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Perceptions of the painful body: The relationship between body perception disturbance, pain and tactile discrimination in complex regional pain syndrome

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Abstract

Background: Disturbances in body perception are increasingly acknowledged as a feature of complex regional pain syndrome (CRPS). Conventional treatments have limited success particularly among those with long-standing disease. Understanding the relationship between body perception disturbance, pain and tactile acuity might provide insight into alternative avenues for treatment. The aim of this study was to test the hypotheses that (1) body perception disturbance is positively related to pain and (2) decreased tactile acuity is related to increased body perception disturbance.
Methods: A controlled observational design was used to measure these features among those with CRPS of one arm. The extent of body perception disturbance was assessed using the Bath CRPS body perception disturbance scale and pain was measured using the neuropathic pain symptom inventory. Two-point discrimination threshold testing was performed as a measure of tactile acuity.
Results: Findings confirmed both hypotheses. Body perception disturbance was found to positively correlate with pain such that those in greater pain had more extensive body perception disturbance ($r = 0.57$, $p < 0.01$). Furthermore, a positive correlation was revealed between body perception disturbance and two-point discrimination thresholds ($r = 0.5$, $p < 0.025$) so those with greater body perception disturbance had worse tactile acuity. Interestingly, those with longer disease duration had significantly greater body perception disturbance ($r = 0.66$, $p < 0.001$).
Conclusion: Aberrant central processing is suggested as the neural correlate of body perception disturbance and tactile impairment. The exact relationship between body perception disturbance, pain and tactile acuity and how they may be modulated for pain relief requires further exploration.

1. Background

Effective treatment of severe and long-standing complex regional pain syndrome (CRPS) is notoriously difficult (Perez et al., 2001; Rowbotham, 2006; McCabe and Blake, 2008). A novel perspective worthy of investigation is the seemingly detrimental influence of pain on the subjective perception of the affected part (Forderreuther et al., 2004; Moseley, 2005; Lewis et al., 2007). A greater understanding of how pain and body perception are related may reveal treatment opportunities.

The majority of those with CRPS (between 54.4% and 84%, Forderreuther et al., 2004; Galer and Jensen, 1999, respectively) report disturbances in body perception. Altered perceptions of their painful body regions

What's already known about this topic?
- Disturbances in body perception are a common feature of complex regional pain syndrome (CRPS).
- Patients perceive the size of their affected hand as larger than in reality and this is associated with impaired touch.

What does this study add?
- CRPS patients in greater pain have more extensive body perception disturbances.
- Those with greater body perception disturbance have worse tactile acuity.
- Patients with longer disease duration have greater body perception disturbance.

are expressed in various ways. Individuals indicate a loss of self-ownership describing the limb as foreign and have a desire for amputation (Geertzen and Eisma, 1994; Galer et al., 1995; Galer and Jensen, 1999; Forderreuther et al., 2004; Lewis et al., 2007). The limb is perceived as distorted in shape and size, heavier, pressurized or different in temperature from objective assessment (Moseley, 2005; Lewis et al., 2007; Lewis and McCabe, 2010; Peltz et al., 2011). Moreover, with their eyes closed, individuals describe distortions in shape and size when mentally visualizing the affected limb and some are unable to picture anatomical parts (Lewis et al., 2007). Clinical observations suggest that patients with body perception disturbances have difficulty relating normally to their painful limb and that this affects rehabilitation outcomes.

Supporting evidence of the proposed relationship between pain and body perception can be found in neural representations within the brain. Body perception, the process of how we perceive our bodies, involves complex interactions between proprioceptive, vestibular, somatosensory and visual inputs interrelating with motor systems. Resultant information forms online neural body representations (termed body schema) stored in cortices such as the primary somatosensory cortex (SI), and other parietal regions (Haggard and Wolpert, 2005; Medina and Coslett, 2010).

The processing of pain is associated with neural activation in SI (Bushnell et al., 1999). Pathologic cortical reorganization of the CRPS affected region in SI is known to occur (Juottonen et al., 2002; Maihofner et al., 2003, 2004; Pleger et al., 2006). Given that SI contributes in part to the body schema suggests that these disturbed central representations may serve to influence how an individual perceives their CRPS limb. The extent of cortical alterations relates to the degree of pain (Maihofner et al., 2004). Importantly, aberrant mapping reverses as pain diminishes (Maihofner et al., 2004; Pleger et al., 2005). In light of this evidence, we tested the hypothesis that pain and body perception disturbance are positively correlated in CRPS.

