibrium between an acceptable level of activity and a tolerable level of pain. However, these findings did

not appear to offer support for catastrophic thinking as a pivotal element of decisions about activity limitation.

Asmundson et al. (2004) proposed that the likelihood of developing a chronic pain-related condition would be significantly elevated if the person has a tendency to be hypervigilant to pain sensations and a tendency to interpret those sensations as dangerous or potentially threatening to their well-being. Consistent with this, there was a suggestion in participants' accounts that where the consequences were regarded as posing risk of serious harm or long term damage, the activity would be more likely to be avoided. However, Asmundson et al.'s notion does not allow for the counterbalance to this in which participants experienced conflict between what they wanted to do, and what they felt their bodies enabled them to do, or the fact that they had a desire to be normal, and sometimes to do things of high personal value irrespective of the consequences for their well-being.

Hypervigilance is an important element in the spiral towards activity limitation in Vlaeyen and Linton's (2000) model. Self-monitoring of their bodies' reactions to activities and pain levels, an awareness of the level of pain beyond which it was not worth pushing themselves and a tendency to regard pain as a message from their bodies, were indeed reported by the participants. However, it seemed that this was in an effort to maintain an adequate level of activity balanced against a tolerable level of pain, as opposed to a tendency towards ever increasing avoidance, as the Vlaeyen and Linton model would suggest.

This finding of participants' efforts to maintain an equilibrium, and the cost-benefit analysis of activities and consequences in order to achieve that, was also at odds with Aldrich et al.'s (2000) conceptualisation of chronic pain as a chronic vigilance to threat aimed at achieving lasting escape from pain. Participants'

accounts suggested that although they would have liked to have achieved lasting escape from pain, in practice, they were attempting to manage pain within a tolerable level. Aldrich et al's notion of chronic pain as a chronic vigilance to threat fails to capture the idea that besides their consideration of the risks posed by the activity and its likely aversive consequences, they also considered the importance of the activity and the benefit which they might have derived from it, sometimes to the extent that the threat it posed was disregarded.

Participants' attempts to manage their activity in order to balance an adequate level of activity with a manageable level of pain illustrated Thwaites et al.'s (2005) contention that it is difficult to distinguish between avoidance and adaptive coping strategies. When participants estimated that the likelihood of aversive consequences outweighed the likely benefit of the activity, they avoided or limited the activity. This may have been adaptive in some situations in which possible injury and several days of incapacity were prevented; however, this may in some cases have been unnecessary avoidance driven by the memory of past aversive experiences with that activity. The unpredictability of pain contributed to the difficulty for participants in identifying where this balance lay, between doing enough to stay healthy and avoiding enough to prevent aversive consequences.

In summary, the complexities of the decision making process which this sample of JHS patients appeared to engage in concerning activity limitation, cannot be adequately represented by Vlaeyen and Linton's (2000) fear-avoidance model. Although fear and anxiety influence this process, they do not appear to be the driving force behind it. The cost-benefit analysis of the activity and its consequences, focusing on each individual activity, may vary depending on the individual's severity of pain at that moment, how certain they are about their estimation of the costs and

benefits, as well as their recall of previous experiences of engaging in that activity and any aversive consequences which may have resulted, together with their estimate of the likelihood of recurrence. Furthermore, their decision making is driven by what they want, which at times may override any rational consideration of consequences.

This study revealed the complex nature of decision making undertaken by people suffering from chronic pain as a result of JHS. Although fear and anxiety did appear to influence decisions about activity, in a specific manner, they did not drive the decision making process as suggested by existing fear-avoidance models. The use of a qualitative method enabled participants to give their perspective on approaching activity limitation, which indicated levels of complexity and individuality not expressed in the existing literature.

These findings bear some relation to the general literature on decision making. In Expected Utility Theory, the consequences of each alternative are considered and the aim of decision making is to maximise the expected utility deriving from that outcome (von Neumann & Morgenstern, 1947). However, it was subsequently recognised that, just as in JHS, decision makers do not have complete information about consequences attached to each alternative, hence they choose a path that satisfies their most important needs, even though this may not be an optimal choice (Simon, 1956). The process of decision making undertaken by the participants in this study, is also consistent with Prospect Theory. This is a widely accepted alternative to Expected Utility Theory, in which it is assumed that gains and losses deriving from decisions, are balanced against each other and people are assumed to be loss averse (Kahneman & Tversky, 1979). A study of everyday decision making recognised that it is not always a rational process, but that individuals consider their own preferences, values and feelings (Marsiske & Margrett, 2006) and the findings

of the present study are consistent with this. A study of everyday self-care decision making in chronic illness (such as cancer and diabetes) suggested some similarities with the process described amongst JHS patients: decisions change over time and in various situations, can be influenced by disease severity, and the process entails a complex interaction among multiple and often conflicting goals and expectations of the decision maker, and arises from an authoritative knowledge which evolves from living with the illness over time (Paterson, Russell, & Thorne, 2001).

However, some limitations of the study need to be borne in mind in the interpretation of the findings. The majority of the participants indicated that they had come to the research interview although they felt sure they would suffer pain as a consequence: they felt it was important to participate in the research because of their strongly held belief that there was insufficient information on JHS available both to the public and amongst the medical profession. Therefore, in spite of the reasonable response rate to the recruitment process, it is possible that this self-selection of participants, combined with the small sample size used in qualitative research, could have resulted in a biased perspective on activity limitation being conveyed. In particular, the fact that participating in the study meant that they had to travel to the appointment, and sit still in one position for a considerable period, could have resulted in those JHS patients suffering higher levels of disablement, distress and activity limitation, not participating. Alternatively, perhaps those who chose not to participate, were limiting their activity very little, and hence felt they would have little to contribute to the study.

