How do people with chronically painful joint hypermobility syndrome make decisions about activity?

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Abstract

Background: The model of activity avoidance prompted by fear of increased pain and/or harm dominates understanding and research into activity limitation in chronic pain. Yet, the accounts of people with chronic pain on decisions about activity limitation are rarely heard beyond the confines of fear and avoidance questionnaires.

Methods: We used semi-structured interviews to explore the decisions of 11 women attending a pain management clinic with chronically painful Joint Hypermobility Syndrome (JHS).

Results: Six themes emerged from Interpretative Phenomenological Analysis: the overall aim of keeping pain to a manageable level, considering whether the planned activity was worth it and, running through all judgements, the influence of pain intensity. The decision was tipped towards avoidance by unpredictability of pain and by high emotional cost and towards going ahead with the activity by the wish to exert control and by low emotional cost. Many accounts described a specifiable cost–benefit analysis of individual decisions, weighing the importance of each activity against its potential aversive consequences, which only in a minority of cases was dominated by fear of pain or injury.

Conclusion: Assumptions of fear as the basis of activity avoidance should not be used uncritically in clinical settings. Decisions about activity should explore beyond pain expectancy, incorporating goals, values, and decision processes.

Summary points

The model of fear of pain or re/injury and associated avoidance, an important insight that has generated effective therapeutic interventions, risks being over-applied and assumed rather than demonstrated. Patients' own accounts, using qualitative analysis of interview in 11 women with long term chronic pain associated with joint hypermobility, give a more nuanced description of complex decision-making around activity. While a few activities were unquestionably avoided because of such fears, others were undertaken when benefits (according to personal values, such as children's needs) outweighed costs in pain and distress. We suggest that activity needs to be discussed with patients beyond asking about avoidance and in the context of their lifestyle choices.

Keywords

Fear, avoidance, disability, decision-making

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Introduction

The fear and avoidance model of chronic pain introduced by Vlaeyen et al.¹ and reformulated since^{2,3} is widely adopted with plentiful data supporting the links between anxiety about pain and damage and (self-reported) avoidance.³ The fear and avoidance model predicts that when envisaging or attempting painful activities, fearful thoughts and images about increased pain or possible injury prompt escape: abandoning the activity or the preparation for it. Since avoidance pre-empts the possibility of disconfirmation – completion of the activity without increased pain or harm – avoidance persists and may extend to other activities.

Recent reviews have raised concern about two issues in particular: the scarcity of tests of some of the model's assumptions with inconsistent findings from the few tests that are published,⁴ and the misleading nature of analogies with phobia given the major differences in presentation and focus.^{3,4} Furthermore, although avoidance is widely observed and reported, associated predictions of disuse have not been consistently supported.⁵ Crombez et al.⁴ in a critical reformulation emphasise the need to embed the model of fear and avoidance in the context of multiple competing goals, to avoid trait-like description of avoidance and to recognise people's efforts to function despite pain.

The voices of people with chronic pain are rarely represented beyond the constraints of questionnaires about fear and avoidance. Might avoidance or limitation of activity, observed or reported, be associated with motivations other than fear? Findings of association between (self-reported) avoidance and greater disability are at a broad level and do not necessarily pertain to

1. The decision to avoid a particular activity;
2. The consistency of such decisions within and across activities within an individual;
3. The extent to which avoidance is associated with strong expectation of immediate pain or damage rather than with moderate estimates of cumulative risk; the former is more easily challenged.

There are several further models applicable to avoidance. One is safety behaviours, behavioural or cognitive attempts to alleviate threat thus preventing disconfirmation of fearful beliefs,⁶ but safety behaviours can be difficult to distinguish from adaptive coping. Another is that apparent avoidance may be an attempt to follow poor advice from health-care professionals.⁷ Third, persistence despite pain can be counterproductive.^{3,4} We were interested in how people with chronic pain described their decisions about attempting or avoiding potentially pain-exacerbating activities.

