



Body perception disturbance: A contribution to pain in complex regional pain syndrome (CRPS)

Jennifer S. Lewis ^{a,b,*}, Paula Kersten ^b, Candida S. McCabe ^{a,c},
Kathryn M. McPherson ^d, David R. Blake ^{a,c}

^a The Royal National Hospital for Rheumatic Diseases NHS Foundation Trust, Bath, UK

^b The School of Health Professions and Rehabilitation Sciences, University of Southampton, Southampton, UK

^c School for Health, University of Bath, Bath, UK

^d Auckland University of Technology, Auckland, New Zealand

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Abstract

In spite of pain in the CRPS limb, clinical observations show patients pay little attention to, and fail to care for, their affected limb as if it were not part of their body. Literature describes this phenomenon in terms of neurological neglect-like symptoms. This qualitative study sought to explore the nature of the phenomenon with a view to providing insights into central mechanisms and the relationship with pain. Twenty-seven participants who met the IASP CRPS classification were interviewed using qualitative methods to explore feelings and perceptions about their affected body parts. These semi-structured interviews were analysed utilising principles of grounded theory. Participants revealed bizarre perceptions about a part of their body and expressed a desperate desire to amputate this part despite the prospect of further pain and functional loss. A mismatch was experienced between the sensation of the limb and how it looked. Anatomical parts of the CRPS limb were erased in mental representations of the affected area. Pain generated a raised consciousness of the limb yet there was a lack of awareness as to its position. These feelings were about the CRPS limb only as the remaining unaffected body was felt to be normal. Findings suggest that there is a complex interaction between pain, disturbances in body perception and central remapping. Clinically, findings support the use of treatments that target cortical areas, which may reduce body perception disturbance and pain. We propose that body perception disturbance is a more appropriate term than 'neglect-like' symptoms to describe this phenomenon.

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1. Introduction

Pain in a limb is the cardinal symptom of complex regional pain syndrome (CRPS). One could assume therefore that patients might be overattentive, wishing to protect or look after their affected limb. However,

both clinical observations and recent literature (Galer et al., 1995; Galer and Jensen, 1999; Lewis et al., 2003) suggest that this is not the case. Patients have been reported to neglect their affected limb despite the pain. They do not attend to their limb often positioning it in such a way that it is outside of their field of view (Lewis et al., 2003). Galer et al. (1995, 1999) proposed that this phenomenon was similar in form to neurological neglect. Some consider their affected hand as foreign or strange (Forderreuther et al., 2004) and larger than it really is (Moseley, 2005). It is clear from these

* Corresponding author. Address: The Royal National Hospital for Rheumatic Diseases NHS Foundation Trust, Upper Borough Walls, Bath BA1 1RI, UK. Tel.: +441225787043; fax: +441225463223.

E-mail address: Jenny.lewis@rnhrd-tr.swest.nhs.uk (J.S. Lewis).

reported observations that patient's behaviour towards their affected limb is altered but quite how an individual with CRPS perceives their affected body parts is not well understood.

Recent brain imaging studies (Maihofner et al., 2003; Pleger et al., 2005) have shown that those with CRPS have a disrupted cortical map of the affected body part. An association between symptom resolution over time and the corrective reorganisation of cortical limb representation suggests that there is a relationship between the amount of pain and the degree of cortical disorganisation (Flor, 2003; Maihofner et al., 2004; Pleger et al., 2005). The existence of these disturbed representations could serve to influence the pain experience and alter how an individual may perceive their CRPS limb. In order to better understand these representations and their impact, it is important to know how people with CRPS actually perceive their own affected body parts. A review of the literature suggests that to date, this perspective remains poorly researched and poorly understood. We suggest a greater knowledge of body perception in CRPS may provide valuable insights into central mechanisms and the relationship with pain. In addition, given that CRPS is known to be resistive to conventional treatments, an improved understanding of body perception in relation to pain may identify areas in which interventions could be specifically targeted.

