

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

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Abstract:	<p>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.</p> <p>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).</p> <p>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.</p> <p>Conclusions 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.</p>

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4 **syndrome make decisions about activity?**
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For Peer Review

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

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.

Conclusions 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.](#) ~~but decisions explored in a broader context.~~

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 overapplied 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.

Introduction

The fear and avoidance model of chronic pain introduced by Vlaeyen and colleagues¹ and reformulated since^{2,3} is widely adopted with plentiful data supporting the links between anxiety about pain and damage and (self-reported) avoidance³. 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 published⁴; and the misleading nature of analogies with phobia given the major differences in presentation and focus^{3,4}. ~~Recent reviews have raised concern about the scarcity of tests of some of its assumptions, and some inconsistent findings⁴, acknowledging that analogies with phobia are misleading given the major differences in presentation and focus³.~~ Further, although avoidance is widely observed and reported, associated predictions of disuse have not been consistently supported⁵. Crombez and colleagues in a critical reformulation⁴ 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

1
2
3 or reported, be associated with motivations other than fear? Findings of association
4
5 between (self-reported) avoidance and greater disability are at a broad level and do not
6
7 necessarily pertain to

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9
10 (1) the decision to avoid a particular activity;
11
12 (2) the consistency of such decisions within and across activities within an individual;
13
14 (3) ~~to what~~ the extent to which avoidance is associated with strong expectation of
15
16 immediate pain or damage rather than with moderate estimates of cumulative risk;
17
18 the former is more easily challenged.
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24 There are several further models applicable to avoidance. One is safety behaviours,
25
26 behavioural or cognitive attempts to alleviate threat thus preventing disconfirmation of
27
28 fearful beliefs⁶, but safety behaviours can be difficult to distinguish from adaptive coping.
29
30 Another is that apparent avoidance may be an attempt to follow poor advice from
31
32 healthcare professionals⁷. Third, persistence despite pain can be counterproductive^{3,4}. We
33
34 were interested in how people with chronic pain described their decisions about attempting
35
36 or avoiding potentially pain-exacerbating activities.
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43 Joint Hypermobility Syndrome (JHS) is a musculoskeletal disorder thought to be a
44
45 subtype of Ehlers-Danlos Syndrome (EDS), an inherited disorder of connective tissue
46
47 protein characterized by varying degrees of joint hypermobility and increased risk of
48
49 subluxation and dislocation⁸. It is not universally recognised in rheumatological
50
51 medicine, and diagnosis is often delayed^{8,9}. Persistent pain in people with JHS is
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53 relatively common¹⁰, affecting joints, muscles and ligaments⁹ and thereby a range of
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55 everyday activities¹¹.
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5 This study aimed to explore, in relation to the models of fear and avoidance and of problem-
6 solving, how JHS patients accounted for the decision to avoid or not to avoid activities
7
8 which that were anticipated to increase pain or to risk dislocation ~~in relation to the models of~~
9 ~~fear and avoidance and of problem-solving~~. Since the aim of this study was to understand
10 the complex processes involved in decision-making, a phenomenological approach was
11 adopted to understand people's perceptions of their experiences, the meanings they attach
12 to their experiences and their underlying assumptions¹².
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25 **Methods**

26 *Participants*

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28 JHS patients were all referred from the same specialist rheumatologist and so constituted a
29 more homogeneous group than any of our other chronic pain patients. Further, their
30 understanding of their diagnosis gave them no expectations of finding a way of abolishing
31 pain, but of adapting as well as possible to a lifelong problem. Patients eligible for
32 participation in this study ~~Eligible patients~~ had been referred by the specialist rheumatologist
33 to the pain management programme. This programme, ~~which~~ shows good outcomes¹³,
34 including reduced interference of pain with everyday life.
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48 Inclusion criteria for this study were pain lasting at least 6 months, diagnosis with JHS by the
49 specialist rheumatologist, ability to attend a face-to-face interview and fluency in English;
50 exclusion criteria were serious psychiatric impairment or substance misuse/abuse problems
51 or other general health condition limiting activity. Thirty-two patients (31 of whom were
52 women) were invited to take part by letter containing information about the study; 15
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3 agreed but four were unable to attend appointments so eleven women (34% of those
4
5 eligible) were interviewed. Ten were white and one was Black British; their ages ranged from
6
7 22 to 55 years, with a mean of 34 years. Age of symptom onset ranged from infancy to 45
8
9 years, with a mean age of onset of 16 years (see Table 1). All but one were using medication
10
11 for pain relief; four had previously had some psychological intervention for their pain.
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17 *[Table 1 about here]*
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21

