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

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### 12 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 this 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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**Keywords:** Complex regional pain syndrome; Reflex sympathetic dystrophy; Patient experience; Body perception disturbance; Mental body representation; Pain; Neglect; Central mechanisms

### 30 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

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

50 Recent brain imaging studies (Maihofner et al., 2003;  
51 Pleger et al., 2005) have shown that those with CRPS  
52 have a disrupted cortical map of the affected body part.  
53 An association between symptom resolution over time  
54 and the corrective reorganisation of cortical limb repre-  
55 sentation suggests that there is a relationship between  
56 the amount of pain and the degree of cortical disorgani-  
57 sation (Flor, 2003; Maihofner et al., 2004; Pleger et al.,  
58 2005). The existence of these disturbed representations  
59 could serve to influence the pain experience and alter  
60 how an individual may perceive their CRPS limb. In  
61 order to better understand these representations and  
62 their impact, it is important to know how people with  
63 CRPS actually perceive their own affected body parts.  
64 A review of the literature suggests that to date, this per-  
65 spective remains poorly researched and poorly under-  
66 stood. We suggest a greater knowledge of body  
67 perception in CRPS may provide valuable insights into  
68 central mechanisms and the relationship with pain. In  
69 addition, given that CRPS is known to be resistive to  
70 conventional treatments, an improved understanding  
71 of body perception in relation to pain may identify areas  
72 in which interventions could be specifically targeted.

73 To achieve these objectives, we set out to explore the  
74 patients' experience of body perception in CRPS, to pos-  
75 tulate a theoretical framework for understanding the  
76 characteristics of body perception disturbance and  
77 how it may contribute to the pain experience. Finally,  
78 we discuss how these findings may inform clinical  
79 practice.

## 80 2. Methods

81 Given the inductive nature of this research (i.e. hypothesis  
82 deriving not hypothesis testing), a qualitative methodology,  
83 drawing on principles of grounded theory, was utilised. This  
84 approach, previously used in the pain field, is of particular  
85 value in exploring areas such as this, where little research  
86 has been undertaken and knowledge is limited (Flick, 2002).  
87 Just as case studies are used to inform and develop hypotheses,  
88 a strength of grounded theory is that it aims to generate theory  
89 from the data from which new hypotheses can be made. Emer-  
90 gent theory is not merely descriptive but seeks to identify key  
91 relationships within and between both existing and newly pro-  
92 posed theory in order to enhance our understanding (Glaser,  
93 1969; Strauss and Corbin, 1998; Seale, 1999; Stanley and  
94 Cheek, 2003).

### 95 2.1. Participants and data collection

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

Additional inclusion criteria were the capability to verbally com- 99  
municate and the absence of co-morbidity such as diabetic neu- 100  
ropathy which may significantly influence their experience. 101  
Recruitment sources included a national CRPS referral hospital, 102  
RSD-UK, the United Kingdom CRPS patient charity, pain clinics 103  
and an orthopaedic department. Purposive sampling was initially 104  
employed to gain a broad spectrum of experiences using 105  
the following variables: age, gender, disease duration and body 106  
part affected. Subsequently, participants were selected in order 107  
to refine and clarify emergent theories (Theoretical sampling: 108  
Glaser, 1978). Sampling continued until the data reached saturation 109  
whereby no new themes emerged. 110

Multi Centre Research Ethics Committee and relevant 111  
NHS Trust approvals were granted prior to commencing data 112  
collection. 113

Following written informed consent, qualitative data 114  
regarding participants' experience of CRPS were collected 115  
using semi-structured interviews. An interview schedule of 116  
open-ended questions was used to explore perceptions about 117  
the affected limb, how it looked and how it felt both physically 118  
and emotionally. The aim was to elicit detailed descriptions 119  
with a view to gaining in-depth experience from the partici- 120  
pant's perspective and hence a better understanding of this 121  
phenomenon (Denzin and Lincoln, 1998). Interviews were 122  
undertaken in the participant's own home to encourage a feel- 123  
ing of relaxation and openness. Themes that emerged through 124  
the analysis of the early interviews informed the schedule for 125  
subsequent interviews. Memos were taken during and after 126  
the interviews documenting ideas and emerging theories as 127  
the interviews progressed. 128