To achieve these objectives, we set out to explore the patients' experience of body perception in CRPS, to postulate a theoretical framework for understanding the characteristics of body perception disturbance and how it may contribute to the pain experience. Finally, we discuss how these findings may inform clinical practice.

2. Methods

Given the inductive nature of this research (i.e. hypothesis deriving not hypothesis testing), a qualitative methodology, drawing on principles of grounded theory, was utilised. This approach, previously used in the pain field, is of particular value in exploring areas such as this, where little research has been undertaken and knowledge is limited (Flick, 2002). Just as case studies are used to inform and develop hypotheses, a strength of grounded theory is that it aims to generate theory from the data from which new hypotheses can be made. Emergent theory is not merely descriptive but seeks to identify key relationships within and between both existing and newly proposed theory in order to enhance our understanding (Glaser, 1969; Strauss and Corbin, 1998; Seale, 1999; Stanley and Cheek, 2003).

2.1. Participants and data collection

Adult participants who met The International Association of the Study of Pain classification criteria for CRPS Type I and II (Stanton-Hicks et al., 1995) were drawn from a UK population.

Additional inclusion criteria were the capability to verbally communicate and the absence of co-morbidity such as diabetic neuropathy which may significantly influence their experience. Recruitment sources included a national CRPS referral hospital, RSD-UK, the United Kingdom CRPS patient charity, pain clinics and an orthopaedic department. Purposive sampling was initially employed to gain a broad spectrum of experiences using the following variables: age, gender, disease duration and body part affected. Subsequently, participants were selected in order to refine and clarify emergent theories (Theoretical sampling: Glaser, 1978). Sampling continued until the data reached saturation whereby no new themes emerged.

Multi Centre Research Ethics Committee and relevant NHS Trust approvals were granted prior to commencing data collection.

Following written informed consent, qualitative data regarding participants' experience of CRPS were collected using semi-structured interviews. An interview schedule of open-ended questions was used to explore perceptions about the affected limb, how it looked and how it felt both physically and emotionally. The aim was to elicit detailed descriptions with a view to gaining in-depth experience from the participant's perspective and hence a better understanding of this phenomenon (Denzin and Lincoln, 1998). Interviews were undertaken in the participant's own home to encourage a feeling of relaxation and openness. Themes that emerged through the analysis of the early interviews informed the schedule for subsequent interviews. Memos were taken during and after the interviews documenting ideas and emerging theories as the interviews progressed.

2.2. Measures

In order to describe the study sample within the context of the general CRPS population it was appropriate to measure pain, depression and health status. Both the McGill Pain questionnaire and Brief Pain Inventory were considered too long and burdensome, therefore the short form Brief Pain Inventory (BPI: Cleeland and Ryan, 1994; Cleeland et al., 1996) was chosen. The inventory measures pain intensity and interference by participants rating (on a scale of 0–10) three factors: (a) their current pain, (b) pain intensity over the previous week and (c) the degree to which pain has interfered with physical, social and psychological aspects of functioning.

Many people with pain suffer from depression. Commonly used in pain studies, the Beck Depression Inventory (BDI: Beck et al., 1961), although slightly longer than the HADS (Zigmond and Snaith, 1983), was considered suitable. Given the exploratory nature of the study, the depth of the BDI was seen as a strength over a screening tool such as the HADS (Love et al., 2004). The BDI evaluates 21 depressive symptoms on a four point intensity scale comprising emotional, behavioural and somatic symptoms. The SF-36 (Ware and Sherbourne, 1992) was chosen to measure health status. Although this measure is generic and some aspects are not specific to this population it was considered important to reflect the health status of the study sample and gave the potential for comparing with normative data.

Questionnaire and assessment tools were administered following the interview so as not to influence interview responses.