Joint Hypermobility Syndrome (JHS) is a musculoskeletal disorder thought to be a subtype of Ehlers–Danlos Syndrome (EDS), an inherited disorder of connective tissue protein characterised by varying degrees of joint hypermobility and increased risk of subluxation and dislocation.⁸ It is not universally recognised in rheumatological medicine, and diagnosis is often delayed.^{8,9} Persistent pain in people with JHS is relatively common,¹⁰ affecting joints, muscles and ligaments⁹ and thereby a range of everyday activities.¹¹

This study aimed to explore, in relation to the models of fear and avoidance and of problem-solving, how JHS patients accounted for the decision to avoid or not to avoid activities that were anticipated to increase pain or to risk dislocation. Since the aim of this study was to understand the complex processes involved in decision-making, a phenomenological approach was adopted to understand people's perceptions of their experiences, the meanings they attach to their experiences, and their underlying assumptions.¹²

Methods

Participants

JHS patients were all referred from the same specialist rheumatologist and so constituted a more homogeneous group than any of our other chronic pain patients. Furthermore, their understanding of their diagnosis gave them no expectations of finding a way of abolishing pain, but of adapting as well as possible to a lifelong problem. Patients eligible for participation in this study had been referred by the specialist rheumatologist to the pain management programme. This programme shows good outcomes,¹³ including reduced interference of pain with everyday life.

Inclusion criteria for this study were pain lasting at least 6 months, diagnosis with JHS by the specialist rheumatologist, ability to attend a face-to-face interview and fluency in English; exclusion criteria were serious psychiatric impairment or substance misuse/abuse problems or other general health condition limiting activity. A total of 32 patients (31 of whom were women) were invited to take part by letter containing information about the study; 15 agreed but four were unable to attend appointments so 11 women (34% of those eligible) were interviewed. Of them, 10 were white and one was Black British; their ages ranged from 22 to 55 years, with a mean of 34 years. Age of symptom onset ranged from infancy to 45 years, with a mean age of onset of 16 years (see Table 1). All but one were using medication for pain relief, and four had previously had some psychological intervention for their pain.

Table 1. Characteristics of participants, in descending order of pain interference.

	Age band	Age at symptom onset, at diagnosis (years)	Highest level of education	Employed	Marital status and children	BPI pain severity	BPI pain interference	Analgesic
P6	40–49	6, 39	School	No	Married + children	7.8	9.7	Compound ^a analgesics
P3	20–29	7, 22	School	Yes	Single	8.0	9.1	NSAID
P11	20–29	7, 11	College/ University	No	Single	6.3	9.0	Compound analgesics, cannabis
P1	30–39	30, 31	Postgraduate	No	Cohabiting	6.0	7.6	Analgesic not specified
P10	30–39	12, 28	College/ University	Yes	Cohabiting + children	7.5	6.9	Compound analgesic
P4	30–39	14, 34	School	No	Single + children	6.5	5.7	None
P9	50–59	30, 49	Postgraduate	No	Single	2.8	5.1	Compound analgesic, NSAID ^a
P7	30–39	12, 28	School	Yes	Single	5.8	4.9	Opioid
P8	50–59	45, 53	College/ University	No	Married + children	6.8	3.0	Paracetamol
P5	20–29	1, 20	College/ University	No	Cohabiting + children	3.8	2.3	Compound analgesic
P2	20–29	14, 18	College/ University	Yes	Single	5.3	1.6	Cocodamol

BPI: Brief Pain Inventory; NSAID: non-steroidal anti-inflammatory drug.

^aContain multiple active ingredients.

Procedure

Ethical approval was obtained from the local Research Ethics Committee. Written informed consent was obtained from each participant. The semi-structured interview lasted approximately 1.5 hours and was audio-recorded. Interviews were conducted by A.S. (main researcher) using guidelines by Smith and Osborn.¹⁴ The first part of the interview covered current pain, pain history and life changes due to pain and established a context for participants' more detailed descriptions of their experience and understanding of pain in the second part of the interview. The latter covered how participants decided what was and was not safe to do and what they avoided, the outcomes of avoidance or of persisting with the activity, advice recalled about activity and a detailed account of a recent decision to avoid an activity.

To provide some context of pain and disability using familiar measures, participants were asked to complete the Brief Pain Inventory (short form) (BPI¹⁵). This assesses pain severity (four items: pain at worst, at least, on average and now) and the impact of pain on activity (seven items). All items are rated from 0 (=no pain/does not interfere) to 10 (=pain as bad as you can imagine/completely interferes). Reliability (Cronbach's α) in non-cancer chronic pain patients is greater than 0.70; the original factor structure has been replicated and the relationship of the BPI to generic measures of pain is strong.¹⁶

Interpretative phenomenological analysis

Interpretative phenomenological analysis (IPA)¹⁴ is described as phenomenological in that it is concerned with personal perception or accounts of an object or event, rather than a quasi-objective statement about it; it is interpretative in that analysis of responses is necessarily influenced by the researcher's own view of the world as well as the nature of the interaction between researcher and participant.¹² The data analyst's (AS's) theoretical orientation was cognitive behavioural, with a particular interest in participants' cognitions and emotions and their effects on behaviour; she had recently begun work in chronic pain management and had no strong views on application of the fear and avoidance model. The last author (AW) had extensive clinical experience in pain management and had concerns about over-application of that model.