Another limitation derives from the fact that all those who took part in the study had a diagnosis of JHS and were attending a pain clinic, suggesting that they had a relative amount of certainty about their condition compared to those suffering the

same symptoms but without a diagnosis. It is possible that those without a diagnosis, and hence with a higher level of uncertainty about aversive consequences arising from activity, may be subject to slightly different influences on their decision making. There was insufficient evidence from this study to determine whether participants' decisions about activity limitation changed once they received their diagnosis.

There were no men in this sample, and little ethnic diversity, which is unrepresentative of the incidence of chronic pain in the population.

Finally, as mentioned above, IPA is an interactive process in which the researcher's own beliefs and preconceptions will influence the interpretative process. Hence it is possible that a different researcher, with a different orientation, may have reached different conclusions.

The findings of this study have some theoretical implications. The results suggest that for people with JHS suffering from chronic pain, fear-avoidance models do not adequately explain the cognitive or emotional process involved in decision making about activity limitation. While fear and anxiety can influence decisions, this does not appear to be universal, and is not the driving force behind the decisions. These findings suggest a multidimensional approach to decision making, not captured by the fear-avoidance models.

These findings also suggest some clinical applications. Although participants all described engaging in a cost-benefit analysis of activities and their consequences, they did not necessarily articulate the process in these terms. It may be of benefit to patients to have this process made clear to them, to assist them in improving their decisions about activity limitation. Helping patients with chronic pain to improve their understanding of the effects of activities on their bodies, and hence the

likelihood of aversive consequences, would assist them in this cost-benefit analysis of activities. Uncertainty could also be reduced by providing patients with information on pain management, providing them with a greater sense of control over their condition, particularly where long waiting lists exist for pain management programmes. Focusing on improving patients' control over their condition, would also go some way towards addressing the fear and anxiety which some participants described.

The constraints discussed above indicate the need for replication of these results, potentially in samples of chronic pain patients with a different underlying pathology, as well as further exploration of this decision making process in large quantitative studies, to enable the generalisability of the findings to be established.

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Part 3: Critical Appraisal

Introduction

This paper reflects on the process of carrying out the empirical research, and encompasses all aspects of that research, including the choices about the design, carrying out the interview with participants, further reflections on the participants' impact on the research process, the analysis and the results. Finally, some directions for future research are considered.

How the choices were made about the design of the study

The aim of this research project at the outset was to examine the existing literature about avoidance in chronic pain, with a particular view to exploring how well the existing fear-avoidance models explained the process in which people with chronic pain engage, in making decisions about activity limitation.

The initial approach adopted was to design a quantitative study which would focus on particular aspects of the existing models, for example Vlaeyen and Linton's (2000) fear-avoidance model. This model incorporates several important assumptions, for example the importance of catastrophic interpretations of pain, the assumption that catastrophic thinking leads to fear of pain and (re)injury, that this pain-related fear results in hypervigilance for threatening pain-related information, and that pain-related fear leads to avoidance of the fear-engendering activity (Vlaeyen & Linton, 2000). However, I soon found that, within the constraints of a major research project, in which time and resources are limited, and which allows for just one experimental study, it would be possible to focus on only one of these assumptions, which would not adequately address what was a rather broad research question.

These considerations, together with the choice of the population of chronic pain patients on which to focus, influenced my choice of design. Joint Hypermobility Syndrome (JHS) is a condition which is not widely recognised by the medical profession, and has been relatively under-researched. Whilst the pain management clinic at which this research was conducted had collected anecdotal evidence about this group of patients' experiences of chronic pain, there was little empirical evidence. This research project presented an opportunity to examine this group's experiences.

Qualitative research methods are particularly suited to research areas where there is little existing knowledge. Therefore, in view of the lack of existing research from which research questions could be hypothesised and tested for this group, together with the difficulty in addressing the research question within the constraints of a single quantitative study, a qualitative phenomenological approach was adopted.

The participants

The interviews yielded a detailed picture of participants' experience of living with JHS. Although this data was not formally analysed as it did not relate directly to the research question, it is included here as it provides a useful contextual background to this study.

Participants described JHS as an "invisible disease" in which one "looked normal" but was actually disabled. They all had experience of "battling" to get a diagnosis, of not being believed by medical professionals who made them feel they were exaggerating their symptoms, seeking attention, or being a hypochondriac. Many had initially been misdiagnosed, and consequently been exposed to treatment, such as surgery or investigations, which were unnecessary and in some cases, they

believed, led to a deterioration in their condition. They felt JHS had restricted their lives, not just in their day to day activities, but in major life choices, such as their career, having a family, and where to live. They also spoke of the emotional impact, chiefly frustration, anger, depression and guilt, and the losses JHS had caused: missing out on activities, loss of their roles as spouse or parent, loss of their personal identity, financial loss, loss of career, ending of relationships and loss of independence.

Approximately half of the patients approached during the recruitment process agreed to take part in the study, although several of them were ultimately unable to do so for practical and logistical reasons. On meeting the participants, it became clear that the majority of them had gone to great lengths to take part in the study. This often involved them in travelling considerable distances, arranging for a family member to accompany them to the interview, and in some cases making arrangements for their children to be taken care of whilst they attended the interview. In almost all cases, they were aware that the physical demands of travelling to the interview, and sitting in one position throughout the course of the interview would result in an increase in their pain and possibly having to rest for a period after the interview in order to recover. However, they felt very strongly that they wanted to take part in this study, so that they could tell their story, and make some contribution towards ensuring that knowledge about JHS amongst health professionals was increased, to prevent others from suffering as they felt they had.

