Validation of the Bath CRPS Body Perception Disturbance Scale

Running title: CRPS Body Perception Disturbance Scale validity

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Highlights

- We validated the B-CRPS-BPDS, which measures alterations in body perception
- We propose a revised B-CRPS-BPDS, removing the item on attention
- The internal consistency, construct validity, and known group validity were good
- Revised scores related to pain intensity, fear of movement, depression, disability
- The r-B-CRPS-BPDS is a valid measure to assess body perception alterations in CRPS

Abstract

The Bath Complex Regional Pain Syndrome Body Perception Disturbance Scale ("B-CRPS-BPDS") measures alterations in body perception. We assessed its internal consistency, known group validity, construct validity, and associations with demographic and clinical characteristics. We also evaluated changes in, and baseline predictors of B-CRPS-BPDS scores at follow-up. We included people with CRPS (*N*=114) and pain-free controls (*N*=69). People with CRPS obtained higher scores than pain-free controls on all B-CRPS-BPDS items, except the item on attention. Because this item also had an insufficient corrected item-total correlation, we propose a revised B-CRPS-BPDS (r-B-CRPS-BPDS) excluding this item. The internal consistency of the r-B-CRPS-BPDS was good. The r-B-CRPS-BPDS showed a large positive relationship with "motor neglect-like symptoms", indicating good construct validity. The r-B-CRPS-BPDS showed positive relationships with pain intensity, fear of movement, depression, and upper limb disability. There were no independent relationships with handedness, affected side, affected limb, disease duration, CRPS severity score, tension, anger, fatigue, confusion, and vigour. Finally, r-B-CRPS-BPDS scores did not consistently change over time. Our results demonstrate the utility of the r-B-CRPS-BPDS for measuring body perception disturbances in CRPS.

Perspective: This article evaluates the validity of the Bath Complex Regional Pain Syndrome Body Perception Disturbance Scale ("B-CRPS-BPDS") in CRPS, and assesses relationships with demographic and clinical variables. The proposed revised B-CRPS-BPDS appears to be a valid measure of body perception disturbances in CRPS.

Key words: complex regional pain syndrome; chronic pain; body perception disturbances; neglect-like symptoms; Bath CRPS Body Perception Disturbance Scale

Introduction

Complex Regional Pain Syndrome (CRPS) is characterized by pain; and autonomic, sensory, and motor symptoms affecting one or more limb(s). An estimated 70-90% of people with CRPS report disturbances in their body perception.[20] These disturbances can involve mismatches between the actual and perceived position, size, weight, pressure, or temperature of the affected limb.[29] People with CRPS may also report that their affected limb feels detached from the rest of their body; or that they avoid looking at, experience negative emotions towards, or have a desire to amputate it. The ability to assess body perception disturbances in a standardized way is clinically relevant. Understanding the relationship between body perception disturbances and CRPS can only be achieved if body perception disturbances can be reliably measured.

The two most used questionnaires to measure body perception disturbance in CRPS are the neurobehavioral questionnaire,[14,15] assessing inattention to and disengagement from the affected limb, and the Bath CRPS Body Perception Disturbance Scale ("B-CRPS-BPDS"),[31] covering a broader range of body perception disturbances.[29,31] Although the B-CRPS-BPDS is widely used in different languages,[1,4,32,36,51,57] its psychometric properties have not been fully explored. In two studies (*N*=22 and 59), the internal consistency of the English B-CRPS-BPDS ranged from poor to good at different time points.[1,32] Several studies have evaluated the construct validity of the B-CRPS-BPDS. With regard to other measures that have been related to body perception disturbances,[20,51] B-CRPS-BPDS scores were associated with worse tactile acuity and higher scores on the neurobehavioral questionnaire.[32,36] However, a study using a French translation of the B-CRPS-BPDS (*N*=13; including fewer items than the original) found no relationship between B-CRPS-BPDS scores and the senses of limb position and movement.[4] Although these studies provide some evidence supporting the validity of B-CRPS-BPDS, no study assessed the validity of the individual items or investigated how well this measure discriminates between people with CRPS and pain-free individuals who are expected to have normal body perception.