22 *Procedure*

23
24 Ethical approval was obtained from the local Research Ethics Committee. Written informed
25
26 consent was obtained from each participant. The semi-structured interview lasted
27
28 approximately one and a half hours and was audio-recorded. Interviews were conducted by
29
30 AS (main researcher) using guidelines by Smith and Osborn¹⁴. The first part of the interview
31
32 covered current pain, pain history and life changes due to pain and established a context for
33
34 participants' more detailed descriptions of their experience and understanding of pain in the
35
36 second part of the interview. The latter covered ~~and covered current pain, pain history and~~
37
38 ~~life changes due to pain, all as context,~~ how ~~the~~ participant~~s~~ decided what was and was not
39
40 safe to do and what ~~they~~/~~he~~ avoided; the outcomes of avoidance or of persisting with the
41
42 activity; advice recalled about activity; and a detailed account of a recent decision to avoid
43
44 an activity.
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51
52 To provide some context of pain and disability using familiar measures, participants were
53
54 asked to complete the Brief Pain Inventory (short form) (BPI¹⁵). This assesses pain severity (4
55
56 items: pain at worst, at least, on average and now) and the impact of pain on activity (7
57
58
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3 items). All items are rated from 0 (= no pain/does not interfere) to 10 (= pain as bad as you
4
5 can imagine/completely interferes). Reliability (Cronbach's α) in non-cancer chronic pain
6
7 patients is greater than 0.70; the original factor structure has been replicated and the
8
9 relationship of the BPI to generic measures of pain is strong¹⁶.
10
11

12 13 14 *Interpretative phenomenological analysis*

15
16 Interpretative Phenomenological Analysis (IPA)¹⁴ is described as phenomenological in that it
17
18 is concerned with personal perception or accounts of an object or event, rather than a quasi-
19
20 objective statement about it; it is interpretative in that analysis of responses is necessarily
21
22 influenced by the researcher's own view of the world as well as the nature of the interaction
23
24 between researcher and participant¹². The data analyst's (AS's) theoretical orientation was
25
26 cognitive behavioural, with a particular interest in participants' cognitions and emotions and
27
28 their effects on behaviour; she had recently begun work in chronic pain management and
29
30 had no strong views on application of the fear and avoidance model. The last author (AW)
31
32 had extensive clinical experience in pain management and had concerns about over-
33
34 application of that model.
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43 All interviews were transcribed verbatim and analysed following IPA methods¹⁴. Each
44
45 interview was examined in detail before reading the next, annotating statements that were
46
47 significant in relation to the research questions. These annotations were summarised as the
48
49 main ideas, then grouped into themes according to theoretical similarities and apparent
50
51 connections between them, with particular reference to the research questions to which
52
53 they pertained. These themes were checked against the transcript to ensure that the
54
55 researcher's interpretation represented what was said. Themes from earlier transcripts were
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3 used to inform analysis of subsequent transcripts, while allowing new themes to emerge.

4
5 Once a final list of themes was constructed, all transcripts were re-read, and a brief narrative
6
7 synopsis ~~that~~ ~~which~~ focused on the research questions was produced for each transcript. The
8
9 list of themes was checked against each synopsis to ensure that it adequately captured the
10
11 content of each transcript.
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17
18 Credibility checks were carried out as follows. Identification and representation of themes ~~by~~
19
20 AS ~~were~~ ~~was~~ discussed with AW at several stages of analysis. ~~The~~ ; results were checked ~~by~~
21
22 AW against transcripts to ensure accurately ~~ly~~ representation of data. ~~A~~ ; a blind derivation of
23
24 themes ~~by~~ KC was compared with the existing themes and all three authors agreed on a
25
26 composite version. Testimonial validity was assessed by checking the results with the
27
28 original participants; all four who responded agreed with the findings, did not suggest any
29
30 changes and two expressed surprise at how similar the other participants' experiences ~~s~~
31
32 ~~were~~ ~~was~~ to their own.
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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 limiting 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 whichthat 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 whichthat it would cause, as well as the consequences of that pain. In particular, participants needed to estimate the steps whichthat would be required to alleviate the pain, such as bed-rest for an uncertain number of days, the activities whichthat would be missed as a result, the