2.3. Data analysis

Interviews were recorded and transcribed verbatim. To protect anonymity, participant names were replaced with identification codes. Interviewees checked the transcripts to ensure reliability of the data. Manual data analysis was initially undertaken followed by further in depth analysis with the aid of a data software program (QSR NVivo (Richards, 2002)). Significant statements from the initial interviews were grouped together into topic areas. In depth questions about these areas were asked at subsequent interviews to form preliminary theory. Cases challenging the emerging theory (negative case analysis: Glaser, 1978) were sought to develop and redefine the theory. A peer analysis review involving the examination of data by qualitative experts was undertaken to ensure that data analysis was credible and robust. Such steps may be considered similar to the requirement in quantitative research to demonstrate the reliability and validity of data.

Data from the questionnaires were statistically analysed using SPSS version 12.0.1 for windows (SPSS, 2003).

3. Results

3.1. Pain, quality of life and depression measures

Results from the BPI, BDI and SF-36 are illustrated in Table 2. As the sample size is small, median scores and interquartile ranges (IQR) have been used rather than means and confidence intervals as is more common

with these data. Whilst caution is necessary given the small sample and wide variation of scores, the sample can be seen to have moderate pain intensity and pain interference (BPI). They were physically limited but with no mental limitations (SF-36) and had mild to moderate depression ratings (BDI).

Twenty-seven participants (12 males, 15 females) were interviewed generating descriptions about body perception experience of considerable scope including those relating to pain. Six themes encapsulated the descriptions of that experience. Themes were noted if they were either repeated by a number of participants or because of the strength of feeling with which they were spoken. These are described and supported with quotes from the interviews. Participant identification codes are given in brackets at the end of each quote as noted below in Table 1. Pain is described in context within the themes where relevant.

The themes are as follows: hostile feelings, spectrum of disassociation; disparity between what is apparent and what is felt; distorted mental image of affected part; awareness of limb position; conscious attention.

The two initial themes are concerned with the attitudes and emotions that were expressed about the affected body part. More abstract perceptions about the appearance and inner felt sense of the affected limb are described in the remaining themes.

Table 1
Participant demographics

Participant ID code	Gender	Age at interview	CRPS type	CRPS trigger	Body part affected	Duration of condition (years)
1	Male	38	I	Soft tissue	Upper limb	5
2	Male	51	I	Soft tissue	Upper limb	3.5
3	Male	50	I	Fracture	Upper limb	3
4	Female	23	I	Post injection	Lower limb	8
5	Female	51	I	Spontaneous	Both	13
6	Female	51	I	Spontaneous	Both	15
7	Male	42	I	Soft tissue	Lower limb	4
8	Male	68	I	Soft tissue	Upper limb	21
9	Female	26	I	Soft tissue	Lower limb	8
10	Male	56	I	Post surgery	Lower limb	13
11	Female	43	II	Post surgery	Upper limb	8
12	Female	18	I	Soft tissue	Both	10
13	Male	19	I	Soft tissue	Lower limb	8
14	Female	47	I	Fracture	Upper limb	8
15	Male	38	II	Soft tissue	Upper limb	1.5
16	Male	33	I	Post surgery	Upper limb	2
17	Female	34	I	Spontaneous	Upper limb	5
18	Female	61	I	Fracture	Upper limb	0.25
19	Female	51	I	Fracture	Lower limb	8
20	Female	53	I	Post surgery	Lower limb	4
21	Female	40	I	Soft tissue	Lower limb	0.4
22	Male	45	I	Fracture	Upper limb	1.25
23	Female	26	I	Soft tissue	Lower limb	8
24	Female	49	I	Spontaneous	Upper limb	4
25	Male	37	I	Spontaneous	Lower limb	7
26	Female	43	I	Soft tissue	Lower limb	1.2
27	Male	62	I	Post shingles	Lower limb	1