Furthermore, the extent of SI cortical reorganization relates to tactile impairment in CRPS (Pleger et al., 2006). Moreover, impaired tactile acuity and overestimation of affected hand size are correlated (Peltz et al., 2011). Based on these studies, we tested the hypothesis that tactile acuity decreases with an increase in body perception disturbance in CRPS.

2. Methods

2.1 Participants

Twenty-two participants aged 18 and over who met the International Association of the Study of Pain (IASP) classification criteria for CRPS type I (Stanton-Hicks et al., 1995) of one upper limb were recruited from a Canadian population. Given the exploratory nature of this study, the number of participants was based on a convenience sample. Additional inclusion criteria were that they could verbally communicate and had no co-morbidity that might influence tactile perception. Participants were recruited from patients who had attended hospitals in the Montreal area in addition to those recruited via relevant patient support groups. Fifteen women and seven men with a mean age of 50.6 years [standard deviation (SD) 10.6 years] took part. CRPS participant data including symptom duration [mean = 37.2 months, SD = 53.7 months, minimum (min)–maximum (max) 3–240 months] and disease presentation are presented in Table 1.

Twenty-two healthy volunteers with no history of chronic pain and who matched the patients' age and gender [mean age = 43 years, SD = 14.2 years, unpaired *t*-test between patients and controls: $p > 0.05$, $t = 1.97$, degrees of freedom (df) = 42] were also recruited via adverts. All participants gave written consent and data were collected in accordance with a protocol approved by Research Ethics Committees of McGill University and the respective hospitals.

2.2 Procedures

2.2.1 Diagnostic assessment

A clinical evaluation was undertaken to ascertain whether the individual met the current IASP diagnos-

Table 1 CRPS participant characteristics.

ID	Age (years)/ gender	Reported inciting incident	Affected upper limb	Dur. (months)	Reported medications	Allodynia and/or hyperalgesia present	Vasomotor sign	Sudomotor sign	Motor sign	Trophic sign	Meets Bruehl et al.'s (1999) research criteria
01	54/F	Fr	L ◆	9	None reported	✔	✔	✔	✔	✔	✔
02	31/F	STI	L	71	Medical marijuana	✔	✔	✔	✔		✔
03	45/M	STI	R ◆	17	Tramadol, pregablin, ibuprofen, cyclonebenzaprine, ranpantoprazole	✔		✔	✔		✔
04	38/M	STI	L◆	30	Methodone, naproxen, tramadol	✔		✔	✔		✔
05	43/F	STI	L	12	alendronate, bromazepam	✔	✔	✔	✔	✔	✔
06	62/F	STI	L	240	Fentanyl patches, medical marijuana, duloxetine, tolterodine, amitripyline, valsartan, rosiglitazone metformin	✔	✔		✔	✔	✔
07	39/F	STI	L	68	Hydromorphine	✔	✔	✔	✔		✔
08	33/F	Fr	R ◆	11	Amitripyline	✔	✔	✔	✔	✔	✔
09	55/M	PS	L	40	Baclofen, ibuprofen, hydromorphine rosuvastatin, atenol, omeprazole, aspirin venlafaxine, quetiapine, oxcarbazepine	✔		✔	✔	✔	✔
10	44/M	STI	L ◆	7	Pregablin, acetaminophen	✔	✔	✔	✔		✔
11	51/F	STI	R ◆	126	Oxycodone	✔	✔	✔	✔	✔	✔
12	57/M	Fr	R ◆	14	Celecoxib	✔		✔	✔		
13	44/F	STI	R◆	50	Naproxen, baclofen, pregablin, acetaminophen	✔	✔	✔	✔		✔
14	50/F	Fr	R ◆	11	Duloxetine, pregablin, naproxen, docusate sodium	✔			✔	✔	
15	65/F	PS	R ◆	11	Amitripyline				✔	✔	
16	45/M	Fr	R	6	Pregablin, amitripyline	✔	✔		✔		✔
17	56/F	Fr	L	18	Ibuprofen	✔			✔		
18	73/M	Fr	L Δ	15	Tramadol	✔	✔		✔	✔	
19	59/F	STI	R	23	Simvastatin, aspirin, calcium	✔			✔		
20	50/F	Fr	R ◆	18	Ramipril			✔	✔	✔	
21	60/F	Fr	R ◆	20	None reported	✔		✔	✔	✔	✔
22	60/F	Fr	L	3	Calcitonin, pregablin, amitripyline, vit c		✔	✔	✔	✔	✔