1
2
3 consequent effects on people within their network of friends and family and the possible
4
5 potential long term deleterious effects on their health. Balanced against this consideration
6
7 was the importance ~~which~~that the participant attached to the particular activity: the value of
8
9 the activity itself, and how completing the activity was egosyntonic¹ in terms of personal
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11 importance. Activities ~~which~~that were considered necessary, urgent, or to which the
12
13 participant attached high personal value were given greater weight.
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19 For some participants this appeared to be an implicit process, described in terms of “just
20
21 knowing” from bodily experience, while for others it appeared to be an explicit process in
22
23 which the nature of the activity, its physical demands, risks and likely consequences were
24
25 carefully considered. Although these factors were taken into account, at times physical
26
27 factors overrode the decision-making process and pain dominated the decision.
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33 The estimate of consequences was made difficult by the unpredictability of pain and the lack
34
35 of a clear relationship between activity and pain. Attempts to solve the problems of keeping
36
37 pain to a manageable level, and of its unpredictability, included careful planning, learning
38
39 from experience, use of coping strategies, pacing and adapting and doing things differently
40
41 or doing different things. Reaching an acceptance of pain and its limitations enabled
42
43 participants to make a positive change in their underlying assumptions about how
44
45 ~~participants~~they lived their lives.
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52 Participants also spoke about the emotional cost of pain and impact on decision-making, in
53
54 terms not just of fear and anxiety, but also depression, anger, frustration, guilt and
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57
58 ¹ [Acceptable and consistent with personality and beliefs](#)
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1
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3 humiliation. Rather than describe explicit serial steps, weighing costs and benefits,
4
5 participants described a process in which their decisions were influenced by existing
6
7 conditions (current intensity of pain), uncertainty and unpredictability, as well as
8
9 considerations of expected emotional cost. The interaction of these themes is illustrated in
10
11 Figure 1.
12
13

14
15 *[Figure 1 about here]*
16

17 Theme 1: Keeping pain at a manageable level

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19 Pain was the central feature in participants' accounts of making decisions about activity: all
20
21 described trying to maintain a balance between an acceptable level of daily activity and a
22
23 tolerable or manageable level of pain.
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25

26
27 "If I can keep my pain at a level where, you know, it's manageable, I can get on
28
29 with a certain amount of pain, that's fine." [P4]
30
31

32 Exceeding this level of activity was predicted to precipitate a spiral of increasing pain,
33
34 fatigue, disability and negative emotional consequences.
35
36

37 "If the pain is reduced I feel myself levels going back up to how happy I'm feeling.
38
39 So I know it's all to do with the excruciating pain. It is just about trying to get the
40
41 pain level down!~~It's like you get a level and you know this level if you do too much~~
42
43 ~~the pain is going to get really worse, and there's a level where you know you can~~
44
45 ~~do things and it's not going to be worse and you can carry on."~~ [P10]
46
47

48
49 Although recognising that they could decide to push themselves beyond this point for the
50
51 sake of a highly valued goal, several participants expressed their reluctance to do so and
52
53 these participants used more catastrophic language and fearful terminology in describing
54
55 their decision-making, were more concerned with loss, with ongoing deterioration in their
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1
2
3 condition, or with the possibility of relapsing to a previous more disabled and distressed
4
5 state.
6

7
8 “Sometimes when you hurt a lot you are scared that it won't go away. When I
9
10 first did my back in it was horrible. I was so scared I was going to be like that,
11
12 that was going to be it. Then you start thinking about all the things you can't do
13
14 and you get really anxious.” [P11]
15
16