In particular, as discussed above, they had often battled to get a diagnosis, and frequently had initially been misdiagnosed and consequently been exposed to inappropriate treatment, which they felt had resulted in an avoidable deterioration in their condition. Therefore, this strong need to tell their story and do something

proactive, suggested the possibility that the group who participated may have had a different experience of JHS from those who chose not to. Without knowledge of those who chose not to participate, there can be no evidence for any differences, however it was possible that there were. For example, perhaps those who did not respond to the initial recruitment letter did not feel as strongly about their experience of JHS, or perhaps were less affected by it, or alternatively were disabled to the extent that they felt unable to take part. There may also have been other differences, but the important consideration is that the sample of patients who took part may not be representative of the general population of patients with JHS, limiting the generalisability of the results.

The interview

The research began coming to life when I began conducting the interviews, and talking to the participants about their experiences. Many of them seemed to appreciate the opportunity to describe the effect of JHS on their lives and the challenges they faced, to be listened to in a non-judgmental manner, and to be believed. Perhaps because of this, some sections of the interview took longer than planned, such as the introductory sections about their experience of JHS and of chronic pain: whereas I had planned to discuss these questions by way of introduction, and to understand the context in which they made decisions about their activities, this very often took a long time, so that I had to be fairly disciplined about moving the discussion on to talk about the topics related more directly to the research questions, while at the same time being flexible, a conflict referred to by Smith and Osborne (2003).

When I analysed the transcripts of these sections, I often found this flexibility rewarded by very relevant details which helped me understand the participant's approach to activity limitation. In a few cases, participants gave very detailed accounts of aspects of their lives which did not relate to JHS or their experience of chronic pain, and I sometimes had to be gently assertive to maintain the focus of the discussion.

Some of the participants became very distressed talking about the impact that JHS had had on their lives, particularly where they felt they had suffered losses as a result of it. This created some tension for me within the interview, between my role as a researcher conducting an interview, and my role as a psychologist.

When participants talked about how they made decisions about activity limitation, there was a range of responses. Participants who felt that pain interfered to a large extent in their lives, gave very thoughtful responses, giving explicit explanations about how they considered the activity, its consequences and whether it was worth it, perhaps because they had had to think very carefully about which activities they could participate in.

Other participants thought it was obvious that the prospect of pain or injury would prevent them from doing an activity, for example, as one participant put it "You hear about people dislocating themselves all the time anyway on skiing holidays, so why would someone who spontaneously dislocates get on a pair of skis?" [P5] At times, when I received such a response, I did experience a small moment of anxiety, wondering how I could discover the thinking behind a response carrying such a sense of finality. However, by probing further, using contrasting examples from the participant's experience, and discussing hypothetical "what if?" scenarios, the thought processes implicit in such a statement became apparent. One participant was

quite apologetic, as she felt she just did things without thinking, or made her decisions subconsciously, based on years of experience, yet as we discussed her choices about activities further, her decision making process became clearer.

The commitment of the participants to taking part in the research, was exemplified by the experiences of two of them. One of them suffered severe fatigue due to very poor sleep as a result of pain, usually only getting about 2 hours unbroken sleep per night. On the morning of her interview she overslept by several hours, but was still very keen to attend the interview, even though she would be over an hour late. This meant that my subsequent interview would have to start late. By the time I contacted the second participant to ask whether her interview could be delayed, the taxi she had organised to bring herself and her husband to the interview was already on its way. Nevertheless, she was very willing to wait, to give the previous participant an opportunity to attend the interview, and both her and her husband waited patiently and uncomplainingly for over an hour.

Having chosen to use a qualitative approach, the value of this method became apparent during the course of the interviews with the participants and the analysis of their transcripts. The semi-structured interview allowed participants the freedom to discuss aspects of their experience of JHS and its effect on their activities, which I probably would not have raised with them within the constraints of a quantitative design, which would have focused on specific aspects of their experience. The qualitative approach, particularly the use of open-ended questions in the semi-structured interview, such as, “How do you decide what is ok to do, or better not to do?” enabled participants to draw on their own experiences, and impart a richness to the descriptions, instead of being limited in their remarks to those aspects of their experience which the existing literature indicated were important. In retrospect, this

was particularly important, in view of the fact that I had no previous experience of working with chronic pain, so that my knowledge and hence my design of the study was based largely on the literature which I had read. Using the qualitative approach, the resulting descriptions of the process of decision making revealed a greater complexity than that suggested by the existing models. The themes derived from the analysis provide ideas for some new research directions which may not have been revealed by a quantitative approach focused on particular aspects of the existing models.

The analysis

However, the analysis of the transcripts using the Interpretative Phenomenological Analysis approach (Smith & Osborn, 2003) presented some difficulties and revealed some potential constraints of this approach. The initial phase of the analysis, which involved annotating anything “interesting or significant” about what the participant said, revealed the depth and complexity of experience contained in their descriptions, often beyond what I expected in the planning of the study and the design of the semi-structured interview.

The description of this process suggests the first difficulty. What was “interesting” and “significant” was clearly a subjective decision and depended upon my own opinion, and my interpretation of the data. These in turn would have been influenced by my theoretical orientation, the research questions being examined and the hypotheses which I had formed about these, based on my interpretation of the existing literature. It seemed that this process was susceptible to a confirmation bias, in which I would be likely to find those aspects of the participants’ accounts which coincided with my own hypotheses, significant. These difficulties highlighted for me

the importance of credibility checks and “owning one’s perspective” (Elliott, Fischer, & Rennie, 1999), which involves the researcher describing their own theoretical orientation and biases, so that the reader is aware of the potential bias in interpretation.

Faced with an abundance of interesting and seemingly relevant annotations, the process of transforming these initial notes “into concise phrases which aim to capture the essential quality of what was found in the text” (Smith et al., 2003) also caused me some difficulties, the most apparent being a fear of losing valuable information in the process of moving from these initial annotations to main ideas or themes. Participants may have talked about a theme in more than one way, giving more than one example of their experience. Combining a range of remarks into one idea or theme seemed to risk losing interesting and relevant information. However, attempting to retain this detail would have hindered the process of finding the essence of participants’ experiences.