All interviews were transcribed verbatim and analysed following IPA methods.¹⁴ Each interview was examined in detail before reading the next, annotating statements that were significant in relation to the research questions. These annotations were summarised as the main ideas, then grouped into themes according to theoretical similarities and apparent connections between them, 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 represented what was said. Themes from earlier transcripts were used to

Figure 1. The process of decision-making.

inform analysis of subsequent transcripts, while allowing new themes to emerge. Once a final list of themes was constructed, all transcripts were re-read, and a brief narrative synopsis that focused on the research questions was produced for each transcript. The list of themes was checked against each synopsis to ensure that it adequately captured the content of each transcript.

Credibility checks were carried out as follows. Identification and representation of themes by AS were discussed with AW at several stages of analysis. The results were checked by AW against transcripts to ensure accurate representation of data. A blind derivation of themes by KC was compared with the existing themes and all three authors agreed on a composite version. Testimonial validity was assessed by checking the results with the original participants; all four who responded agreed with the findings and did not suggest any changes while two expressed surprise at how similar the other participants' experiences were to their own.

Results

Analysis of the patient data on activity avoidance yielded six themes:

1. Keeping pain at a manageable level;
2. Is it worth it;
3. Influence of pain intensity;
4. Unpredictability of pain;
5. Exerting control;
6. Emotional cost of pain.

The themes are elaborated below and illustrated using quotations from participants' transcripts (the number following the quotation is the participant's identification number). They are related to one another in Figure 1: the first three are linked in the process of decision-making and the second three act to push the decision towards avoidance or towards being active.

Process of decision-making about activity

Participants' accounts suggested that an overriding aim is to minimise pain and reinjury and to keep pain within a manageable level. When faced with the possibility of engaging in an activity that did not cause much anxiety, they adopted a cost-benefit analysis approach, weighing up the personal value of doing the activity against their estimation of the likelihood of aversive consequences occurring as a result of doing it. The central consideration in terms of consequences was the intensity and duration of pain that it would cause, as well as the consequences of that pain. In particular, participants needed to estimate the steps that would be required to alleviate the pain, such as bed-rest for an uncertain number of days, the activities that would be missed as a result, the consequent effects on people within their network of friends and family and the potential long term deleterious effects on their health. Balanced against this consideration was the importance that the participant attached to the particular activity: the value of the activity itself and how completing the activity was egosyntonic (acceptable and

consistent with personality and beliefs) in terms of personal importance. Activities that were considered necessary, urgent or to which the participant attached high personal value were given greater weight.

For some participants this appeared to be an implicit process, described in terms of ‘just knowing’ from bodily experience, while for others it appeared to be an explicit process in which the nature of the activity, its physical demands, risks and likely consequences were carefully considered. Although these factors were taken into account, at times physical factors overrode the decision-making process and pain dominated the decision.

The estimate of consequences was made difficult by the unpredictability of pain and the lack of a clear relationship between activity and pain. Attempts to solve the problems of keeping pain to a manageable level, and of its unpredictability, included careful planning, learning from experience, use of coping strategies, pacing and adapting and doing things differently or doing different things. Reaching an acceptance of pain and its limitations enabled participants to make a positive change in their underlying assumptions about how they lived their lives.

Participants also spoke about the emotional cost of pain and impact on decision-making, in terms not just of fear and anxiety, but also depression, anger, frustration, guilt and humiliation. Rather than describing explicit serial steps, weighing costs and benefits, participants described a process in which their decisions were influenced by existing conditions (current intensity of pain), uncertainty and unpredictability, as well as considerations of expected emotional cost. The interaction of these themes is illustrated in Figure 1.