### 2.2. Measures 129

In order to describe the study sample within the context of 130  
the general CRPS population it was appropriate to measure 131  
pain, depression and health status. Both the McGill Pain ques- 132  
tionnaire and Brief Pain Inventory were considered too long 133  
and burdensome, therefore the short form Brief Pain Inventory 134  
(BPI: Cleeland and Ryan, 1994; Cleeland et al., 1996) was cho- 135  
sen. The inventory measures pain intensity and interference by 136  
participants rating (on a scale of 0–10) three factors: (a) their 137  
current pain, (b) pain intensity over the previous week and 138  
(c) the degree to which pain has interfered with physical, social 139  
and psychological aspects of functioning. 140

Many people with pain suffer from depression. Commonly 141  
used in pain studies, the Beck Depression Inventory (BDI: 142  
Beck et al., 1961), although slightly longer than the HADS 143  
(Zigmond and Snaith, 1983), was considered suitable. Given 144  
the exploratory nature of the study, the depth of the BDI 145  
was seen as a strength over a screening tool such as the HADS 146  
(Love et al., 2004). The BDI evaluates 21 depressive symptoms 147  
on a four point intensity scale comprising emotional, behav- 148  
ioural and somatic symptoms. The SF-36 (Ware and Sher- 149  
bourne, 1992) was chosen to measure health status. 150  
Although this measure is generic and some aspects are not spe- 151  
cific to this population it was considered important to reflect 152  
the health status of the study sample and gave the potential 153  
for comparing with normative data. 154

Questionnaire and assessment tools were administered fol- 155  
lowing the interview so as not to influence interview responses. 156

## 157 2.3. Data analysis

158 Interviews were recorded and transcribed verbatim. To protect  
159 anonymity, participant names were replaced with identification  
160 codes. Interviewees checked the transcripts to ensure  
161 reliability of the data. Manual data analysis was initially  
162 undertaken followed by further in depth analysis with the  
163 aid of a data software program (QSR NVivo (Richards,  
164 2002)). Significant statements from the initial interviews were  
165 grouped together into topic areas. In depth questions about  
166 these areas were asked at subsequent interviews to form preliminary  
167 theory. Cases challenging the emerging theory (negative  
168 case analysis: Glaser, 1978) were sought to develop and  
169 redefine the theory. A peer analysis review involving the exam-  
170 ination of data by qualitative experts was undertaken to ensure  
171 that data analysis was credible and robust. Such steps may be  
172 considered similar to the requirement in quantitative research  
173 to demonstrate the reliability and validity of data.

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

## 176 3. Results

## 177 3.1. Pain, quality of life and depression measures

178 Results from the BPI, BDI and SF-36 are illustrated  
179 in Table 2. As the sample size is small, median scores  
180 and interquartile ranges (IQR) have been used rather  
181 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 atti-  
tudes 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	0.25
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

### 209 3.1.1. Hostile feelings

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

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

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

223  
224 *“I got more cross with it, like if you pick up a bottle, I  
225 picked up a bottle the other day from the step and it  
226 dropped straight out of my hand only because I hadn’t  
227 gripped it as I thought I’d gripped it.”* (18)

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

230  
231 *“I don’t think, I sort of centralise my annoyance at the  
232 pain, blaming my actual leg. I don’t know. I don’t think  
233 I have any feelings towards it.”* (4)

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

239  
240 *“It’s got worse, yeah I’d say it’s got worse, like I’ve got, I  
241 think I’ve got more self-conscious about them [um] I hate  
242 them more kind of thing.”* (13)

### 243 3.1.2. Spectrum of disassociation

244 Participants spoke of how much the affected limb felt  
245 a part of their body. A spectrum of disassociation  
246 emerged from the data such that many participants  
247 described that psychologically the affected limb felt  
248 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.

249  
250  
251  
252  
253  
254  
255  
*“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)

256 Other participants described a stronger sense of disas-  
257 sociation such that the limb felt a separate entity from  
258 the body with its own control system that rebelled  
259 against the body’s intentions.