17 18 19 Theme 2 Is it worth it? 20

21
22 Participants described weighing the value of the activity and the importance of doing it
23
24 against the expected aversive consequences. Consideration of the consequences went
25
26 beyond risk of pain or damage to health and disruption of immediate plans, to impact on
27
28 other people and egodystonic² outcomes of not living up to expectations, personal values
29
30 and roles.
31
32

33
34 “Something that is potentially high risk of dislocation then it's just not worth
35
36 doing it, because then you got to take someone's time getting you to the
37
38 hospital, so they've got to stop doing what they want to be doing, you got to
39
40 waste someone's time the next day looking after me and the baby. It's just not
41
42 worth it, so you just don't do it.” [P5]
43
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45
46 Participants' accounts suggested that the decision inevitably involved conflicting pressures,
47
48 or a cost in terms of pain if the activity was completed versus guilt if it was left undone.
49

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51 “It makes you feel really guilty and it makes you feel like you have let people down and
52
53 it makes you feel like you constantly let people down.” [P11]
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58 ² [denoting aspects of a person's thoughts, impulses, and behavior that are felt to be repugnant, distressing, unacceptable, or inconsistent with the self-conception](#)
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3 Consideration was given to both the value of the activity itself and how engaging in the
4
5 activity was egosyntonic in terms of personal values and what was important to the person.
6
7 In many cases participants were able to articulate their weighing of the importance of the
8
9 activities.
10

11
12 “I have things, you know, different levels of importance. Me missing a meal isn’t
13
14 important, but the dogs have to be sorted out, but the ironing doesn’t have to be
15
16 done, it’s not important.” [P7]
17

18
19 However, at times, an overriding desire to do something ~~which that that~~ was of high personal
20
21 value would lead participants to bypass the weighing of costs and benefits, resulting in a
22
23 rather liberating decision to engage in the activity and deal with the consequences later,
24
25 instead of anticipating them.
26
27

28
29 “I mean, some things are worth it. If it’s something I really want to do, then I just
30
31 do it. You think, I’m going to feel that tomorrow, but I want it.” [P5]
32

33
34 Not only the activity itself but the sense of control it represented were highly valued, and
35
36 this was most often expressed in two ways. One of these was feeling and/or seeming
37
38 “normal” (unrestricted) to others.
39

40
41 “When I go out when I’m seen by other people, I’m trying to do things like the
42
43 others so I try, I want people to see me like normal.” [P1]
44

45
46 The second was in demonstrating to themselves their determination and resilience.
47

48
49 “I’d say there’s nothing that I would let it stop me [from doing], because that’s
50
51 just the kind of person I am.” [P2]
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55 Theme 3: Influence of pain intensity
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3 Sometimes, in contrast to this careful deliberation, pain dominated and participants felt that
4
5 they were simply responding to pain, not taking a decision. ~~At a lesser level of pain,~~
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7
8 ~~Decisions~~ about activity were modulated by current intensity of pain, often expressed in
9
10 terms of “good” or “bad” days. For example, when pain levels were high:

11
12 “Well it’s easy when I’m flaring because there’s no choice, because the pain’s so
13
14 bad, I just can’t do it.” [P4]

15
16
17 Similarly, when pain levels were low:

18
19 “I would feel there is nothing wrong with me on a good day. I can get up and do
20
21 what I like. Just carry on, carry on as if there’s no tomorrow.” [P8]

22
23
24 At higher current pain intensity, only an activity of paramount importance could outweigh it.
25
26 Furthermore, a high level of pain resulted in a higher level of fatigue and lower energy and
27
28 therefore less inclination to undertake the activity. On such occasions participants described
29
30 their bodies as autonomous and pain as a message of warning.

31
32
33 “I avoid doing totally anything at the moment, ‘cos anything will bring it on and it
34
35 doesn’t allow me. It doesn’t allow me.” [P6]

36
37
38 “The pain is the way of my body trying to tell me that I’m doing something wrong or
39
40 I’m hurting something so I have to lessen it or stop.” [P3]

41 42 43 44 45 Theme 4: Struggling with unpredictability

46
47 While participants could attach a particular value to an activity, the unpredictability of
48
49 pain and of their bodies’ responses to the activity could not be accurately estimated.