Mixed results have been reported regarding the relationships between body perception disturbance as measured with the B-CRPS-BPDS and disease characteristics in people with CRPS. Positive relationships between body perception disturbance and pain intensity have been found in some studies,[32,51] but not others.[57] One study found positive relationships between body perception disturbances and depression, anxiety, stress, and quality of life.[51] A prospective study found that body perception disturbance decreased in the first 12 months following the onset of CRPS, along with a decrease of pain, CRPS severity, and disability.[1] However, cross-sectional studies found either no effect of disease duration[51,57] or greater body perception

disturbance in people with longer disease durations.[32] It is important to note that the latter findings do not mean these symptoms progress and worsen over time, as cases that have been resolved are not captured by these studies.[1] Indeed, reduced body perception disturbance has been reported following treatment for CRPS.[27,28]

The aims of the current study were to assess the following aspects of the English B-CRPS-BPDS: 1) internal consistency; 2) known group validity (i.e. comparing scores between people with CRPS and pain-free controls); 3) construct validity (i.e. assessing the relationship with the neurobehavioral questionnaire);[14] 4) independent associations with demographic and clinical variables, including age, sex, handedness, affected side, affected limb, disease duration, pain intensity, CRPS severity score, fear of movement, upper limb disability, and mood states; and 5) consistent changes in body perception disturbances over time and relevant clinical baseline predictors of these changes.

Methods

Data

In this retrospective study, we combined databases of eight different published and unpublished studies (Table 1). The sample size and included variables were based upon availability. Crucially, all included studies were conducted by the same research group and the procedure of B-CRPS-BPDS administration was the same across different researchers. Data was collected in the United Kingdom (UK) between October 2013 and February 2020. All participants gave informed written and verbal consent. All research was approved by the UK National Health Service Health Research Authority (REC references 12/SC/0557 and 18/LO/1430), and by the University of Bath Psychology Department Ethics Committee (PREC approval codes 16-333, 18-251, and 16-236), in accordance with the Declaration of the World Medical Association (www.wma.net).

Table 1. The eligibility criteria and number of participants included in the current study, listed per study for which the data was originally collected. Some participants took part in more than one study. The studies combined resulted in a total of 516 sessions for people with CRPS and 117 for pain-free controls. Also listed are relevant additional measures used for the current study.

Study	**Additional eligibility criteria for participants with CRPS**	***N* CRPS**	***N* pain-free**	**Additional measures**
Bultitude et al.[6]	CRPS-1 or 2 (clinical or research criteria)[a]	24	-	Profile of Mood States
Bultitude and Petrini[5]	CRPS-1 or 2 (clinical or research criteria)	22	-	-
Halicka et al.[18,19]	CRPS-1 (research criteria) in upper limb; current pain intensity ≥2/10; no physical impairment that would prevent treatment; aged 18-80 years	54 (6 sessions)	22	Profile of Mood States
Halicka et al., unpublished[b]	CRPS-1 or 2 (clinical or research criteria)	28	19	-
Stanton et al., unpublished[c]	CRPS-1 or 2 (research criteria); aged 18-80 years	25	-	Neurobehavioral questionnaire
Ten Brink et al.[55]	CRPS-1 or 2 (clinical or research criteria); aged 18-85 years	40	40	-
Vittersø, Buckingham, Halicka, et al.[60]	CRPS-1 or 2 (research criteria); sufficient arm strength to manoeuvre the tools	36	36	-
Vittersø, Buckingham, Ten Brink, et al.[59]	CRPS-1 (clinical or research criteria)[a] in upper limb; no pacemaker, spinal cord stimulator or similar devices; not pregnant or breastfeeding; no physical impairment that would prevent arm movements	17	-	Quick DASH, Neurobehavioral questionnaire

Abbreviation: CRPS, Complex Regional Pain Syndrome; Disabilities of the Arm, Shoulder and Hand (DASH).

[a]For these studies, two people were diagnosed with CRPS not otherwise specified.