I used the method set out in Smith et al.’s (2003) paper, which involved a case by case analysis of the transcripts, so that after analysis of the first transcript, an initial set of themes had been created. However, whether one chooses to analyse the subsequent transcripts from scratch, or to use the themes identified from the first transcript to inform the analysis of subsequent transcripts, it seemed inevitable that the themes from the first transcript influenced the annotation and description of themes in subsequent transcripts. I was concerned that had I begun the analysis with a different transcript, I may have derived a different set of initial themes, and consequently, my analysis of subsequent transcripts may have progressed in a slightly different direction. It seemed that the first transcript analysed would be the most influential, as analyses of subsequent transcripts tended to conform to the

thematic structure created in the first. As my analysis of subsequent transcripts progressed, these later transcripts appeared to be diminishingly important in influencing the identification and definition of unique themes.

At the stage of combining the themes into a narrative account of participants' experiences, I found that the individual participant's narratives had become lost behind the framework of themes which had been created. The product of the analysis is initially a list of themes, from which I found it difficult to produce a narrative which retained the richness of participants' accounts and avoided oversimplifying them. Therefore to overcome this, before writing the narrative account, I re-read each participant's transcript and summarised it in the form of a brief narrative synopsis, attempting to capture the essential aspects of how they went about making decisions about activity limitation, focusing on the research questions. I found that combining the process of producing a brief synopsis with the process of filtering out a list of themes, seemed to allow for a systematic identification of themes, as well as ensuring that the final narrative closely reflected the original data.

Reflections on the results

After interviewing eleven participants and carrying out a detailed analysis of their transcripts, it was revealed that JHS patients make decisions about activity limitation in much the same way that decisions are made in the day to day lives of people in all walks of life: they weigh up the costs and the benefits of the alternatives, considering the likely consequences and the potential benefits, and making a choice which achieves a satisfactory balance between the two. At times this rational process is abandoned, consequences are ignored or not considered, and people do what they

feel like. As one participant put it, “I’m sure this is just the same in everybody’s life” [T9], and this process could be applied to any decision.

However the context of chronic pain in which these decisions are made, does add some unique dimensions to the process. The factors which participants consider in reaching these daily decisions appear to pertain to the effect on their body and their health, risks to their own sense of self, impact on their relationships, and the impact on their lives both in terms of the next few days or weeks, and in terms of possible permanent damage and disability. These factors suggest that their decisions carry a potential risk of large personal loss. Furthermore, their accounts suggested that their decision relating to a particular activity will not always be the same, but may be influenced by factors which vary from day to day, such as their level of pain at that moment, how certain they feel that their health and personal resources would enable them to carry out that activity, and the relative importance of other plans which they may have in the near future, which may lead them to be more cautious to ensure that they are able to fulfil later plans. The emotional outcomes of engaging in an activity also appear to be considered, for example fear and anxiety relating to particular consequences, embarrassment, guilt or depression. Implicit in their accounts was the suggestion that their decisions also appeared to be influenced by their perception of their current level of health and the stability or deterioration of their condition, as well as the distress which it causes them. Their decisions appeared to be made more complex by the unpredictability which they attach to the consequences of an activity, based on their previous experience of it.

Therefore, while it was interesting, albeit with the benefit of hindsight, slightly predictable, to discover that JHS patients make decisions using a cost-benefit analysis, it turned out that what was most interesting, was participants’ accounts of

the factors *influencing* the process of decisions around activity limitation. To establish whether these influences are unique to chronic pain patients, it would be interesting to carry out the same study with a sample of healthy volunteers, focusing on the process of their decisions relating to activities of varying degrees of importance. This would contribute to isolating any aspects of the decision making process which are unique to JHS patients suffering from chronic pain.

Intuitively, it seems unlikely that a healthy individual would engage in such a detailed and deliberate process about engaging in an activity unless the decision was very important, had considerable consequences or presented a significant challenge. It seems more likely that the overriding factor in reaching the decision for a healthy individual, would be what they *wanted* to do. This may highlight an important feature of JHS patients' decision making about activities: to them, the consequences may be far reaching, in that it is likely that they may suffer injury, or severe pain of several days' duration, or even permanent damage, which could lead to further disablement and emotional consequences. Hence the likelihood of suffering loss, as well as the size of those potential losses appear to be bigger for the JHS patient, than for the healthy individual.

Another difference seems to be the uncertainty which JHS patients face in making their decisions: a healthy individual (with no particular physical problems or emotional difficulties to contend with such as fear of heights) would encounter little unpredictability in the way their body would react to activities or in their ability to cope with those reactions, unless they were undergoing some change, for example facing the onset of older age. By contrast, JHS patients found their pain unpredictable, difficult to relate to particular activities, and felt anxious about their ability to control pain when it reached a certain level. This introduced a dimension

of caution into their decision making which may not be so apparent in the healthy individual's decision making process.

However, the hypothetical nature of the above discussion highlights the need for empirical examination of the aspects of decision making in the context of chronic pain, which sets it apart from the decision making of healthy individuals.

This consideration of healthy individuals also highlights the potentially pathologising nature of fear-avoidance models. Vlaeyen and Linton's (2000) fear-avoidance model proposes that the judgement of the meaning or purpose of the pain determines whether the individual follows a pathway of "no catastrophising" or "no fear" leading to recovery, or alternatively places a catastrophic meaning on the pain, leading to avoidance and hence disability. Whilst it seems that healthy individuals would be regarded as quite rational in refraining from activity which caused pain or discomfort, the use of the word avoidance in this model, seems to imply a shirking, or not doing something that one really ought to be doing. This seems to suggest that people suffering from chronic pain are expected to do more than a healthy individual would be expected to do in similar circumstances, echoing the view that "disabled people have to overcompensate to be accepted into the community. The negative psychological implications for the majority struggling to cope in a largely hostile environment are clear" (Barnes, 1992).