Table 2
Pain, health status and depression measures

Measure	Median (IQR)	Mean (SD)	Range
Brief Pain Inventory (BPI) (Short form)			
Pain intensity	6.25 (5.25–7.1)	6 (1.5)	3–8.5
Pain interference	7.14 (4.7–8.35)	6.21 (2.82)	0.14–10
SF-36			
Physical scale	20 (5–65)	33.1 (31)	0–90
Mental scale	52 (36–68)	54.1 (22.3)	16–96
Beck Depression Inventory (BDI)	19.5 (10.25–33.5)	21.9 (14.8)	0–57

3.1.1. Hostile feelings

The majority of participants described hostile feelings about the affected limb whilst unaffected parts were felt to be normal. The intensity of feeling was expressed in varying degrees irrespective of disease duration. These feelings ranged from mild frustration to a considerably more intense experience of hate, disgust and repulsion;

“I feel disgust, I know it sounds a very strong word to use but I’m disgusted that my arm is this way.” (15)

Participants explained that one of the reasons they had negative feelings was because their pain and other symptoms prevented them doing what they were once able. They blamed the affected limb as the cause and as such centred anger and hate towards it.

“I got more cross with it, like if you pick up a bottle, I picked up a bottle the other day from the step and it dropped straight out of my hand only because I hadn’t gripped it as I thought I’d gripped it.” (18)

However, this was not the case for everybody as the following quote illustrates;

“I don’t think, I sort of centralise my annoyance at the pain, blaming my actual leg. I don’t know. I don’t think I have any feelings towards it.” (4)

It was clear from the descriptions that these hostile feelings changed over time. Some expressed that their negative feelings eventually diminished, whilst others reported an increase in intensity during the course of the disease;

“It’s got worse, yeah I’d say it’s got worse, like I’ve got, I think I’ve got more self-conscious about them [um] I hate them more kind of thing.” (13)

3.1.2. Spectrum of disassociation

Participants spoke of how much the affected limb felt a part of their body. A spectrum of disassociation emerged from the data such that many participants described that psychologically the affected limb felt detached to varying degrees from the remainder of their unaffected body. The spectrum ranged from no feelings of detachment to an extreme form in the desperate desire of wanting to get rid of the limb.

“It was just like this foreign body you were carrying around with you cause it didn’t feel like it was part of you.” (17)

Other participants described a stronger sense of disassociation such that the limb felt a separate entity from the body with its own control system that rebelled against the body’s intentions.

“On a good day it’s, it’s fine, it’s behaving itself you know. On a bad day that feeling comes back to me and I’d go through a period throughout the day where I feel like it’s taking control and then I have to sort of muster myself and think, “No, I’m in control” so you know it’s different from day to day and just sometimes if it creeps up on you it sort of overpowers you.” (20)

At the severe end of the disassociation spectrum was the extreme desire of wanting to get rid of the pain by removing the affected limb. This was a common view spontaneously described by many participants who talked in graphic detail of a strong yearning to ‘chop off’ the affected limb;

“I’ve sometimes felt if I could get an axe and chop it off I would do because to me as it stands, at this minute in time sat here it’s a useless, it’s a useless thing.” (2)

Despite the commonality of this experience, a few participants felt differently as noted in the following excerpt;

“It all feels, still feels a part of my body, but, yeah, it does.” (27)

3.1.3. Disparity between what is apparent and what is felt

Participants described how the affected part felt in relation to how it looked whilst viewing their affected limb. Many participants reported that what they saw was often at variance with how the limb felt. Rather than a general distortion, feelings of pain, size, and temperature in discrete parts of the limb were discordant with how their limb appeared. Intense pain was widely described as being a spontaneous felt experience yet not explained by the appearance of the affected part;

“At the moment my hand feels like it, the worse sort of burning that I can imagine and yet I can look at my hand and say there’s nothing burning it.” (15)

The size of the limb was commonly felt to be larger than its’ actual appearance;