[b]Unpublished material [Halicka M, Cousins O, Ten Brink AF, Vittersø AD, Proulx MJ, Bultitude JH. Visual attention bias in personal space in upper and lower limb CRPS. Manuscript in preparation]

[c]Unpublished material [Stanton TR, Jones H, Spence C, Bultitude J. Self-prioritisation of limbs in people with unilateral complex regional pain syndrome. Manuscript in preparation]

Participants

For all studies, the following inclusion criteria applied: all participants had to be aged over 18, have no visual deficits substantial enough to interfere with completing the tasks of the original study, have no history of neurological disorders (e.g. stroke) or epilepsy, and have sufficient fluency in written and verbal English to understand the instructions. Participants with CRPS were required to have received a diagnosis of CRPS affecting primarily one limb at least 3

months before the research session; and CRPS had to be the primary pain-related complaint. On the day of testing, they had to meet the Budapest diagnostic criteria for CRPS.[21,22] Pain-free controls could not have chronic pain (defined as having experienced pain on most days for 3 months or more). Pain-free controls were matched regarding age, sex, and handedness, either with an individual person with CRPS or as a group (i.e. this differed per study). Additional inclusion criteria per study are listed in Table 1.

We used longitudinal data from the same 35 participants collected on six occasions over 30 weeks as part of the RCT conducted by Halicka et al.[19]After the initial baseline session, the follow-up intervals were four weeks, six weeks (including two weeks of prism adaptation or sham treatment), 10 weeks, 18 weeks, and 30 weeks. As the study showed no differences between the two treatment groups regarding any of the outcome measures,[19] we did not dissociate between these groups in the current study.

A flowchart of data selection of people with CRPS for the current study is depicted in Figure 1. We included all participants for whom identifiable information was available and for whom the B-CRPS-BPDS was administered at least once. Several people with CRPS participated in multiple studies and/or research sessions. Some of these took place within 48 hours of each other. For example, some people who lived a significant distance from the university took part in one or more studies over two days while staying locally overnight. For the purpose of the current study, all questionnaires that were assessed within 48 hours of filling out a B-CRPS-BPDS, were considered to be part of the same research session (regardless of whether they were assessed as part of the same or a different original study). Next, we removed all consecutive sessions without data on any of the variables that were included in analysis of B-CRPS-BPDS scores over time. We specified which sample was analysed for each research question (see section ‘Statistical analyses’).

For the pain-free controls, there were 117 research sessions. Of these sessions, 14 were excluded because no identifiable information was available, 5 because no B-CRPS-BPDS was administered, and 29 because they were consecutive sessions of the same participant. This resulted in data of the B-CRPS-BPDS for 69, unique, pain-free participants.

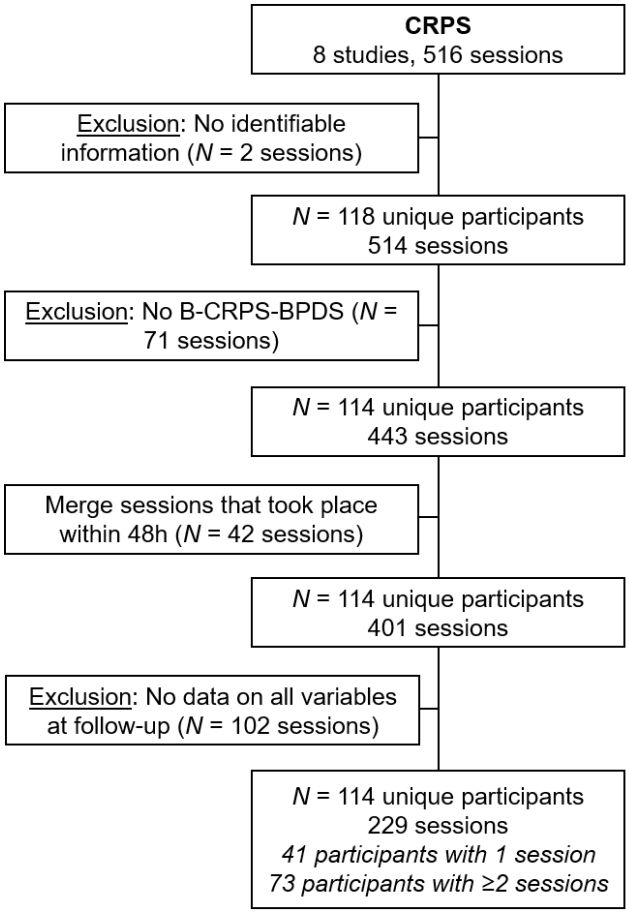

Figure 1. Flowchart of data collection for people with CRPS. We combined data that was collected as part of 8 different studies, including 516 research sessions. The left-hand boxes indicate the screening process. We removed sessions for which no identifiable information was present and in which no Bath CRPS Body Perception Disturbance Scale (B-CRPS-BPDS) was administered. Next, sessions that were administered within 48h from each other were merged. Finally, follow-up sessions without data on predictor variables for the regression model were removed. This resulted in a database of 114 unique participants with a total of 299 sessions.