Theme 1: keeping pain at a manageable level. Pain was the central feature in participants’ accounts of making decisions about activity: all described trying to maintain a balance between an acceptable level of daily activity and a tolerable or 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)

Exceeding this level of activity was predicted to precipitate a spiral of increasing pain, fatigue, disability and negative emotional consequences:

If the pain is reduced I feel my self levels going back up to how happy I’m feeling. So I know it’s all to do with the excruciating pain. It is just about trying to get the pain level down. (P10)

Although recognising that they could decide to push themselves beyond this point for the sake of a highly

valued goal, several participants expressed their reluctance to do so, and these participants used more catastrophic language and fearful terminology in describing their decision-making, were more concerned with loss, with ongoing deterioration in their condition or with the possibility of relapsing to a previous more disabled and distressed state:

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: is it worth it? Participants described weighing the value of the activity and the importance of doing it against the expected aversive consequences. Consideration of the consequences went beyond risk of pain or damage to health and disruption of immediate plans, to impact on other people and egodystonic (denoting aspects of a person’s thoughts, impulses and behaviour that are felt to be repugnant, distressing, unacceptable or inconsistent with the self-conception) outcomes of not living up to expectations, personal values and roles:

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’ accounts suggested that the decision inevitably involved conflicting pressures, or a cost in terms of pain if the activity was completed versus guilt if it was left undone:

It makes you feel really guilty and it makes you feel like you have let people down and it makes you feel like you constantly let people down. (P11)

Consideration was given to both the value of the activity itself and how engaging in the activity was ego-syntonic in terms of personal values and what was important to the person. In many cases, participants were able to articulate 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. (P7)

However, at times, an overriding desire to do something that was of high personal value would lead participants to bypass the weighing of costs and benefits,

resulting in a rather liberating decision to engage in the activity and deal with the consequences later, instead of anticipating them:

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)

Not only the activity itself but the sense of control it represented was highly valued, and this was most often expressed in two ways. One of these was feeling and/or seeming 'normal' (unrestricted) to others:

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)

The second was in demonstrating to themselves their determination and resilience:

I'd say there's nothing that I would let it stop me [from doing], because that's just the kind of person I am. (P2)

Theme 3: influence of pain intensity. Sometimes, in contrast to this careful deliberation, pain dominated and participants felt that they were simply responding to pain, not taking a decision. Decisions about activity were modulated by current intensity of pain, often expressed in terms of 'good' or 'bad' days. 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)

At higher current pain intensity, only an activity of paramount importance could outweigh it. Furthermore, a high level of pain resulted in a higher level of fatigue and lower energy and therefore less inclination to undertake the activity. On such occasions, participants described their bodies as autonomous and pain as a message of warning:

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)

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. (P3)

Theme 4: struggling with unpredictability. While participants could attach a particular value to an activity, the

unpredictability of pain and of their bodies' responses to the activity could not be accurately estimated:

As I've said from the start of this it's something inside my body and is just in there. It does what it wants to do. If it wants to have a bad day for three weeks it will. It sort of has its own mind. (P10)

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 decisions about future activities, constraining advance planning. There was implicit or explicit expression of anxiety in many of these statements but it was balanced against the other concerns articulated:

I don't know how I'm going to be in two or three days so I cannot, for example, [decide] to see my friends, because I don't know how I'm going to be in three days. I might be in pain. (P1)

Theme 5: exerting control. An important part of participants' attempts to address their uncertainty and the unpredictability of pain was ensuring that they felt able to exercise control over their pain. Considerations included the distance from help, options for escape from the situation and ability to manage pain:

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 extended to exercising control in a more global sense:

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)

Mental simulation helped participants consider the physical demands and expected consequences of the activity. For example,

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)

When participants could not calculate or visualise the activity in detail, they described a trial and error learning approach, recalling their previous experience of that activity:

This is another thing with this condition: it's not always instantly that you're going to get the flare. It'll be that

evening or the following day that you'll flare and so it's kind of like trial by error really. (P4)

In order to accomplish their daily goals while avoiding fatigue and incorporating rest periods, participants frequently paced their activities by doing things more slowly or in a different way and preparing themselves for planned activities:

If I've got something planned, I'd sort of try to take it easy so that I can go. (P5)

At times, participants' accounts suggested not only practical adaptations, but a change in the underlying assumptions pertaining to how they lived their lives and a reassessment of life priorities:

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)

Participants broke tasks down into smaller chunks that could be accomplished in a shorter time, involved briefer use of the same muscles, or spread tasks such as housework over a whole week instead of a day:

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)