F, female; M, male; STI, soft tissue injury; Fr, fracture; PS, post-surgery; L, left; R, right; ◆, dominant hand is affected; Dur. (months), duration in months since symptom onset; Δ, amputated distal phalanx of index finger on affected side; CRPS, complex regional pain syndrome.

tic criteria for CRPS type 1. Presenting signs and symptoms of the condition were assessed. The presence of mechanical allodynia and hyperalgesia was determined via the use of von Frey hairs applied to the affected limb. Temperature asymmetry between both hands was assessed using a spot infrared thermometer directed at the dorsal surface proximal to the third metacarpophalangeal joint of each hand. Examining the appearance of both upper limbs assessed asymmetries in skin colour, sweating, oedema, hair and nail growth. A physical examination determined any abnormalities in range of movement of the affected limb. Disease history and current medication were documented.

2.2.2 Outcome measures

2.2.2.1 Body perception disturbance rating

The Bath CRPS body perception disturbance scale was used to measure changes in body perception of the affected limb (Lewis and McCabe, 2010). This scale is underpinned by extensive qualitative patient-orientated research and reflects in detail the breadth of body perception disturbance expressed by CRPS patients (Lewis et al., 2007). Full assessment of psychometric properties of the Bath CRPS body perception disturbance scale has not yet been performed. We have calculated internal consistency using Cronbach's alpha coefficient and interrater reliability using Cohen's kappa for our study sample (Nunnally and Bernstein, 1994; Streiner and Norman, 2008; Abell et al., 2009). Values of 0.66 for Cronbach's alpha and 0.87 for Cohen's kappa suggest adequate internal consistency (Streiner and Norman, 2008; Abell et al., 2009) and adequate interrater reliability (Nunnally and Bernstein, 1994) of the scale.

In order to obtain the nature and extent of these perceptual disturbances, the questionnaire comprises seven items covering different aspects related to the affected limb. These are: (1) a sense of ownership; (2) limb position awareness; (3) attention paid to the limb; (4) emotional feelings towards the limb; (5) perceptual disparities in size, temperature, pressure and weight (compared with the unaffected limb); (6) a desire to amputate the limb; and (7) a mental representation of the affected limb.

Five of these seven items are rated on a 0–10 scale to establish the extent of abnormality within that item. Item 5 (perceptual disparity between affected and unaffected limbs) requires a dichotomous response (yes = 1, no = 0) for each quality (size, temperature, etc.). The final item involves an illustration of the mental representation of both the affected and unaffected limbs. With eyes closed, the participant is asked to generate a mental picture of both upper limbs commencing with his/her unaffected side. As the participant verbalizes his/her mental image, the investigator draws a picture of both limbs based on these descriptions. The resultant drawing is assessed by the participant for accuracy and adjusted if necessary. The drawing is independently graded on a three-point scale: no distortion = 0, distortion = 1, severe distortion = 2. If either a distortion in size or shape is depicted within the drawing or the accompanying textual descriptions, i.e. that it is not anatomically consistent with the actual size or shape of the limb, the rating 'distortion' is given. A rating of 'severe distortion' is given if one or more segments of the limb are missing. The total score is calculated by summing the individual scores of the seven items (Lewis and McCabe, 2010). The higher the score the greater is the degree of disturbance.

2.2.2.2 Pain assessment

The neuropathic pain symptom inventory (NPSI) (Bouhassira et al., 2004), a validated measure for the severity of neuropathic pain, was administered. The questionnaire determines subjective intensities (for the preceding 24 h) of spontaneous superficial, spontaneous deep, paroxysmal and evoked pain as well as paraesthesia. These different neuropathic symptoms are rated on an 11-point numerical rating scale. A total score is calculated by summing the five categories. Higher scores denote greater intensity.