Future research

The discussion above highlights the need for replication of this study, not only in samples of chronic pain patients with different underlying pathology, but also in groups of healthy volunteers, which would help to elucidate those aspects of the process which are unique to chronic pain sufferers. Further exploration of these

findings in large quantitative studies would contribute to establishing the generalizability of results, and enable more detailed analysis of particular aspects of the process. For example, it may be useful to explore the individual's perception of loss and their level of distress, which may affect the weight they attach to the aversive consequences of an activity. The results of this study suggested that an elevated level of distress, a heightened perception of loss and previous aversive experiences, made the individual more cautious in balancing the benefits and consequences of an activity. A better understanding of this process may also have useful clinical implications, in that focusing on an individual's distress and perception of loss would not only be therapeutic in itself, but may also enable the individual to increase their level of activity.

Conclusion

This research has given some insights into a relatively neglected group in the chronic pain research literature. Using a qualitative approach, it has revealed the complex process of decision making around activity limitation, not previously described in the literature. This has suggested some areas for future research and possible areas of focus for clinical application.

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Appendices

Appendix A: Letter to patients

Date

M

Dear M

Research study on Joint Hypermobility Syndrome

Following your referral to the COPE pain management programme, I would like to tell you about a research study which is taking place, which may be of interest to you.

I have enclosed an Information Sheet describing the research and what it would involve. Please take the time to read the sheet carefully. If you would like to take part in the study, please complete the tear off slip attached to the Information Sheet, and return it in the stamped addressed envelope provided, or reply by email to the following address:

Thank you for your help.

Yours sincerely

Professor
Consultant Rheumatologist



UCL Hospitals is an NHS Foundation Trust incorporating the Eastman Dental Hospital, Elizabeth Garrett Anderson & Obstetric Hospital, The Heart Hospital, Hospital for Tropical Diseases, National Hospital for Neurology & Neurosurgery, The Royal London Homoeopathic Hospital and University College Hospital.



Appendix B: Patient information sheet

Patient Information sheet

Activity Limitation in Joint Hypermobility Syndrome

I would like to ask you if you would be willing to take part in a research study. In order for you to decide, I have provided information about why the research is being done and what it will involve, for you to read and discuss with others if you wish. If there is something you want to know which I haven't covered, please feel free to contact me and ask.

After reading this information sheet, if you decide you would like to take part, please turn to the back page, which tells you how to contact me.

What is the purpose of the study?

This study aims to explore how patients' lives are affected by Joint Hypermobility Syndrome (JHS), and will focus in particular on what activities or demands JHS patients avoid and how they make decisions about whether or not to avoid them. The aim will be to understand the process of patients' decisions around avoidance and to explore whether the theories about chronic pain described in the literature, can be used to help understand JHS patients' experience of avoidance and chronic pain. A better understanding of these decisions will help us in assessing patients, designing treatment, and in further research.

Why have I been chosen?

I am looking for people who are JHS patients who have experienced pain lasting at least 6 months. Between 10 and 15 participants will be included in the study.

Do I have to take part?

No. It is up to you to decide whether or not to take part. If you do, you will be given this information sheet to keep and be asked to sign a consent form at a later date. You are still free to withdraw at any time and without giving a reason. If you withdraw, we will ask your permission to use any interview material we have collected from you up to that point. A decision to withdraw



UCL Hospitals is an NHS Foundation Trust incorporating the Eastman Dental Hospital, Elizabeth Garrett Anderson & Obstetric Hospital, The Heart Hospital, Hospital for Tropical Diseases, National Hospital for Neurology & Neurosurgery, The Royal London Homoeopathic Hospital and University College Hospital.



at any time, or a decision not to take part, will not affect your treatment in any way.

What will happen to me if I take part?

You will be asked to take part in a meeting at the clinic at which you attend your normal appointments and this will be conducted in a venue which will enable the meeting to remain confidential :

- 1) I will interview you for about one to one and a half hours. During this interview I will ask you about your experience of living with JHS, with a particular focus on how it has led you to avoid certain activities.
- 2) After completion of the analysis of all the interviews, I will ask for your comments on the results of the analysis. This is likely to take place at least 3 months after completion of all the initial interviews and will take approximately 30 minutes.

If possible, interviews will be arranged to take place when you are attending an appointment. If this is not possible, specific appointments will be arranged at times mutually convenient to you and me. I expect that all interviews and follow-ups will be completed by April 2007.

Interviews will be audio-taped so that they can be transcribed for analysis. Once the transcript has been completed the audio-tape will be destroyed. The transcript will remain confidential, and will contain no information which would enable you to be identified. My supervisors at University College London may read some of the transcript material, but they would not be able to identify you. It is possible that when the study is written up, transcript material may be quoted to enhance the report, however your permission would be sought to do so, and all quotations would be anonymous.

Expenses and payments:

You will be offered payment of your travel expenses to attend the meeting.

What do I have to do?

I would like you to do the initial interview with me, and later, to provide your comments on the analysis of all the interviews. If you are unable to attend a meeting, I would like you to let me know in advance, so that we can re-arrange it.

What are the possible disadvantages and risks of taking part?

Some participants may find it distressing to discuss the effect of JHS on their lives, but as a trainee clinical psychologist, I have experience and training in such situations and will do what I can to help. I am also being supervised by a clinical psychologist. You will be interviewed in the hospital at which you

usually attend appointments, so you will have access to a clinician should that be necessary.