Adaptations in work included retraining in a job that was more physically manageable, or reducing working hours; some stopped working completely. Some participants accepted pain as a normal part of existence, therefore adapting to it was not letting it limit their activities, but rather it was a positive choice, contributing to an increased sense of coping and control:

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 6: emotional cost of pain. Participants' accounts reflected an emotional cost of pain and a consequent impact on decision-making, in terms wider than fear and anxiety, or in terms of anxiety about issues beyond pain and harm. Fear and anxiety did affect decisions about activity, but were described in relation to specific activities, not as the pervasive dynamic underlying decisions:

I get really anxious before doing it because I know I will get a lot of pain, so I don't go hiking anymore. (P1)

Four participants spoke of fearing escalation of pain: two of these described very restricted activities, high levels of pain and a previous rapid deterioration in

physical condition. One of them had only 'bad days, extra bad or double extra bad days' [P6] and was afraid of worsening her current level of disability and distress through increasing pain. The other two participants had experienced such states but were now recovered, so they feared that making the wrong decisions could lead to 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 hinge on their expectation of coping with 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)

Participants described the emotional cost in terms of anger, depression and guilt arising from the limitations imposed on them by pain, not being able to do the things they wanted to and the losses (such as of career) suffered due to pain:

... and then you get angry, you get very angry, like you can't do this. It's like a bereavement, it's like I've lost someone. (P6)

There were also several accounts of frustration and anger directed towards the medical establishment, particularly over the difficulty obtaining a diagnosis and not knowing enough about how to treat the condition. There was also self-directed anger when they felt they had avoidably exacerbated pain and had to give up activities; guilt, depression and frustration were also expressed in relation to not being able to fulfil important roles:

If I'm having a flare up I can't cook a meal and that's quite depressing because then I have to get my eldest daughter to make a dinner, but then, it depresses me because I feel like I'm not doing my role as a mother. (P4)

Having to ask for help and depending on others, brought humiliation as well as guilt and frustration. Participants also described worry about not working, about money and about the consequences of pain.

Discussion

A total of 11 patients with JHS were interviewed to explore how it limited their activity, and their process of decision-making about activity limitation. The long duration of pain for most participants, their definitive

diagnosis and their opting for pain management, meant that they were largely focused on adaptation to chronic pain rather than expecting or hoping for pain remission or cure. In this way, they perhaps differ from many people with chronic pain who still seek and hope for complete resolution.

Our participants did describe fear about increased and unmanageable pain and (not without reason) about dislocation and subluxation of joints, or about other injuries that could further disable them. In relation to this, they also described deciding against particular activities, at a particular point or permanently. But their accounts corresponded more to the reformulation by Crombez et al.⁴ than to the classical fear-avoidance model, describing flexible and contextual decisions and balancing multiple competing goals. Fear was entirely absent from a substantial number of accounts that rather described a calm analysis of costs and benefits, consistent with the motivational formulation⁴ in which pain that is no more than expected and understood as unthreatening can be overridden in the pursuit of valued goals.

Even those participants who rated pain interference very high gave accounts of proceeding with activities when pain was not too bad, or of tackling it strategically, as well as of avoidance. Participants appeared to be attempting to maintain a fine balance between an acceptable level of daily activity and a manageable level of pain. This is consistent with the accounts of less highly selected groups of people with JHS⁹ and with the contention by Crombez et al.⁴ that pain-resilient individuals have found ways to balance activity and rest so as to minimise the physical toll of persistence. Factors influencing the decision were not consistent across activities, or from day to day, such as the importance of the activity, the current intensity of pain and the resources available to support possible outcomes. Risks and physical demands of the activity and the likelihood of aversive consequences were weighed up but occasionally for highly valued activities this was revised, in spite of anticipated aversive consequences.

The unpredictability of pain and the difficulty in relating physical consequences to specific activities (described in this population previously¹¹) made the decision more difficult. This was because, under these circumstances, both the likelihood of the consequences occurring and the intensity and duration of resulting pain were uncertain. Participants used experience from previous occasions as a guide where possible. We were unable to distinguish between safety behaviours and adaptive coping strategies⁶ since individuals' retrospective rationales for their decisions may have obscured the original intention and perceived function of their behaviours. Nor was reference made to following cautious (or other) advice from health-care professionals.