2.2.2.3 Tactile acuity

As a measure of tactile acuity, a two-point discrimination test was undertaken on the distal pulp of the index fingers (IFs) of both the affected and unaffected hands according to Moberg's (1990) method. This test assesses the smallest distance at which an individual can clearly distinguish between two points applied simultaneously to the skin. With the participant's eyes closed, the aesthesiometer (Homecraft Rolyan, Nottingham, UK) was applied to the surface using the smallest possible distance between the two points until the skin blanched. The participant reported whether they felt the touch of one or two points. The distance between the two points was gradually increased for each trial until the participant correctly discerned two rather than one point. Three consecutive correct answers at the smallest discernable distance determined the two-point discrimination threshold (2ptDT). Sham trials involving stimulation of one point only were randomly included. Three threshold measurement trials were taken on each side. The mean of these trials was calculated for each IF.

2.2.2.4 Hand volume

Hand volume as a measure of swelling was included as an objective clinical sign of CRPS. The degree of swelling was recorded by measuring hand volume with a hand volumeter. The amount of lukewarm water displaced (measured in mL) when the whole hand up to the crease of the wrist was submerged into the volumeter was measured three consecutive times for both the affected and unaffected hands. Participants were instructed to align the crease of their wrist with the water line in order to minimize error. The mean of three trials for each hand was calculated.

2.3 Statistical analysis

2.3.1 Primary analyses

One-sided Pearson's correlation analyses were used to test the primary hypotheses that (1) pain and body perception disturbance are positively correlated and (2) decreased tactile acuity and body perception disturbance are positively correlated in the CRPS group. Tactile acuity ratios are presented in order to express affected hand tactile acuity relative to that of the unaffected hand. Ratios were calculated by dividing the mean of the 2ptDTs on the affected hand by the mean of the 2ptDTs on the unaffected hand.

To reduce the risk of false positive findings, outcome measure values that were equal to or greater than three SDs from the mean were considered outliers. Where such cases were identified, the values are presented in the findings and excluded from correlation analyses. To control for type 1 errors due to multiple comparisons, a Bonferroni correction (Abdi, 2007) was applied, resulting in a significance level of $\alpha = 0.025$.

2.3.2 Secondary analyses

Body perception disturbance and pain were hypothesized to be greater in patients than controls and tested using one-sided unpaired *t*-tests. Similarly, one-sided unpaired *t*-tests were applied to compare tactile acuity between patients and controls as tactile acuity is known to be impaired in CRPS (Pleger et al., 2005, 2006; Maihofner and Decol, 2007; Moseley et al., 2008c; Peltz et al., 2011). All data points were included in the group comparison analyses in order to capture the full range of clinical presentations. Outliers identified using the method previously described were excluded from correlation analyses.

Similar to the ratios for 2ptDT as described above, the ratio affected over unaffected side was calculated for hand volume. For the control group, the proportions of dominant and non-dominant hands were matched to the proportions in the CRPS group to obtain the same number of dominant (and non-dominant hands) in the nominator (and denominator) in both groups. This was performed to ensure that any group differences are not related to hand dominance.

A one-sided paired *t*-test was used to compare tactile acuity between affected and unaffected sides in the CRPS group. A two-sided paired *t*-test was used for the control group to compare dominant and non-dominant sides. A comparison of tactile acuity ratios between the groups utilized a one-sided test. Two-sided paired *t*-tests were used for hand volume in the patient as well as in the control group as there was no clear hypothesis about the direction of change. Pearson's correlation analyses were performed between pain and tactile acuity (one-sided), body perception disturbance and disease duration (two-sided), and body perception disturbance and hand volume (two-sided) to investigate potential relationships between these factors. Statistical significance levels for the secondary analyses were set to $\alpha = 0.05$, not corrected for multiple comparisons. All analyses were undertaken using PASW Statistics 17 (SPSS Inc., Chicago, IL, USA).

3. Results

Hypothesis 1: Body perception disturbance is positively correlated to pain

Correlation analysis revealed a significant linear relationship between body perception disturbance and pain (r = 0.57, one-sided test, $p < 0.01$) (see Fig. 1A).

Hypothesis 2: Body perception disturbance is correlated with tactile acuity impairment

An outlier with a 2ptDT ratio value of ≥4 (three SDs above the mean) (ID10, 2ptDT ratio = 4.33) was excluded from the analysis. 2ptDT was not performed on ID18 due to an amputated distal pulp of the left IF. A significant correlation was found between body perception disturbance and 2ptDT (r = 0.5, one-sided test, $p < 0.025$) (see Fig. 1B), indicating a positive relationship between impairment of tactile acuity and body perception disturbance.