Demographic and pain-related characteristics

We collected information on age, sex, self-reported handedness, affected limb (i.e. upper, lower), affected side (i.e. left, right), and the disease duration (i.e. the time since the inciting event or, if there was none, the pain onset). For all research sessions, we measured the current pain intensity on a scale from 0 to 10 using the Numeric Pain Rating scale.[25,49]

The Bath CRPS Body Perception Disturbance Scale (B-CRPS-BPDS)

The B-CRPS-BPDS was used to assess body perception disturbances.[31] The questionnaire has seven items that cover different aspects of body perception of the affected part (Table 2). Pain-

free control participants were matched with a CRPS participant and were instructed to answer the questionnaire items with respect to the limb that was affected in the CRPS participant. The sum of numerical ratings from items 1 to 4 and 6b are added to items 5 and 6a (scored no = 0, yes = 1). The mental representation drawing is graded on a three-point scale: no distortion = 0, distortion = 1, severe distortion = 2.[31] See Figure 2 for some example drawings and their scorings. The total score of the B-CRPS-BPDS ranges from 0 to 57, with higher scores indicating more severe body perception disturbances.

Table 2. Items of the Bath CRPS Body Perception Disturbance Scale (B-CRPS-BPDS), taken from Lewis and McCabe.[31]

Item	Question
1)	On a scale of 0-10 how much a part of your body does the affected part feel? Very much a part = 0_1_2_3_4_5_6_7_8_9_10 = Completely detached
2)	On a scale of 0-10 how aware are you of the physical position of your limb? Very aware = 0_1_2_3_4_5_6_7_8_9_10 = Completely unaware
3)	On a scale of 0-10 how much attention do you pay to your limb in terms of looking at it and thinking about it? Full attention = 0_1_2_3_4_5_6_7_8_9_10 = No attention
4)	On a scale of 0-10 how strong are the emotional feelings that you have about your limb? Strongly positive = 0_1_2_3_4_5_6_7_8_9_10 = Strongly negative
5)	Is there a difference between how your affected limb looks or is on touch compared to how it feels in terms of the following: Size yes □ no □ Comment_____ Temperature yes □ no □ Comment_____ Pressure yes □ no □ Comment_____ Weight yes □ no □ Comment_____
6a)	Have you ever had a desire to amputate the limb? Yes □ No □
6b)	If yes, how strong is that desire now? Not at all = 0_1_2_3_4_5_6_7_8_9_10 = Very strong Desired amputation site_____
7)	With eyes closed describe a mental image of your affected and unaffected body parts (drawn by assessor during patient description then verified by the patient)