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51
52
53 ~~“As I've said from the start of this it's something inside my body and is just in there. It~~
54 ~~does what it wants to do. If it wants to have a bad day for three weeks it will. It sort of~~
55 ~~has its own mind. I know I have to do it, I'm going to do it, but I always wonder if I'm~~
56 ~~going to be able, and I know I'm going to be able, but how much pain am I going to get~~
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3 out of it, I'm going to go through, and how many days problems I'm going to have
4 after?" -[P10]
5
6
7

8
9 "I mean I could do something and there'll be no repercussion that time and I
10 could do something again and there will be a repercussion. There doesn't seem
11 to be any pattern." [P7]
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16 This unpredictability affected decisions about future activities, constraining advance
17 planning. There was implicit or explicit expression of anxiety in many of these
18 statements but it was balanced against the other concerns articulated.
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21
22

23 "I don't know how I'm going to be in 2 or 3 days so I cannot, for example,
24 [decide] to see my friends, because I don't know how I'm going to be in 3 days. I
25 might be in pain." [P1]
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33 Theme 5: Exerting control

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35 An important part of participants' attempts to address their uncertainty and the
36 unpredictability of pain was ensuring that they felt able to exercise control over their pain.
37 Considerations included the distance from help, options for escape from the situation and
38 ability to manage pain.
39
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44 "If it's something, say, round the corner or something short, where I know I can leave
45 and go home and relieve the pain or something, then I do [it]." [P3]
46
47
48

49 This need for control extended to exercising control in a more global sense.

50
51 "I like to be able to be in control of what I do. It's important to me. I don't want
52 to knock myself out and spend two days in bed and have the children come in
53 and see me and go away thinking that mum's really ill." [P8]
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3 Mental simulation helped participants consider the physical demands and expected
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5 consequences of the activity, for example,
6

7
8 “Walking the dogs I have to be careful where I walk them, what I do, whether the
9
10 ground’s level, is it a route that I know, just because I have to be really aware of
11
12 my surroundings.” [P7]
13

14
15 When participants could not calculate or visualise the activity in detail, they described
16
17 a trial and error learning approach and recalling their previous experience of that
18
19 activity.
20

21
22 “This is another thing with this condition: it’s not always instantly that you’re
23
24 going to get the flare. It’ll be that evening or the following day that you’ll flare
25
26 and so it’s kind of like trial by error really.” [P4]
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28

29
30 In order to accomplish their daily goals while avoiding fatigue and incorporating rest
31
32 periods, participants frequently paced their activities by doing things more slowly or in
33
34 a different way and preparing themselves for planned activities.
35

36
37 “If I’ve got something planned, I’d sort of try to take it easy so that I can go.”
38

39
40 [P5]

41
42 At times, participants’ accounts suggested not only practical adaptations, but a change
43
44 in the underlying assumptions pertaining to how they lived their lives, and a
45
46 reassessment of life priorities.
47

48
49 “So it’s got to the stage where it’s yes, it’s dusty, it’s going to get dusty and it can
50
51 stay dusty. But that was never me.” [P4]

52
53 Participants broke tasks down into smaller chunks ~~which~~that could be accomplished in a
54
55 shorter time, involved briefer use of the same muscles, or spread tasks such as housework
56
57 over a whole week instead of a day.
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1
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3 “So I can’t, I won’t be able to do something throughout, I have to sort of break it up
4
5 into pieces and do it bit by bit by bit.” [P3]
6

7 Adaptations in work included retraining in a job that was more physically manageable, or
8
9 reducing working hours; some stopped working completely. Some participants accepted
10
11 pain as a normal part of existence, therefore adapting to it was not letting it limit their
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13 activities, but rather it was a positive choice, contributing to an increased sense of coping
14
15 and control.
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19 “I have to phrase it as things that I avoid in order to explain to other people, but
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21 in fact it’s how I choose it.” [P9]
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24 25 26 Theme 6: Emotional cost of pain 27