Only a minority of descriptions of decision-making around activity focused on fear or weighted it higher than all other factors. Largely, although fear of pain and dislocation were described, these were balanced against other factors in the equation, despite a degree of uncertainty that tends to exacerbate fear. This is consistent with a model within which pain expectancies intensify escape and avoidance tendencies,⁴ but not one in which they determine escape or avoidance.² In the fear and avoidance model,² catastrophic interpretation of the meaning of pain is pivotal in determining avoidance. However, the catastrophes anticipated by participants in this study were largely drawn from previous experience and were less influential in determining avoidance than careful recalculation of possible outcomes and resources to manage them. The value of the activity to the participant and concern about consequences for others should the participant experience a setback from the activity emerged as particularly important factors, both related mainly to role within the family (including companion animals) or wider social network,¹⁷ as exemplified particularly in Theme 5, Exerting Control. Current pain intensity also played an important part.

Strengths and limitations of study

This was a small sample of women attending specialist services for pain related to JHS, and it could be argued that the findings are specific to people who have to a great extent accepted that they have chronic pain; other findings might emerge from sampling people with JHS who were earlier in their journey. Participants were self-selected from those invited to take part and many stated that despite the anticipated increased pain (from travel and prolonged sitting), they opted in because of their strongly held beliefs that JHS was insufficiently understood by the public and by health-care professionals. It is possible that people with this orientation have achieved a way of managing their pain that they described accurately but that does not characterise those who did not participate. However, Table 1 shows a range of levels of pain and of interference by pain with life, indicating that we did not only sample those with low levels of pain or disability.

One characteristic of our sample that may be of particular importance is that all had (often after considerable delay) achieved a definitive diagnosis of JHS and felt that they were understood and had been taken seriously within this specialist service. Achieving a diagnosis, or even an explanation that makes sense of their experience – something that many people with chronic pain continue to pursue for many years – may facilitate cognitive and emotional adaptation that produces a shift from fear-based decision-making to a more cost-benefit

approach. Unfortunately, we have no contrasting population in which to test this hypothesis.

For participants, there is also the social desirability of presenting themselves as rational agents, but they were not unwilling to describe the influence of various emotions on their decisions. Their experience of pain, of unpredictability and their (not unrealistic) fears of unpleasant physical outcomes are highly reminiscent of accounts by people with chronic musculoskeletal pain of non-JHS origin,¹⁸ with the difference mainly in the specific risk of dislocations and subluxations for those with JHS. We would argue, however, that the focus on identifying and quantifying fear and avoidance and their correlates in people with chronic pain has produced an unnecessarily narrow vision of what motivates people with chronic pain in their daily lives and on the context in which fear and avoidance are balanced against other emotions and other options.

Qualitative studies are notoriously liable to bias from the researcher, so several credibility checks were designed and carried out to attempt to minimise bias. The main researcher (AS) was new to work in chronic pain so brought no particular preconceptions to her interviews, whose design could as easily have elicited an account of decisions driven by fear and avoidance as it elicited the current findings. The model (Figure 1) is very tentative, but developed as an attempt to represent the interaction of factors, rather than a linear process. It requires critical analysis of both components and connections.

Implications for clinical and research work

Our findings are consistent with models of everyday decision-making,¹⁹ influenced by preferences, values and emotions, although avoidance of losses may be counted more important than achievement of goals. We see this as a part of the canon of a normal psychology of pain and described in terms of normal psychology rather than psychopathology.²⁰ Our findings are particularly consistent with the motivational account of fear and avoidance⁴ that starts from a normal not a psychopathological viewpoint and emphasises ‘that the [fear-avoidance] model was never meant to be unconditionally embraced’ (p. 476).⁴ Rather, it recognises avoidance as one of several competing goals, decisions as contextual rather than trait-determined and pain interference with valued goals rather than pain intensity as the central determinant of patients’ lack of wellbeing.

If we only ask our patients and research participants about fear and avoidance, then we can only understand their behaviour in those terms. A wider context and a patient perspective should enable us to understand more of the variance in patients’ behaviour and emotion

than is explained by fear and avoidance. Although fear and anxiety in patients studied here 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 activity limitation, which indicated levels of complexity and individuality not expressed in the existing literature. We propose that decision-making theory offers useful concepts on understanding and mis/estimation of risk,¹⁹ even in conditions of uncertainty such as pertaining in chronic pain.

The constraints discussed above indicate the need for replication of these results, potentially in samples of chronic pain patients with different underlying pathology and diagnostic certainty, as well as further exploration of this decision-making process using quantitative methods, to test the generalisability of our findings.

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Declaration of Conflicting Interests

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