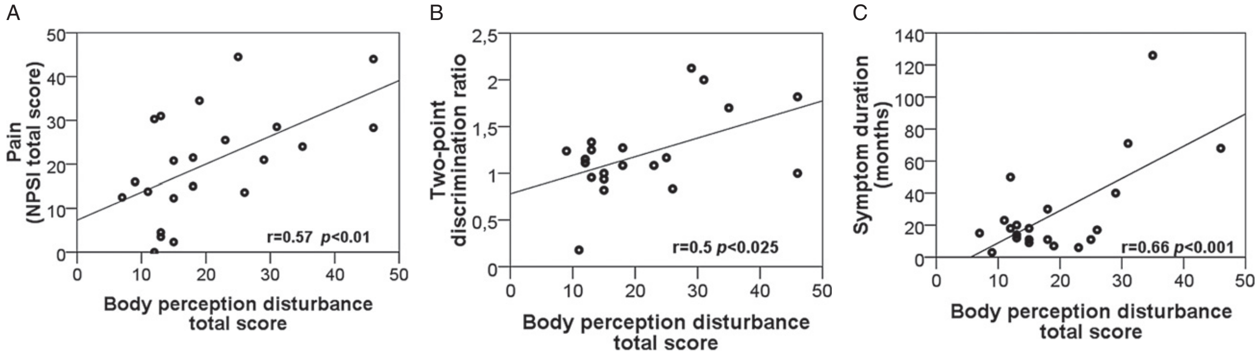

Figure 1 (A) Scatter plot illustrating the relationship between body perception disturbance and pain. (B) Scatter plot illustrating the relationship between body perception disturbance and tactile acuity. (C) Scatter plot illustrating the relationship between the extent of body perception disturbance and symptom duration. NPSI, neuropathic pain symptom inventory.

3.1 Secondary analyses (Table 2)

3.1.1 Body perception disturbance

Body perception disturbance for the CRPS group (mean = 20.5, SD = 11.1, min–max 7–46) was significantly worse (one-sided *t*-test, $p < 0.0001$, t = 5, df = 42) than for the healthy control (HC) group (mean = 7.8, SD = 4.1, min–max 0–15) as determined by the total score of the Bath CRPS body perception disturbance scale (Lewis and McCabe, 2010). The respective item of the scale revealed that 16 (73%) of the CRPS group perceived their affected hands to be different in size (either larger or smaller) compared with the unaffected hand. None of the HC group expressed abnormalities in the size perception of their hands. Details of body perception disturbance scores can be found as online supplementary material (Supporting Information Table S1).

3.1.2 Pain assessment

The NPSI total score was significantly higher (one-sided *t*-test, $p < 0.0001$, t = 7.6, df = 42) in the CRPS group (mean = 20.34, SD = 12.5, min–max 0–44.5) than in the HC group (mean = 0.23, SD = 0.75, min–max 0–3). Hence as one would expect, those with CRPS had significantly greater pain than healthy participants.

Individual CRPS participant scores are available as online supplementary data in Supporting Information Table S2.

3.1.3 Tactile acuity

A significant difference was found between the affected IF 2ptDT (mean = 4.2 mm, SD = 1.8 mm) and the unaffected IF (mean = 3.4 mm, SD = 0.97 mm) in the CRPS group (one-sided *t*-test, $p < 0.05$, t = −1.95, df = 20). There was no significant difference between HC dominant IF 2ptDT (mean = 3.35 mm, SD = 0.63 mm) and non-dominant IF (mean = 3.4 mm, SD = 0.65 mm) (two-sided *t*-test, $p > 0.05$, t = −0.55, df = 21).

Between-group comparison of 2ptDT ratios revealed a significant difference (one-sided *t*-test, $p < 0.05$, t = 2.1, df = 41) between the CRPS group

Table 2 Secondary analysis. Comparison of outcome measures between CRPS and healthy controls.

Group	Bath CRPS body perception disturbance total score	2pt discrimination ratio	Pain NPSI total score	Volumeter ratio
CRPS				
Mean (SD)	20.5 (11.1)	1.35 (0.8)	20.34 (12.5)	0.98 (0.08)
Min–max	7–46	0.18–4.33	0–44.5	0.73–1.12
HC				
Mean (SD)	7.8 (4.1)	0.98[a] (0.15)	0.23 (0.75)	0.98[a] (0.07)
Min–max	0–15	0.53–1.22	0–3	0.87–1.12
Significance	$p < 0.0001$	$p < 0.05$	$p < 0.0001$	$p > 0.05$

CRPS, complex regional pain syndrome; SD, standard deviation; HC, healthy control; NPSI, neuropathic pain symptom inventory.
[a]Proportions of dominant/non-dominant hand ratios matched to CRPS group.