What are the possible benefits of taking part?

You may find that the interview is an interesting opportunity to think about how JHS has affected you. The insights which you provide into living with JHS will get fed back into our knowledge of treating JHS, and we hope that this will help improve both our understanding and treatment of people with JHS.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed.

Any complaints should be addressed to the Supervisor of this research (Dr Amanda C de C Williams, Sub-Dept of Clinical Health Psychology, University College London,). If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure. Details can be obtained from the hospital.

Harm:

Appropriate redress and/or compensation will be available in the event of negligent harm and this cover is provided by the Clinical Negligence Scheme for Trusts. There are no indemnity arrangements for non-negligent harm.

Will my taking part in this study be kept confidential?

Yes. All the information about your participation in this study will be kept confidential. The audio-tape of your interview will be destroyed once the transcript has been completed. The transcript of your interview will be identified by a reference number only and will contain no information which would enable you to be identified. A list of reference numbers linked to personal details to enable us to contact you after the initial interview will be stored securely in a locked cupboard in the Sub-Dept of Clinical Health Psychology at UCL. Transcript material and analysis will be shared with supervisors at UCL for the purposes of training and to ensure the validity of the interpretation. However, none of this material will contain information which would enable you to be identified.

Transcripts will be retained for 5 years after publication of the study, which is normal scientific practice, but will not be used for future studies. Thereafter they will be disposed of securely.

Our procedures for handling, processing, storage and destruction of your transcript are compliant with the Data Protection Act 1998.

The custodian of the data is _____, Reader in Clinical Health Psychology, University College London.

What will happen to the results of the research study?

The results of this research will form part of my Doctorate in Clinical Psychology. The intention would also be to publish the results in a peer-reviewed scientific journal. You will have the opportunity to give feedback on the results before they are published. You will also be provided with a summary of the results. It is possible that when the study is written up, transcript material may be quoted to enhance the report, however your permission would be sought to do so, and all quotations will be anonymous.

Who is organising and funding the research?

This research is being sponsored by the University College London Hospitals. The research is being funded by the Sub-Dept of Clinical Health Psychology, University College London.

Who has reviewed the study?

This study was given a favourable ethical opinion for conduct in the NHS by the National Hospital for Neurology and Neurosurgery and The Institute of Neurology Joint Ethics Research Committee. It has also been approved by the University College London Hospitals Research and Development Directorate.

Contact Details:

If you would like some further information about the study, or if you have any questions please contact me. My details are:

Anne Schmidt (Chief Investigator)
Tel:
Email:

Should you wish to participate in the study you will be given a copy of the information sheet and a signed consent form to keep.

Thank you for taking time to read this sheet.

If you would like to take part in this study please return the tear-off sheet below, to:

Anne Schmidt

Please respond within one week of receiving this information sheet.

-----Tear off this sheet

Please provide the following details about yourself:

Name:

Address:

Post code:

Telephone:

Email:

I am interested in taking part in this study. Please contact me to arrange an appointment.

Signature: _____

Please send to:

*Anne Schmidt,
Sub-Department of Clinical Health Psychology,
University College London,*

OR Email your details to:

OR Text me on:

Appendix C: Ethical approval

**The National Hospital for Neurology and Neurosurgery
& Institute of Neurology Joint REC**
Research & Development

Mrs D Anne Schmidt
Trainee Clinical Psychologist
University College London
Sub-Dept of Clinical Health Psychology
University College London

Our Ref: 06L 350

06 November 2006

Dear Mrs Schmidt

Full title of study: **Avoidance in Joint Hypermobility Syndrome**
REC reference number: **06/Q0512/57**

Thank you for your letter of 23 October 2006, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information was considered at the meeting of the Sub-Committee of the REC held on 03 November 2006. A list of the members who were present at the meeting is attached.

Kindly note that a completed substantial amendment form is requested for Point 3 of your letter, with any supporting documents.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised.

Conditions of approval

The favourable opinion is given provided that you comply with the conditions set out in the attached document. You are advised to study the conditions carefully.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Application	5.1	17 May 2006
Investigator CV	1	17 May 2006
Protocol	1	24 March 2006
Covering Letter	1	23 October 2006
Peer Review	1	03 May 2006
Participant Information Sheet: Participant Information Sheet	1	28 April 2006
Participant Information Sheet	2	29 August 2006
Participant Consent Form: Participant Consent Sheet	1	28 April 2006
Response to Request for Further Information		23 October 2006

Draft Covering Letter	1	23 October 2006
Draft Reminder letter	1	23 October 2006

Research governance approval

The study should not commence at any NHS site until the local Principal Investigator has obtained final research governance approval from the R&D Department for the relevant NHS care organisation.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

06/Q0512/57	Please quote this number on all correspondence
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With the Committee's best wishes for the success of this project

Yours sincerely

Chair

Email:

*Enclosures: List of names and professions of members who were present at the meeting.
Standard approval conditions
Site approval form*

Copy to: R&D Department for UCLH

**The National Hospital for Neurology and Neurosurgery & Institute of Neurology Joint
REC**

Attendance at Sub-Committee of the REC meeting on 03 November 2006

Committee Members:

<i>Name</i>	<i>Profession</i>	<i>Present?</i>	<i>Notes</i>
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(Chair)

Consultant Nurse Y

(Vice-Chair)

Consultant Nephrologists Y

The National Hospital for Neurology and Neurosurgery & Institute of Neurology Joint REC

LIST OF SITES WITH A FAVOURABLE ETHICAL OPINION

For all studies requiring site-specific assessment, this form is issued by the main REC to the Chief Investigator and sponsor with the favourable opinion letter and following subsequent notifications from site assessors. For issue 2 onwards, all sites with a favourable opinion are listed, adding the new sites approved.