“It feels like it’s really, really fat. I mean sometimes I actually look at it to, cause I think god my leg’s swollen and then I’ll look actually look at my leg and I think oh no it’s not but it feels like it is.” (26)

Pain was felt to be a reason for excessive pressure in the affected limb;

“It’s almost as though there’s too much pain to fit into one limb and it does feel like it’s the pain that’s trying to get out.” (11)

Some explained there was no size disparity between the appearance of the limb and how it felt;

“That swelling’s not there any more and the foot’s gone back to the same size as the other one and I haven’t got that sensation of it feeling larger than the other one.” (06)

Participants discussed the spontaneous felt experience of temperature and how that was at odds with the temperature when touching the limb;

“Although my leg can sometimes feel cold to the touch, to me it’s absolutely burning. I can literally feel that my legs are on fire. [um] But if you were to come along and touch them, then they would feel ice cold.” (09)

They found these mismatches between what they were seeing and what they were feeling as confusing and distressing, as they were unable to make sense of them.

“It was just as though the, the sensations were totally mixed up. What I was seeing wasn’t what I was feeling and that, that was, looking back it was confusing.” (10)

Furthermore, participants described receiving a negative reaction from professionals when sharing these experiences.

“All the different types of sensations and feelings and everything you get... when you explain it to a specialist that doesn’t know about this, I mean, they just look at you and think, “Well, you know, you’re pretty stupid.” (1)

3.1.4. Distorted mental image of affected parts

Participants detailed a very distorted image when asked to describe with their eyes closed, a mental picture of their affected limb in comparison to their unaffected contralateral limb. These imagined representations were very different to the participant’s descriptions of actual appearance whilst looking at the affected limb. Typically, discrete parts rather than the whole limb were distorted particularly in size;

“My leg, from my thigh to my knee, it feels almost a normal size, it does hurt, my knee feels swollen and then as I get from the calf to my ankle, the swelling gets worse and then from my ankle to my toes, then it feels enormous.” (19)

Similarly, there was a distortion in shape. Participants talked about a change in anatomical proportions such that parts of the limb were seen as foreshortened;

“They actually feel as if my finger tips are me knuckles, that doesn’t make sense. They, they feel a lot shorter than they should be.” (22)

More intriguingly however, some described that specific parts of their affected limb were missing from their mental image;

“It’s bigger than this leg (gestures to left leg) and then the rest is completely void, there’s nothing there at all, I can actually see the floor. I can see a big toe and I can’t see anything else from my knee down.” (26)

By contrast, a few participants had a normal mental image of their affected limb;

“Both my arms are the same colour there’s no difference there [um] my hands are probably both the same as well.” (17)

3.1.5. Awareness of limb position

Participants explained that their awareness of the affected limb was heightened, particularly as a consequence of the pain and they were conscious of its presence. Paradoxically though, participants typically expressed a difficulty in knowing its position;

“Even if, it gets to the point where if the pain is very intense, even if I am touching something else I’ve got no idea where my arm exists. The first time I realised it I think is when I woke up in bed and it, I, I just couldn’t, I didn’t know where my right arm was, I could feel my left arm fine but, and it wasn’t like the numbness like you’ve rolled over and it’s gone to sleep, I just had no concept of where my arm was and it was like a feeling of panic.” (15)

Furthermore, when their attention was drawn to the position of the limb they became aware of how abnormal the position appeared to be as expressed by participant 11 with CRPS of the left arm.

“It’s strange actually because I didn’t realise until a few weeks ago what I’d been doing and I had actually been putting it behind my back a lot. Keeping it out of the way, partly because I didn’t have to look at it. I wasn’t aware of that until a few weeks ago when somebody mentioned it.” (11)

Participants found the conflicting experience of a heightened awareness of the limb and disorientation to its position to be inexplicable and therefore confusing.