(mean = 1.35, SD = 0.8, min–max 0.18–4.33) and the HC group (mean = 0.98, SD = 0.15, min–max 0.53–1.22).

3.1.4 Hand volume

There was no difference in hand volume ($p > 0.05$, t = 1.2, df = 21) between the affected (mean = 286 mL, SD = 75 mL, min–max 179–425 mL) and unaffected hands (mean = 292 mL, SD = 75 mL, min–max 181–449 mL) in the CRPS group. Furthermore, no difference in hand volume ratios (two-sided *t*-test, $p > 0.05$, t = –0.9, df = 42) was found between the CRPS group and HC group (CRPS ratio = 0.98, SD = 0.08, min–max 0.73–1.12; HC ratio = 0.98, SD = 0.07, min–max 0.87–1.12).

3.1.5 Pain and tactile acuity

There was no significant correlation between pain and tactile acuity (r = 0.31, one-sided test, $p > 0.05$).

3.1.6 Body perception disturbance and disease duration

An outlier with a disease duration value ≥217.34 months (three SDs above the mean) was identified (ID06, disease duration = 240 months) and removed prior to correlation analysis. A significant correlation (r = 0.66, two-sided test, $p < 0.001$) was found between body perception disturbance and disease duration among those with CRPS such that those with longer disease duration had more extensive disturbances in body perception (see Fig. 1C).

3.1.7 Hand volume and body perception disturbance

No significant correlation was found between body perception disturbance total score and hand volume (r = –0.26, two-sided test, $p > 0.05$).

4. Conclusions

Findings from this study have confirmed our hypotheses that firstly, body perception disturbance is positively correlated with pain, and secondly, that decreased tactile acuity is positively correlated with body perception disturbance.

These findings will be explored within the context of current literature. A theoretical model is proposed and the implications for practice and future research will be discussed.

4.1 Body perception disturbance and pain

The notion of a link between pain and body representation in CRPS has been previously described by others but not empirically determined. Schwoebel et al. (2001, 2002) were the first to indicate a link between body perception and pain by interpreting their findings as suggestive of a disruption in body representation. Those with unilateral upper limb CRPS showed impaired reaction times to hand laterality tasks only on the affected side. The authors proposed that the process by which laterality of the pictured hand was determined involved recruiting the central body representation, or 'body schema', when mentally rotating the limb into the pictured position (Schwoebel et al., 2001, 2002). Pain in the affected hand was stated as influencing performance although no formal testing of this hypothesis has been reported (Schwoebel et al., 2001, 2002). Evidence that impaired reaction times in hand laterality tasks are related to how painful the patient predicted a movement to be if the movement was undertaken has also been provided (Moseley, 2004).

Features that we consider to be individual aspects of body perception disturbance have been evaluated by others in relation to pain. A feeling of foreignness as described by Förderreuther et al. (2004) is akin to a sense of disownership of the affected limb (Lewis et al., 2007). Förderreuther et al. (2004) found that foreignness did not correlate with evoked pain. Perceived size distortions, specifically perceived enlargement of the affected hand, were found to have no relationship with pain intensity (Moseley, 2005; Peltz et al., 2011).

In contrast to previous research, the construct 'body perception disturbances' as used in our study encompasses several aspects of self-perception about the affected limb. A comprehensive evaluation of body perception disturbance might provide a more accurate assessment of the phenomenon. This could explain why we detected a relationship between pain and body perception disturbance that had not been found when individual aspects of body perception were investigated, as discussed in the preceding paragraph.

Recent evidence suggests interplay between body perception and pain such that one may modulate the enhancement or deterioration of the other factor suggesting exciting potential for treatment. The illusion of a hand visually altered in size compared with reality has been shown to alleviate pain in CRPS and osteoarthritis (Moseley et al., 2008b; Preston and Newport, 2011). Similarly, in healthy volunteers, controlled studies have revealed that viewing visual size distortions of the hand to which noxious stimuli are applied has an analgesic effect. This analgesic effect is modulated by the direction of change in size, i.e. an enlarged hand enhances the analgesic affect (Longo et al., 2009; Mancini et al., 2011). However, whether body perception disturbances are induced and/or driven by pain or quite the reverse remains unclear.