REC reference number:	06/Q0512/57	Issue number:	1	Date of issue:	06 November 2006
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Chief Investigator:	Mrs D Anne Schmidt
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Full title of study:	Avoidance in Joint Hypermobility Syndrome
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This study was given a favourable ethical opinion by The National Hospital for Neurology and Neurosurgery & Institute of Neurology Joint REC on 03 November 2006. The favourable opinion is extended to each of the sites listed below. The research may commence at each NHS site when management approval from the relevant NHS care organisation has been confirmed.

Principal Investigator	Post	Research site	Site assessor	Date of favourable opinion for this site	Notes ⁽¹⁾
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Approved by the Chair on behalf of the REC:

..... (Signature of Chair/Co-ordinator)
 (delete as applicable)

..... (Name)

(1) The notes column may be used by the main REC to record the early closure or withdrawal of a site (where notified by the Chief Investigator or sponsor), the suspension or termination of the favourable opinion for an individual site, or any other relevant development. The date should be recorded.

Confirmed 22/11/06 by phone. Research in UCLH is approved. Not site specific hence no extra sites listed. A. write letter & minor amendments & new into sheet.



Mrs D Anne Schmidt
Trainee Clinical Psychologist
Sub-Dept of Clinical Health Psychology
University College London

Research & Development Department

Our Ref: 06L 379

Website: www.uclh.nhs.uk

05 December 2006

Dear Mrs Schmidt

Study title: Avoidance in Joint Hypermobility Syndrome
REC reference: 06/Q0512/57

Amendment number: 1
Amendment date: 22 November 2006

The above amendment was reviewed at the meeting of the Sub-Committee of the REC held on 05 December 2006.

Ethical opinion

The members of the Committee present gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
Participant Information Sheet	3	23 November 2006
Cover letter for patients been referred to the COPE program	1	23 October 2006
Reminder letter following I.S	1	23 October 2006
Notice of Substantial Amendment (non-CTIMPs)	1	22 November 2006

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Research governance approval

All investigators and research collaborators in the NHS should notify the R&D Department for the relevant NHS care organisation of this amendment and check whether it affects research governance approval of the research.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

06/Q0512/57:

Please quote this number on all correspondence

Yours sincerely

Committee Co-ordinator

E-mail:

Copy to: R&D Department for NHS UCLH

Enclosures List of names and professions of members who were present at the meeting.

The National Hospital for Neurology and Neurosurgery & Institute of Neurology Joint
REC

Attendance at Sub-Committee of the REC meeting on 05 December 2006

<i>Name</i>	<i>Profession</i>	<i>Capacity</i>
<i>(Chair)</i>	<i>Consultant Nurse</i>	<i>Y</i>
<i>(Vice-Chair)</i>	<i>Nephrologists</i>	<i>Y</i>

Appendix D: Research and Development approval



Joint UCLH/UCL Biomedical Research (R&D) Unit
Director

Web-site: www.uclh.nhs.uk

24 November 2006

Mrs D A Schmidt
Sub-Dept of Clinical Health Psychology
UCL

Dear Mrs Schmidt

Project ID: 06/0085 (*Please quote in all correspondence*)
Title: Avoidance in Joint Hypermobility Syndrome (JHS)

Thank you for registering the above study with the R&D Directorate. I am pleased to give the approval of UCL Hospitals NHS Foundation Trust for the study to proceed.

You will be aware that as principal investigator you have various responsibilities under the Department of Health's *Research Governance Framework for Health and Social Care*. Please note that you are required:

- to comply with the UCLH Information Security Policy (the R&D Directorate's data protection toolkit "Consent and Security" will help you meet the requirements of the Data Protection Act and is available at <http://www.uclh.org/services/research/>).
- to ensure that any co-investigator who is not an employee of UCLH has in place an up-to-date honorary contract.
- to keep copies of all consent forms with your project documentation. UCLH carries out audits of informed consent and if your project is selected for audit, you will need to provide access to the consent forms.

Please ensure that you have addressed any outstanding issues raised by the research ethics committee and have ethical approval before you start your project. Also you must ensure that you comply with all the requirements of the ethics committee regarding progress reports, notification of protocol amendments and adverse events.

You are strongly recommended to use an investigator file to store all the documentation relating to this research project. This will help facilitate the research audit process which is now a research governance requirement. The attached list of headings is designed to help you assemble your investigator file.

Yours sincerely

Director of R&D, UCL Hospitals NHS Foundation Trust



Appendix E: Consent Form

Appendix F: Demographic questionnaire

Background Questionnaire

Name	
Age	
Are you currently working	
Occupation	
Marital status	
Do you have any children	
Highest level of education attained	
Please list previous illnesses or injuries	
Have you seen a psychologist before? Please describe.	
What medication are you using at the moment?	
How much alcohol do you consume per week?	
Do you use any drugs? If so, please describe.	
Do you use any other substances? If so, please describe.	

Appendix G: Semi-structured interview schedule

Semi-structured interview

Introduction

Prompts

- Describe research, review information sheet
- Purpose of interview
- Reimbursement of travel expenses
- Consent form
- Ask participant to complete socio-demographic questionnaire (see appendix)

The experience of JHS

- **What's it like having JHS?**

Prompts

- o How long was it until you were given a diagnosis?

- **What's it like being in pain?**

Prompts

- o Where is your pain?
- o How long have you had it?
- o Has it changed over time?
- o What's it like for people around you when you're in pain?