260  
261  
262  
263  
264  
265  
266  
267  
*“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)

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

274  
275  
276  
277  
*“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)

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

281  
282  
283  
*“It all feels, still feels a part of my body, but, yeah, it  
does.”* (27)

### 284 3.1.3. Disparity between what is apparent and what is felt

285 Participants described how the affected part felt in  
286 relation to how it looked whilst viewing their affected  
287 limb. Many participants reported that what they saw  
288 was often at variance with how the limb felt. Rather

289 than a general distortion, feelings of pain, size, and tem-  
 290 perature in discrete parts of the limb were discordant  
 291 with how their limb appeared. Intense pain was widely  
 292 described as being a spontaneous felt experience yet  
 293 not explained by the appearance of the affected part;

294  
 295 *“At the moment my hand feels like it, the worse sort of*  
 296 *burning that I can imagine and yet I can look at my hand*  
 297 *and say there’s nothing burning it.” (15)*

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

300  
 301 *“It feels like it’s really, really fat. I mean sometimes I*  
 302 *actually look at it to, cause I think god my leg’s swollen*  
 303 *and then I’ll look actually look at my leg and I think oh*  
 304 *not it’s not but it feels like it is.” (26)*

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

307  
 308 *“It’s almost as though there’s too much pain to fit into one*  
 309 *limb and it does feel like it’s the pain that’s trying to get*  
 310 *out.” (11)*

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

313  
 314 *“That swelling’s not there any more and the foot’s gone*  
 315 *back to the same size as the other one and I haven’t got*  
 316 *that sensation of it feeling larger than the other one.” (06)*

317 Participants discussed the spontaneous felt experience  
 318 of temperature and how that was at odds with the tem-  
 319 perature when touching the limb;

320  
 321 *“Although my leg can sometimes feel cold to the touch, to*  
 322 *me it’s absolutely burning. I can literally feel that my legs*  
 323 *are on fire. [um] But if you were to come along and touch*  
 324 *them, then they would feel ice cold.” (09)*

325 They found these mismatches between what they were  
 326 seeing and what they were feeling as confusing and dis-  
 327 tressing, as they were unable to make sense of them.

328  
 329 *“It was just as though the, the sensations were totally*  
 330 *mixed up. What I was seeing wasn’t what I was feeling*  
 331 *and that, that was, looking back it was confusing.” (10)*

332 Furthermore, participants described receiving a nega-  
 333 tive reaction from professionals when sharing these  
 334 experiences.

335  
 336 *“All the different types of sensations and feelings and*  
 337 *everything you get... when you explain it to a specialist*

*that doesn’t know about this, I mean, they just look at* 338  
*you and think, “Well, you know, you’re pretty stupid.” (1)* 339

#### 3.1.4. Distorted mental image of affected parts 340

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. Typi-  
 cally, discrete parts rather than the whole limb were dis-  
 torted particularly in size;

349  
 350 *“My leg, from my thigh to my knee, it feels almost a nor-*  
 351 *mal size, it does hurt, my knee feels swollen and then as I*  
 352 *get from the calf to my ankle, the swelling gets worse and*  
 353 *then from my ankle to my toes, then it feels enormous.”*  
 354 *(19)*

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

358  
 359 *“They actually feel as if my finger tips are me knuckles,*  
 360 *that doesn’t make sense. They, they feel a lot shorter than*  
 361 *they should be.” (22)*

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

365  
 366 *“It’s bigger than this leg (gestures to left leg) and then the*  
 367 *rest is completely void, there’s nothing there at all, I can*  
 368 *actually see the floor. I can see a big toe and I can’t see*  
 369 *anything else from my knee down.” (26)*

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

372  
 373 *“Both my arms are the same colour there’s no difference*  
 374 *there [um] my hands are probably both the same as well.”*  
 375 *(17)*

#### 3.1.5. Awareness of limb position 377

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

383  
 384 *“Even if, it gets to the point where if the pain is very intense,*  
 385 *even if I am touching something else I’ve got no idea where*  
 386 *my arm exists. The first time I realised it I think is when I*



387 woke up in bed and it, I, I just couldn't, I didn't know where  
388 my right arm was, I could feel my left arm fine but, and it  
389 wasn't like the numbness like you've rolled over and it's gone  
390 to sleep, I just had no concept of where my arm was and it  
391 was like a feeling of panic." (15)

392 Furthermore, when their attention was drawn to the  
393 position of the limb they became aware of how abnormal  
394 the position appeared to be as expressed by partic-  
395 ipant 11 with CRPS of the left arm.