“Often I feel I’m going mad, totally and utterly bonkers because a lot of you, the logical part of you is saying this isn’t and yet part of you believes it is. It’s almost like you’re split in two, part of you is trying to deal with it on a very logical level and the other part is dealing with how you actually feel about it.” (9)

By contrast, a few participants did not describe a mismatch as they were clear that the felt position and the actual limb position were consistent;

“They feel pretty much as to where they are.” (21)

3.1.6. Conscious attention

The last category describes the level of conscious attention participants paid to their affected body parts in terms of thinking about, looking and touching them. Thinking about the limb, for some, generated such strong feelings of disgust that they avoided doing so;

“You’re watching television you see, say, a paedophile and there’s something in you which is almost gut wrenching, you can’t understand how an adult would treat a child like that or a young baby like that or abuse cases. That is about as close as you can get to disgust, and that’s how I feel about my arm. If I can avoid thinking about it, I will.” (9)

Some explained that they consciously distracted their attention away from the limb as a way of dealing with the pain;

“The pain side of it was there so I used to probably distract myself with other things and try and forget about it as much I could which in the end wasn’t a very helpful idea because it, it did you know actually, I did forget about it as much as it just wasn’t part of me eventually.” (23)

Moreover, participants explained that they had to intensely concentrate on their limb in order to undertake what they thought to be a straightforward movement;

“If I went to move a finger, two or three fingers would move. And, it wasn’t like, you know, move that finger and that happened, it was move that finger and, and this went down and you thought, “Well it should be that one.” So I felt like my hand wasn’t attached to my brain, if you know what I mean. It felt like I really had to concentrate to get that arm going, to get that hand going. And I did have to concentrate. And it was, it was a very hard thing to do.” (3)

Many participants spoke of a difficulty in paying visual attention to their limb. They avoided looking at it and in some cases deliberately hid their limb from their field of view;

“I used to try and hide it. [Um] If I was sat over there, which is where I normally sit, I would have my arm on

the other side of that cushion so I couldn’t see it. I still do that now. So, if I can’t see it I can’t be angry with it.” (3)

Difficulty in touching the limb was also expressed and was mainly due to the possibility of causing pain;

“It really was very, very hard to do it. when I touched it, in lots of ways it actually reinforced the idea that it wasn’t mine at all because it didn’t feel like it and I couldn’t make it feel like mine and I didn’t want to make it feel like mine in a lot of ways because it hurt so much.” (23)

By contrast some touched their limb routinely to check the limb in order to gauge a sense of normality;

“I touch it everyday to check that it still feels the same as the other foot.” (21)

Participants spoke about how they were encouraged to think about, look at and touch their limb as part of rehabilitation and how this helped the process of re-engaging with the limb and perceiving it in a more normal way. For example, desensitisation, a rehabilitation technique of touching the limb through stroking, massage and with the use of different textures was experienced as being beneficial;

“I started to touch it, just gentle touching for a second and I, I’ve built it up and built it up so that I then could massage it and talk to it and love it and accept that it was still part of me even though it was still painful, I was still angry with it but I felt like I took control.” (20)

In summary, our participants expressed strong negative feelings about a part of their own body and there was a desperate desire to amputate this part despite the prospect of further pain and loss of function. A mismatch was experienced between the sensation of the limb and how it looked. Anatomical parts of the CRPS limb were erased in mental representations of the affected area. Pain generated a raised consciousness of the limb yet there was a lack of awareness as to its position. Participants voiced a deep tension as emotion fought with logic in trying to make sense of what was happening.

All participants described some form of disturbance in how they perceived their affected body parts regardless of age, gender, trigger or disease duration. That is to say that the presence of disturbance did not seem to be associated with any one of these factors.

However, those with mild disease described fewer components of the themes suggesting that the extent of disturbance was less than those with more severe disease. Those with resolving disease spoke about feelings of disassociation being more apparent during periods when the disease was more active. When talking about the acute stages of onset, participants mentioned being aware of disturbances in body perception within as few as four days of symptom presentation.