4.2 Body perception disturbance and tactile acuity

We have shown that patients with a greater degree of body perception disturbance have worse tactile acuity than those with less disturbance. Our findings corroborate with a recent study showing that overestimation of affected hand size, one aspect of body perception disturbance, correlated with increased 2ptDTs (Peltz et al., 2011). A relationship between disturbances in mental representation of the body region and tactile acuity has been found in chronic back pain. Tactile thresholds were higher in the regions of their back that patients had difficulty in depicting through drawing compared with those areas that they were able to draw (Moseley, 2008a). Limb disownership, a further component of body perception disturbance, was also found to be related to tactile processing deficits in CRPS. Moseley and colleagues investigated judgements regarding the timing of tactile stimuli presented to both unaffected and affected sides at various asynchronies and found that tactile processing deficits were related to the extent of affected limb disownership (Moseley et al., 2009). However, a further CRPS study found no relationship between altered tactile processing and abnormal body perception: misidentification of affected digits on tactile stimulation without vision was not related to foreignness (Förderreuther et al., 2004).

A plausible explanation for the observed relationship between body perception disturbance and tactile acuity might be that cortical shrinkage of affected limb representation that is known to occur within SI (a region responsible for tactile discrimination) also contributes to alterations in the multi-modal central representation of the affected limb (Graziano and Botvinick, 2002; Juottonen et al., 2002; Maihofner et al., 2003, 2004; Haggard and Wolpert, 2005; Pleger et al., 2005, 2006). We propose that this distorted body schema represented in lower (SI) and higher order cortices in superior and inferior parietal regions influences an individual's perception of their affected body part. The combination of altered body schema coupled with anomalous SI representation in tactile perception may account for why tactile stimulation to the affected limb was more difficult to discriminate when disturbances in body perception were greater. These disrupted representations might also contribute to altered central representations of limb position given that impairments in affected limb positioning have been observed (Forderreuther et al., 2004; Lewis et al., 2007, 2010).

4.3 Tactile acuity and pain

It is well established in CRPS that those in greater pain have worse tactile acuity (Pleger et al., 2005, 2006; Maihofner and Decol, 2007; Moseley et al., 2008c). Albeit our findings and those of a recent study (Peltz et al., 2011) showed no significant correlation of tactile acuity and pain severity, other studies have found a robust association (Pleger et al., 2005, 2006; Moseley et al., 2008c). Pleger et al. (2005, 2006) found a correlation between two-point discrimination and sustained pain such that those in greatest pain (subjects rated their mean pain intensity experienced over 4 weeks on an 11-point numerical rating scale) had worse tactile acuity. Interestingly, current pain levels were found not to correlate with tactile acuity, suggesting that pain rated over a longer duration is a better predictor of tactile acuity. The pain measure used in the present study refers to the past 24 h and, hence, might explain why we did not find a relationship between tactile acuity and pain.

4.4 Perceived size and hand volume

Interestingly, despite perceived alterations in hand size there was no objective size difference in hand volume between affected and unaffected sides. These findings corroborate with other studies where no relationship between swelling and perceived enlargement of the CRPS hand was found (Moseley, 2005; Peltz et al., 2011). Hence, our findings provide further evidence that those with CRPS have an altered size perception of the affected limb that is at odds with reality.

4.5 Body perception and disease duration

The significant correlation observed in the present study between disease duration and body perception disturbance is in line with previous findings. A greater percentage of those expressing foreignness in Förderreuther's (2004) study had chronic disease (greater than 6 months from inciting event). Furthermore, a relationship between perceived enlarged hands and disease duration has been shown (Moseley, 2005; Peltz et al., 2011). Earlier work has indicated that body perception disturbances are expressed by patients soon after symptom onset (Lewis et al., 2007). The observation that body perception disturbances might worsen over time is of considerable clinical relevance.