396  
397 "It's strange actually because I didn't realise until a few  
398 weeks ago what I'd been doing and I had actually been  
399 putting it behind my back a lot. Keeping it out of the  
400 way, partly because I didn't have to look at it. I wasn't  
401 aware of that until a few weeks ago when somebody men-  
402 tioned it." (11)

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

406  
407 "Often I feel I'm going mad, totally and utterly bonkers  
408 because a lot of you, the logical part of you is saying this  
409 isn't and yet part of you believes it is. It's almost like  
410 you're split in two, part of you is trying to deal with it  
411 on a very logical level and the other part is dealing with  
412 how you actually feel about it." (9)

413 By contrast, a few participants did not describe a mis-  
414 match as they were clear that the felt position and the  
415 actual limb position were consistent;

416  
417 "They feel pretty much as to where they are." (21)

### 418 3.1.6. Conscious attention

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

424  
425 "You're watching television you see, say, a paedophile and  
426 there's something in you which is almost gut wrenching, you  
427 can't understand how an adult would treat a child like that  
428 or a young baby like that or abuse cases. That is about as  
429 close as you can get to disgust, and that's how I feel about  
430 my arm. If I can avoid thinking about it, I will." (9)

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

434  
435 "The pain side of it was there so I used to probably dis-  
436 tract myself with other things and try and forget about

it as much I could which in the end wasn't a very helpful 437  
idea because it, it did you do actually, I did forget about 438  
it as much as it just wasn't part of me eventually." (23) 439

Moreover, participants explained that they had to 440  
intensely concentrate on their limb in order to undertake 441  
what they thought to be a straightforward movement; 442

443  
444 "If I went to move a finger, two or three fingers would  
445 move. And, it wasn't like, you know, move that finger  
446 and that happened, it was move that finger and, and this  
447 went down and you thought, "Well it should be that  
448 one." So I felt like my hand wasn't attached to my brain,  
449 if you know what I mean. It felt like I really had to con-  
450 centrate to get that arm going, to get that hand going,  
451 to get that arm going. And I did have to concentrate.  
452 And it was, it was a very hard thing to do." (3)

Many participants spoke of a difficulty in paying 453  
visual attention to their limb. They avoided looking at 454  
it and in some cases deliberately hid their limb from 455  
their field of view; 456

457  
458 "I used to try and hide it. [Um] If I was sat over there,  
459 which is where I normally sit, I would have my arm on  
460 the other side of that cushion so I couldn't see it. I still  
461 do that now. So, if I can't see it I can't be angry with  
462 it." (3)

Difficulty in touching the limb was also expressed and 463  
was mainly due to the possibility of causing pain; 464

465  
466 "It really was very, very hard to do it. when I touched it, in  
467 lots of ways it actually reinforced the idea that it wasn't  
468 mine at all because it didn't feel like it and I couldn't make  
469 it feel like mine and I didn't want to make it feel like mine  
470 in a lot of ways because it hurt so much." (23)

By contrast some touched their limb routinely to 471  
check the limb in order to gauge a sense of normality; 472

473  
474 "I touch it everyday to check that it still feels the same as  
475 the other foot." (21)

Participants spoke about how they were encouraged 476  
to think about, look at and touch their limb as part of 477  
rehabilitation and how this helped the process of re- 478  
engaging with the limb and perceiving it in a more nor- 479  
mal way. For example, desensitisation, a rehabilitation 480  
technique of touching the limb through stroking, mas- 481  
sage and with the use of different textures was experi- 482  
enced as being beneficial; 483

484  
485 "I started to touch it, just gentle touching for a second and  
486 I, I've built it up and built it up so that I then could mas-  
487 sage it and talk to it and love it and accept that it was still

488 *part of me even though it was still painful, I was still angry*  
489 *with it but I felt like I took control.”* (20)

490 In summary, our participants expressed strong nega-  
491 tive feelings about a part of their own body and there  
492 was a desperate desire to amputate this part despite  
493 the prospect of further pain and loss of function. A mis-  
494 match was experienced between the sensation of the  
495 limb and how it looked. Anatomical parts of the CRPS  
496 limb were erased in mental representations of the  
497 affected area. Pain generated a raised consciousness of  
498 the limb yet there was a lack of awareness as to its posi-  
499 tion. Participants voiced a deep tension as emotion  
500 fought with logic in trying to make sense of what was  
501 happening.