The effects of pain on your life: activity limitation and restriction

- **How has your life changed since you started having pain/since your pain got really bad?**

Prompts

- o Work
- o Home
- o Interests, leisure, social
- o Relationships
- o Personal identity

- **Are there things that you prefer not to do, or that you cannot do, when you're in pain?**

Prompts

- o stimulation, movement, activity, social interaction, leisure pursuits.
- o Work
- o Home
- o Interests, leisure activities, social
- o Exercise, sport

The activity limitation decision making process

- **How do you decide what is ok to do, or safe to do, or better not to do?**

Prompts

- Discuss areas raised by participant in previous section
- Does being uncertain about your diagnosis make it seem more important to avoid stuff in case you do some damage?

- **How does avoiding these things affect you?**

Prompts

- Does it make you feel better or worse physically?
- How does it affect your mood?
- How does it affect your relationships, work, family, leisure?

- **What would happen if you did not avoid these things?**

Prompts

- Do you have any images of what might happen, e.g. to your body?

- **What advice have you been given about doing things or not doing things/about what you can and can't do/should and shouldn't do?**

Prompts

- From professionals
- From other people suffering pain, [e.g. self-help groups, have you taken part in any of these]
- From family/friends?
- From things you've read?

- **Can you think of a specific example of something you did not do recently?
How did you make the decision not to do that activity?**

Prompts

- How did you feel about being faced with that activity?
- How did you decide not to do that?
- When did you make the decision?
- Did anyone else influence your decision?

- **How will you decide whether to go back to doing something you've given up or limited, or to give up or limit other things in future? (change your level of activity in future?)**

Prompts

- What would make you give up other things?
- What would make you go back to an activity that you've given up?

Conclusion

- Is there anything I haven't asked that you think might be important?
- Do you have any questions for me?
- How have you found talking to me today?

Appendix H: Stages of the analysis

Stages of the analysis

Stage one: annotation of interesting items

Trancript one	Initial Annotation
T What is it like having joint hypermobility syndrome?	
P I don't want to think negatively, I try to think positively but it's very limiting I cannot do everything I want to do, I cannot do the sport I want to do, I cannot do all the things I want to do in one day, I have to rest quite a lot, I cannot see my friends when I want, where I want, there's a lot of things I cannot do, and I have to deal with this, but not thinking too much about it, and just think I have a normal life, so I just try to manage my life but try to do the maximum I can, even if I have pain.	<ul style="list-style-type: none"> • <i>Positive mental attitude</i> • <i>Activity limitation</i> • <i>Doing less in one day</i> • <i>Need for rest</i> • <i>Limits on social life</i> • <i>Desire to be normal</i> • <i>Do as much as I can</i>
T Yeah, ok, so it sounds like it's quite a lot of limitations but you try to get through those?	
P Yes or I do them in a different way, or I'm not thinking too much about them if I cannot do something I'm just ...trying to keep off my mind and do something different.	<ul style="list-style-type: none"> • <i>Do things differently</i>
T Ok. So where are the main places that you have the pain?	
P My knee....when I was skiing I said oh yeah I have the knee pain also if I go hiking in the mountain I cannot do it.	<ul style="list-style-type: none"> • <i>Pain prevents activity</i>

Stage two: transform into main ideas

Initial annotation

Idea arising from annotation

Positive mental attitude

Positive attitude

Activity limitation

Limitation – giving up activities

Doing less in one day

Doing less

Need for rest

Need for rest

Limits on social life

Limitation of social activity

Desire to be normal

Desire to be normal

Do as much as I can

Balancing pain and activity

Do things differently

Adaptations

Pain prevents activity

Effect of pain

Stages three to six:

Stage 3: group similar ideas from first transcript into clusters	Stage 4: addition of new ideas from analysis of remaining transcripts	Stage 5: production of a final list of themes	Stage 6: organise final list of themes into two domains
Limitation – giving up activities Limitation – positions and movements Effect on sleep Effect of fatigue	Emotional impact – loss Emotional impact – frustration Loss of independence Relying on others	1.1 Limitation and restriction	Domain one: Impact on activities
Adaptations Doing less	Fatigue – need for rest Change of career Varying activities	1.2 Changing how I do things	
Difficulty driving	Effect of physical environment Use of public transport Planning trips	1.3 The difficulty of travel	
Limitation of social activity Loss of spontaneity	Social withdrawal Use of aids Inability to plan in advance	1.4 Curtailment of social life	
Role of parent Effect on personal identity Effect on family	Impact on relationships	1.5 Narrowing of roles within the family	

Stages three to six (continued)

Stage 3: group similar ideas from first transcript into clusters	Stage 4: addition of new ideas from analysis of remaining transcripts	Stage 5: production of a final list of themes	Stage 6: organise final list of themes into two domains
Emotional consequences Effect on my body Responsibility to others Importance of activity	Consequences for other people Missing out on other plans Prioritising	2.1 Is it worth it?	Domain two: Process of decision making about activity limitation
What I want to do Positive attitude Desire to be normal	Determination Think about consequences later	2.2 What I want	
Adaptation Pacing Managing fatigue	Changing priorities	2.3 Pacing and adapting	
Balancing pain and activity Conflict between pain and activity Exercise as treatment Need for rest	Regulating pain Finding a level of pain Self-monitoring	2.4 Balancing the pain	
Characteristics of activity Physical environment of activity	Physical demands of activity Risks for injury Time available Travelling involved	2.5 Thinking about the activity	
Effect of pain No choice	Pain as a message from body Listening to my body	2.6 There's nothing to think about	
Uncertainty about consequences Unpredictability of pain	Difficulty planning Uncertainty of body's reaction	2.7 Struggling with unpredictability	
Level of pain	Feeling good or bad Good day or bad day Progress of condition	2.8 How I'm feeling	
Fear of deterioration	Anxiety about activity Fear of pain Uncertainty about ability to cope	2.9 Fear and anxiety	
Need for control Ability to escape		2.10 Staying in control	