28 Participants’ accounts reflected an emotional cost of pain and a consequent impact on
29
30 decision-making, in terms wider than fear and anxiety, or in terms of anxiety about issues
31
32 beyond pain and harm. Fear and anxiety did affect decisions about activity, but were
33
34 described in relation to specific activities, not as the pervasive dynamic underlying decisions.
35
36

37
38 “I get really anxious before doing it because I know I will get a lot of pain, so I
39
40 don’t go hiking anymore.” [P1]
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42

43 Four participants spoke of fearing escalation of pain: two of these described very restricted
44
45 activities, high levels of pain and a previous rapid deterioration in physical condition. One of
46
47 them had only “bad days, extra bad or double extra bad days” [P6] and was afraid of
48
49 worsening her current level of disability and distress through increasing pain. The other two
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51 participants had experienced such states but were now recovered, so they feared that
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53 making the wrong decisions could lead to regression to that state.
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3 “Yeah I’m always scared when I go back into big heavy pain because it always
4
5 reminds me how I was before and I always get scared that I’ll get like that, back
6
7 like that.” [P1]
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10 Other participants’ anxiety seemed to hinge on their expectation of coping with particular
11
12 situations.
13

14 “I get a bit anxious. What if I pop [dislocate], or what if I fall? If people do help
15
16 you, you’ve then got to try and explain to them, why you’re on the deck, why
17
18 you’re on the floor in the first place.” [P5]
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21

22 Participants described the emotional cost in terms of anger, depression and guilt arising
23
24 from the limitations imposed on them by pain, not being able to do the things they wanted
25
26 to and the losses (such as of career) suffered due to pain.
27

28
29 “...and then you get angry, you get very angry, like you can’t do this. It’s like a
30
31 bereavement, it’s like I’ve lost someone.” [P6]
32
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34 There were also several accounts of frustration and anger directed towards the medical
35
36 establishment, particularly over the difficulty obtaining a diagnosis and not knowing enough
37
38 about how to treat the condition. There was also self-directed anger when they felt they had
39
40 avoidably exacerbated pain and had to give up activities; guilt, depression and frustration
41
42 were also expressed in relation to not being able to fulfil important roles.
43
44

45 “If I’m having a flare up I can’t cook a meal and that’s quite depressing because
46
47 then I have to get my eldest daughter to make a dinner, but then, it depresses
48
49 me because I feel like I’m not doing my role as a mother.” [P4]
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52 Having to ask for help and depending on others, brought humiliation as well as guilt and
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54 frustration. Participants also described worry about not working, about money and about
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56 the consequences of pain.
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For Peer Review

Discussion

Eleven 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 ~~which~~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 and colleagues⁴ were than to the classical fear-avoidance model, describing much more flexible and contextual decisions and, not least in counterbalancing multiple competing goals, than described in the classical fear-avoidance model, corresponding more to the reformulation by Crombez and colleagues⁴. F and fear was entirely absent from a substantial number of accounts ~~which~~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