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

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

#### 516 4. Discussion

517 This study has extended our understanding of dis-  
518 turbed body perception and its contribution to pain in  
519 CRPS by detailing intriguing insights from the patients’  
520 perspective adding to previous studies in this area (Galer  
521 et al., 1995; Galer and Jensen, 1999; Forderreuther  
522 et al., 2004). Although the methodology and size of  
523 the sample preclude generalisation to the total CRPS  
524 population (as a quantitative study seeks to achieve),  
525 findings reveal how people with this condition perceive  
526 their affected limbs. Our data illustrate the bizarre atti-  
527 tudes patients have about parts of their own body,  
528 regardless of gender, disease duration and affected body  
529 part, and for most, in the absence of major nerve dam-  
530 age. Further to feeling foreign or strange, as Forderreu-  
531 ther et al. and Galer relate (1995, 1999, 2004) the CRPS  
532 limb was felt to be another entity and the whole of the  
533 affected limb was difficult for sufferers to accurately  
534 locate, rather than solely a mislocation of the fingers  
535 as previously described (Forderreuther et al., 2004).

536 To put these strange phenomena in the context of  
537 other conditions where body perception disturbances  
538 are reported, Galer has previously suggested that they  
539 are similar to those seen in neurological neglect. He pro-  
540 posed the term ‘motor neglect’ (Galer et al., 1995; Galer

and Jensen, 1999) to describe difficulty in initiating and  
542 coordinating, and the need for mental and visual atten-  
543 tion when moving the CRPS limb. A parallel can be  
544 drawn with our category ‘conscious attention’ as partic-  
545 ipants described the need for intense concentration when  
546 undertaking movements which were often difficult and  
547 inaccurate. Furthermore, cognitive neglect (Galer  
548 et al., 1995; Galer and Jensen, 1999) was reflected in  
549 aspects of our category ‘spectrum of disassociation’.  
550 Participants described the limb as feeling as if it were  
551 not part of their body similar to Galer’s notion of the  
552 limb not being part of their being. However, despite  
553 these similarities our findings do not wholly support  
554 Galer’s hypothesis that body perception disturbances  
555 seen in CRPS mimic the traditional definition of neuro-  
556 logical neglect-like syndrome (Galer et al., 1995; Galer  
557 and Jensen, 1999). Our data suggest that there are three  
558 important differences. First, body perception distur-  
559 bances were experienced in both the acute and chronic  
560 stages whereas traditional neurological neglect is obser-  
561 vable in the acute stages of a central lesion and often  
562 resolves spontaneously within a few weeks (Cutting,  
563 1978). Second, participants had some insight into these  
564 disturbances, which is uncommon in neurological  
565 neglect, and finally, two participants had both upper  
566 and lower limb involvement in contralateral quadrants,  
567 whereas neurological neglect is commonly unilateral.

568 Our participants’ descriptions could be suggestive of  
569 similarities to those seen in body dysmorphic disorder  
570 (Phillips, 1991), where patients have an imagined or  
571 minor defect in their physical appearance leading to a  
572 dislike of that area. Severe cases can result in corrective  
573 surgery to an otherwise normal body part. Our partici-  
574 pants also described a perceived distortion of their  
575 affected limb, exacerbated when imagining it with their  
576 eyes closed, alongside a strong dislike and a desire for  
577 surgical removal of that limb. The important difference  
578 between the two conditions is that in body dysmorphic  
579 disorder the perceived distorted body part has never  
580 appeared abnormal to the external observer, whereas  
581 in CRPS changes in colour, size and temperature may  
582 have all been observed.

583 Unlike eating disorders where patients have a distor-  
584 tion in the experience of overall body shape and weight  
585 (Skrzypek et al., 2001) our participants described a dis-  
586 tortion in the CRPS affected body part only, whilst  
587 unaffected areas were perceived as normal. Further-  
588 more, these distortions were experienced as a variety  
589 of sensations and were more specific to discrete areas  
590 of the CRPS limb than previous studies would suggest  
591 (Moseley, 2005).

592 Given the discrepancies in the perception anomalies  
593 reported by our study population and those from other  
594 conditions, we suggest that rather than ‘neglect-like’  
595 symptoms, the term body perception disturbance may  
596 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 totos meaning cut). Although the nosological appropriateness of this term is under debate (Devor, in press; Wilkie et al., in press), 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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*Abstract*

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