Taken together, we have shown that those with greater disturbances in body perception have worse pain and poorer tactile acuity. On this basis, a theoretical model comprising body perception disturbance, pain and tactile acuity is proposed. We hypothesize that aberrant cortical reorganization is the central nervous system correlate of body perception disturbance and impaired tactile acuity. Although other cortical regions are believed to be involved, three important aspects related to SI support our hypothesis of how this relationship occurs. Firstly, SI is primarily responsible for the central representation of tactile acuity and contributes to the multi-modal representation of the affected part (Haggard et al., 2003; Haggard and Wolpert, 2005). Secondly, experimental manipulation in healthy volunteers provides evidence of changes in perceived body size and tactile acuity that are directly related to alterations in SI representation (Tegenthoff et al., 2005; Schaefer et al., 2007). Finally, primate research has shown that pain leads to alterations in SI neuronal response to tactile stimulation (Whitsel et al., 2010).

Integration of cognitive (thoughts and feelings about the limb), sensory discriminative and size perception aspects suggests a multi-representation of the affected part within the brain. Our model supports the proposed 'body matrix', a multi-level cortical representation of the body that directly interrelates multisensory, cognitive and homeostatic areas in order to maintain the integrity of the body (Moseley et al., 2012). Such a model provides an explanation for the interplay between body perception and tactile acuity and how this might be disrupted in chronic pain.

Further investigation particularly using brain imaging aimed at determining the cortical correlates of body perception disturbance and tactile acuity is required to test this hypothesis.

Changes in perception of the limb can occur rapidly and be initiated by peripheral stimulation (Gandevia and Phegan, 1999) or via visual manipulation of enlargement or reduction in limb size (Moseley et al., 2008b; Preston and Newport, 2011). Given that both peripheral and central pathophysiological changes occur in CRPS, it is unclear from which direction body perception disturbances are triggered.

The findings of the present study are clearly relevant to treatment. Although the majority of patients have disturbances in body perception (Galer and Jensen, 1999; Forderreuther et al., 2004), it is those in greater pain and/or with a longer disease duration who are more likely to experience extensive disturbances. However, given the subtle nature of these perceptual disturbances, careful, directed questioning accompanied by appropriate objective screening is necessary to reveal their presence (Lewis and McCabe, 2010). Considering that those with long-standing CRPS have greater body perception disturbance, there is a clear need for early identification and appropriate intervention to ameliorate symptoms as the condition progresses.

Clinical observations suggestive of poor engagement in rehabilitation indicate that providing corrective input to normalize attitudes and perceptions about the affected limb may facilitate engagement and improve rehabilitation outcomes. Thus, treatment interventions aimed at reducing body perception disturbance to alleviate pain appear important from a clinical perspective.

Recent research in both experimental and chronic pain has revealed how visually manipulating body perception of the painful body part can provide pain relief (Mancini et al., 2011; Preston and Newport, 2011). Although further controlled testing is required to determine whether this approach provides sustained relief, using innovative techniques to alter body perception presents exciting potential for the treatment of pain. That tactile discrimination training (TDT) reduces pain in CRPS is well understood (Moseley et al., 2008c; Moseley and Wiech, 2009). Given the relationship between tactile acuity and body perception disturbance, it would seem reasonable to predict that TDT may be beneficial in correcting body perceptual abnormalities. Furthermore, TDT has been shown to normalize cortical representations of the affected region in other chronic pain conditions (Flor et al., 2001; Flor, 2002). Similarly, in CRPS where cortical maps are altered, appropriate tactile inputs might correct central body representations which in turn could positively influence perceptions about the limb, thus facilitating functional gain.

A limitation of the present study is that although relationships have been identified, whether pain causes, perpetuates and/or is the consequence of body perception disturbance is yet to be elucidated. Prospective investigations would help to clarify which of these variables are causative. Furthermore, this study explored only three aspects of the disease, consequently findings cannot account for other as yet unknown factors that might be influencing these relationships. Lastly, it is also acknowledged that the HCs were slightly younger than the CRPS group.

In conclusion, we have shown that body perception disturbance positively correlates with pain and decreased tactile acuity. Future prospective studies involving brain imaging to clarify the role of cortical regions associated with the body matrix in these interactions may provide further insight into manipulating central representations with the aim of more effectively treating this recalcitrant condition.

Author contributions

J.L. is responsible for the integrity of the complete work from inception and design, acquisition of data, analysis, drafting and revisions to publication of the manuscript. P.S. has made a substantial contribution to the intellectual content, conception and design of the study, and revision of the manuscript.

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Supporting Information

Additional Supporting Information may be found in the online version of this article:

Table S1. Bath CRPS body perception disturbance scores by scale item.
Table S2. Outcome measures of CRPS participants.

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