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2
3 acceptable level of daily activity and a manageable level of pain. This is consistent with the
4
5 accounts of less highly selected groups of people with JHS⁹ and with the contention by
6
7 Crombez and colleagues⁴ that pain-resilient individuals have found ways to balance activity
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9 and rest so as to minimize the physical toll of persistence. Factors influencing the decision
10
11 were not consistent across activities, or from day to day, such as the importance of the
12
13 activity, the current intensity of pain and the resources available to support possible
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15 outcomes. Risks and physical demands of the activity and the likelihood of aversive
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17 consequences were weighed up, but occasionally for highly valued activities, this was revised
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21 in spite of anticipated aversive consequences.
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26 The unpredictability of pain and the difficulty in relating physical consequences to specific
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28 activities (described in this population previously¹¹) made the decision more difficult. This
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30 was because, under these circumstances, both the likelihood of the consequences occurring
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32 and the intensity and duration of resulting pain were uncertain. ~~The unpredictability of pain,~~
33
34 ~~and the difficulty in relating physical consequences to specific activities, described in this~~
35
36 ~~population previously¹¹, meant that under these circumstances, both the likelihood of the~~
37
38 ~~consequences occurring and the intensity and duration of the pain were uncertain, making~~
39
40 ~~the decision more difficult.~~ Participants used experience from previous occasions as a guide
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42 where possible. We were unable to distinguish between safety behaviours and adaptive
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44 coping strategies⁶ as since individuals' retrospective rationales for their decisions may have
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46 obscured the original intention and perceived function of their behaviours. ~~which may be~~
47
48 ~~obscured by retrospective representation of decisions, and adaptive coping strategies⁶.~~ Nor
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55 was reference made to following cautious (or other) advice from healthcare professionals.
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3 | Thus ~~o~~Only a minority of descriptions of decision-making around activity focussed on fear or
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5 weighted it higher than all other factors. Largely, although fear of pain and dislocation were
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7 described, these were balanced against other factors in the equation, despite a degree of
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10 | uncertainty ~~which~~that tends to exacerbate fear. This is consistent with a model within which
11
12 pain expectancies intensify escape and avoidance tendencies⁴, but not one in which they
13
14 determine escape or avoidance². In the fear and avoidance model², catastrophic
15
16 interpretation of the meaning of pain is pivotal in determining avoidance. However, the
17
18 catastrophes anticipated by participants in this study were largely drawn from previous
19
20 experience and were less influential in determining avoidance than careful recalculation of
21
22 possible outcomes and resources to manage them. The value of the activity to the
23
24 participant and concern about consequences for others should the participant experience a
25
26 setback from the activity emerged as particularly important factors, both related mainly to
27
28 role within the family (including companion animals) or wider social network¹⁷, as
29
30 exemplified particularly in theme 5, Exerting Control. Current pain intensity also played an
31
32 important part.
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41 *Strengths and limitations of study*

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43 This was a small sample of women attending specialist services for pain related to JHS and it
44
45 could be argued that the findings are specific to people who have to a great extent accepted
46
47 that they have chronic pain; other findings might emerge from sampling people with JHS
48
49 who were earlier in their journey. Participants were self-selected from those invited to take
50
51 part and many stated that despite the anticipated increased pain (from travel and prolonged
52
53 sitting), they opted in because of their strongly held beliefs that JHS was insufficiently
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55 understood by the public and by healthcare professionals. It is possible that people with this
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3 orientation have achieved a way of managing their pain which that they described accurately
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5 but which that does not characterise those who did not participate. However, Table 1 shows
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7 a range of levels of pain and of interference by pain with life, indicating that we did not only
8
9 sample those with low levels of pain or disability.
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17 One characteristic of our sample which that may be of particular importance is that all had
18
19 (often after considerable delay) achieved a definitive diagnosis of JHS and felt that they were
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21 understood and had been taken seriously within this specialist service. Achieving a diagnosis,
22
23 or even an explanation which that makes sense of their experience – something which that
24
25 many people with chronic pain continue to pursue for many years – may facilitate cognitive
26
27 and emotional adaptation which that produces a shift from fear-based decision-making to a
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29 more cost-benefit approach. Unfortunately, we have no contrasting population in which to
30
31 test this hypothesis.
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38 For participants, there is also the social desirability of presenting themselves as rational
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40 agents, but they were not unwilling to describe the influence of various emotions on their
41
42 decisions. Their experience of pain, of unpredictability and their (not unrealistic) fears of
43
44 unpleasant physical outcomes are highly reminiscent of accounts by people with chronic
45
46 musculoskeletal pain of non-JHS origin¹⁸, with the difference mainly in the specific risk of
47
48 dislocations and subluxations for those with JHS. We would argue, however, that the focus
49
50 on identifying and quantifying fear and avoidance and their correlates in people with chronic
51
52 pain has produced an unnecessarily narrow vision of what motivates people with chronic
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3 pain in their daily lives and on the context in which fear and avoidance are balanced against
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5 other emotions and other options.
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10 Qualitative studies are notoriously liable to bias from the researcher, so several credibility
11
12 checks were designed and carried out to attempt to minimise bias. The main researcher (AS)
13
14 was new to work in chronic pain so brought no particular preconceptions to her interviews,
15
16 whose design could as easily have elicited an account of decisions driven by fear and
17
18 avoidance as it elicited the current findings. The model (Figure 1) is very tentative, but
19
20 developed as an attempt to represent the interaction of factors, rather than a linear process.
21
22 It requires critical analysis of both components and connections.
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29 *Implications for clinical and research work*