4. Discussion

This study has extended our understanding of disturbed body perception and its contribution to pain in CRPS by detailing intriguing insights from the patients' perspective adding to previous studies in this area (Galer et al., 1995; Galer and Jensen, 1999; Forderreuther et al., 2004). Although the methodology and size of the sample preclude generalisation to the total CRPS population (as a quantitative study seeks to achieve), findings reveal how people with this condition perceive their affected limbs. Our data illustrate the bizarre attitudes patients have about parts of their own body, regardless of gender, disease duration and affected body part, and for most, in the absence of major nerve damage. Further to feeling foreign or strange, as Forderreuther et al. and Galer et al. relate (1995, 1999, 2004) the CRPS limb was felt to be another entity and the whole of the affected limb was difficult for sufferers to accurately locate, rather than solely a mislocation of the fingers as previously described (Forderreuther et al., 2004).

To put these strange phenomena in the context of other conditions where body perception disturbances are reported, Galer et al. has previously suggested that they are similar to those seen in neurological neglect. He proposed the term 'motor neglect' (Galer et al., 1995; Galer and Jensen, 1999) to describe difficulty in initiating and coordinating, and the need for mental and visual attention when moving the CRPS limb. A parallel can be drawn with our category 'conscious attention' as participants described the need for intense concentration when undertaking movements which were often difficult and inaccurate. Furthermore, cognitive neglect (Galer et al., 1995; Galer and Jensen, 1999) was reflected in aspects of our category 'spectrum of disassociation'. Participants described the limb as feeling as if it were not part of their body similar to Galer's notion of the limb not being part of their being. However, despite these similarities our findings do not wholly support Galer's hypothesis that body perception disturbances seen in CRPS mimic the traditional definition of neurological neglect-like syndrome (Galer et al., 1995; Galer and Jensen, 1999). Our data suggest that there are three important differences. First, body perception disturbances were experienced in both the acute and chronic stages whereas traditional neurological neglect is observable in the acute stages of a central lesion and often resolves spontaneously within a few weeks (Cutting, 1978). Second, participants had some insight into these disturbances, which is uncommon in neurological neglect, and finally, two participants had both upper and lower limb involvement in contralateral quadrants, whereas neurological neglect is commonly unilateral.

Our participants' descriptions could be suggestive of similarities to those seen in body dysmorphic disorder (Phillips, 1991), where patients have an imagined or

minor defect in their physical appearance leading to a dislike of that area. Severe cases can result in corrective surgery to an otherwise normal body part. Our participants also described a perceived distortion of their affected limb, exacerbated when imagining it with their eyes closed, alongside a strong dislike and a desire for surgical removal of that limb. The important difference between the two conditions is that in body dysmorphic disorder the perceived distorted body part has never appeared abnormal to the external observer, whereas in CRPS changes in colour, size and temperature may have all been observed.

Unlike eating disorders where patients have a distortion in the experience of overall body shape and weight (Skrzypek et al., 2001) our participants described a distortion in the CRPS affected body part only, whilst unaffected areas were perceived as normal. Furthermore, these distortions were experienced as a variety of sensations and were more specific to discrete areas of the CRPS limb than previous studies would suggest (Moseley, 2005).

Given the discrepancies in the perception anomalies reported by our study population and those from other conditions, we suggest that rather than 'neglect-like' symptoms, the term body perception disturbance may better describe the phenomenon in CRPS.