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31 Our findings are consistent with models of everyday decision-making¹⁹, influenced by
32
33 preferences, values and emotions, although avoidance of losses may be counted more
34
35 important than achievement of goals. We see this as a part of the canon of a normal
36
37 psychology of pain and described in terms of normal psychology rather than
38
39 psychopathology²⁰. Our findings are particularly consistent with the motivational
40
41 account of fear and avoidance⁴ ~~which that that was published after we started writing up this~~
42
43 ~~study: it~~ starts from a normal not a psychopathological viewpoint and emphasises “that the
44
45 [fear-avoidance] model was never meant to be unconditionally embraced”⁴ (p476). Rather, it
46
47 recognises avoidance as one of several competing goals, decisions as contextual rather than
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49 trait-determined and pain interference with valued goals rather than pain intensity as the
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51 central determinant of patients’ lack of wellbeing.
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3 If we only ask our patients and research participants about fear and avoidance, then we can
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5 only understand their behaviour in those terms. A wider context and a patient perspective
6
7 should enable us to understand more of the variance in patients' behaviour and emotion
8
9 than is explained by fear and avoidance. Although fear and anxiety in patients studied here
10
11 did appear to influence decisions about activity, in a specific manner, they did not drive the
12
13 decision-making process as suggested by existing fear-avoidance models. The use of a
14
15 qualitative method enabled participants to give their perspective on activity limitation,
16
17 which indicated levels of complexity and individuality not expressed in the existing literature.
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19 ~~and~~ We propose that decision-making theory offers useful concepts on understanding and
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21 mis/estimation of risk¹⁹, even in conditions of uncertainty such as pertain in chronic pain.
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29 The constraints discussed above indicate the need for replication of these results, potentially
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31 in samples of chronic pain patients with different underlying pathology and diagnostic
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33 certainty, as well as further exploration of this decision-making process in large using
34
35 quantitative studies methods, to enable test the generalisability of the our findings to be
36
37 established.
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43 **Disclosures**

44 There was no funding source or sponsorship.

45 The authors have no conflicts of interest to declare.

50 Interview schedule

51 The interview schedule is available on request from the corresponding author.
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Legends

Figure 1: The process of decision making

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

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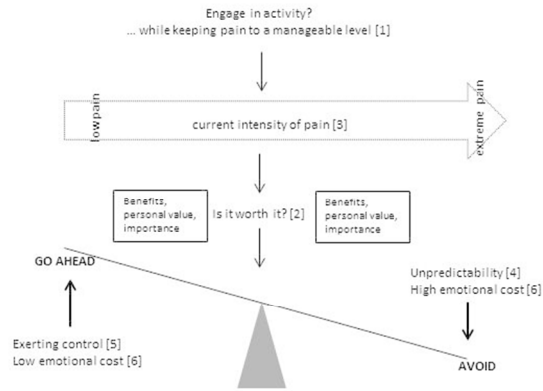
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Figure 1: The process of decision making

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

For Peer Review

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254x190mm (96 x 96 DPI)

Review

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

	Age band	Age at symptom onset, at diagnosis	Highest level of education	Employed	Marital status & children	BPI pain severity	BPI pain interference	Analgesic
P6	40-49	6, 39	School	No	Married + children	7.8	9.7	Compound ¹ analgesics
P3	20-29	7, 22	School	Yes	Single	8.0	9.1	NSAID ²
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 ¹
P7	30-39	12, 28	School	Yes	Single	5.8	4.9	Opioid

¹ Contain multiple active ingredients

² Non-steroidal Anti-inflammatory Drug

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3	P8	50-59	45, 53	College/University	No	Married + children	6.8	3.0	Paracetamol
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5	P5	20-29	1, 20	College/University	No	Cohabiting + children	3.8	2.3	Compound
6									
7									analgesic
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9									
10	P2	20-29	14, 18	College/University	Yes	Single	5.3	1.6	Cocodamol
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