Besides the generation of novel theory, this qualitative approach enables the exploration of new links to existing theory. For example, one might propose that the desire for amputation of an otherwise healthy limb, as reported by the majority of our population, is similar to autotomy (autos meaning self and tomos meaning cut). Although the nosological appropriateness of this term is under debate (Devor, 2007; Wilkie et al., 2007), it was defined by Patrick Wall et al. (1979) to describe the maiming behaviour of rodents when they gnawed their otherwise healthy, anaesthetised limb following nerve ligation. Descriptions of a desire for intentional self mutilation to an existing painful part appear to contravene the need to guard and protect the limb against generating further pain and perhaps illustrate a modified version of animal autotomy. As only two participants had demonstrable nerve damage this theory has limitations. However, there does appear to be a hereditary single gene autosomal recessive trait within animal autotomy (Devor and Raber, 1990). If this is the case, it may indicate that a centrally driven reflex underlies the onset of body perception disturbance in CRPS. Further research is required to define whether this is genetically derived.

A reduction in body perception disturbance appeared to parallel symptom reduction, in particular pain, as those with resolving disease expressed these disturbances to a lesser extent. Brain imaging studies (Maihofner et al., 2004; Pleger et al., 2005) demonstrating that corrective cortical reorganisation correlates with a

reduction in CRPS pain provide reasonable evidence to suggest that there is a close relationship between altered central limb representation and a disturbance in affected limb perception. The relationship between pain and body perception disturbance is however, more elusive. Given that disturbances in body perception become apparent within a few days of symptom onset and appear to fluctuate in line with the intensity of symptoms this would suggest that possible central reorganisation is dynamic and that a complex interaction between body perception disturbance and pain exists. Whether a disturbance in body perception precipitates or perpetuates pain remains unclear.

The erasing of discrete anatomical parts of the CRPS limb from the mental body representation suggests a specific alteration in the central body schema map. Participants expressed surprise when describing these mental distortions indicating that these changes occurred outside of their conscious state of awareness. This finding further supports the view that an altered central representation contributes to a disturbance in body perception. An intact cortical body schema is an essential component of movement control (Graziano and Botvinick, 2002) therefore absence of limb segments within the central representation of the CRPS limb may have implications for function.

Participants expressed more difficulty in articulating the subtler aspects of body perception disturbances than when describing pain and spoke of an inner turmoil in attempting to make sense of them. Raising such concerns with health professionals may cause distress for fear of being regarded as absurd and possibly dismissed. However, many participants expressed relief at being able to share these previously untold experiences and we would suggest that clinicians encourage and reassure patients when discussing these issues, as they are clearly not alone in experiencing disturbing thoughts and feelings. Furthermore, gaining an understanding of the individuals' body perception disturbance may inform treatment choice.

As our findings suggest a close relationship between body perception disturbance and altered central representations, it would seem reasonable to target treatments at correcting this remapping with the aim of reducing pain and normalising limb perception. Novel treatments for CRPS such as mirror visual feedback (McCabe et al., 2003) and motor imagery (Moseley, 2004, 2006) are designed to target cortical areas, and appear to provide pain relief. Yet they may also be influencing other, as yet unmeasured, symptoms associated with body perception.

Participants expressed that other less well researched interventions such as desensitisation helped them to perceive their limb in a more normal way. As these strategies involve looking, touching and thinking about the affected limb they too may have an influence on central areas.

Recommendations for further research include developing a clinical tool to measure the extent and nature of body perception disturbance in CRPS. Testing the potential for new interventions in reducing body perception disturbance, as well as pain, is warranted. Such testing may also establish the efficacy of existing rehabilitation techniques. A more definitive relationship between body perception disturbance and pain could be established by determining whether CRPS patients with no ongoing pain have a disturbance in body perception.

In conclusion, this qualitative study has provided a more detailed insight into how patients with CRPS perceive the body perception anomalies associated with their affected limbs. We have demonstrated that the definitions previously applied to these perceptions do not accurately or adequately reflect the patients' experience and suggest that 'body perception disturbance' is an appropriate term to describe this phenomenon. Findings suggest a complex interaction between pain, body perception disturbance and central remapping. Clinically, our findings support the use of treatments that target cortical areas, which may reduce body perception disturbance and pain. Further study is required to measure the extent of body perception disturbance and clarify the relationship with pain in CRPS.

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Computer Program

Abstract

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