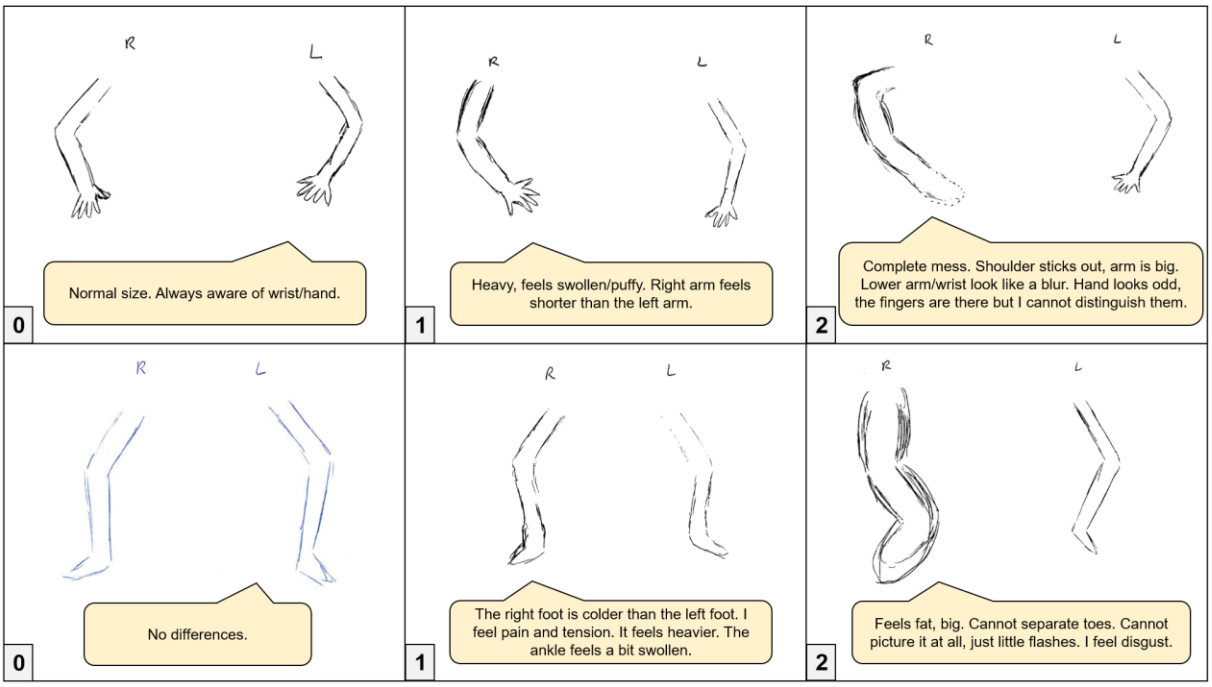

Figure 2. Examples of responses for item 7 of the Bath CRPS Body Perception Disturbance Scale, scored as 0 (left panels), 1 (central panels), or 2 (right panels), for both upper and lower limb patients with chronic limb pain (either CRPS or 'other' limb pain). The drawings were made based upon descriptions of participants, and adjusted until the participant confirmed that the drawing was an accurate depiction of the mental representation of their affected limb.

CRPS Severity Score

To assess whether or not they fulfilled the Budapest (clinical or research) criteria for CRPS, all participants with CRPS were interviewed about whether they had experienced eight symptoms, and we objectively quantified the presence or absence of eight signs of CRPS.[21,22] The CRPS severity score is a validated measure[22] that reflects the number of patient-reported symptoms and signs scored as present during examination (range 0-16). Higher scores indicate more CRPS signs and symptoms, which could be one indication of CRPS severity (although this measure does not attempt to encapsulate other possible severity indicators such as the intensity of individual signs or symptoms). The locations for sensory testing were the most painful site on the affected limb and the corresponding site on the unaffected limb. The procedures described below were skipped or adapted if the participant indicated that it was too painful or if the location would not allow using the standardized protocol (e.g. some locations could not be touched with a testing instrument held at a perpendicular angle).

Hypoesthesia (higher mechanical detection threshold) and hyperalgesia (lower mechanical pain threshold) were assessed by a single pinprick, and were indicated to be present

if the participant answered that the sensation was intensely painful or lasted longer than the duration of the pinprick itself (i.e. hypoesthesia); or that it felt numb or not painful at all (i.e. hyperalgesia). Allodynia (pain experienced from stimulation that normally does not cause pain) was examined using a cotton ball, Q-tip, and brush (MRC Systems PinPrick Stimulator Set), and/or a 128HZ tuning fork, and cold metal pen. Temperature asymmetry was measured using handheld infrared thermometer with an 8:1 distance to spot size ratio, and a red laser aim. An absolute difference between the affected and unaffected side greater than 1°C was classed as a temperature asymmetry.[21] We used the figure-of-eight procedure that uses a soft tape measure to quantify swelling of feet and ankles[44,54] or hands and wrists.[34,43] If locations other than the hand or foot were affected, swelling was assessed by visual inspection by the experimenter. Asymmetries in skin colour, sweating, and changes in hair and nail growth were assessed by visual inspection by the experimenter. Motor weakness for the upper limb was assessed using an electronic hand-held dynamometer, or by asking participants to squeeze as hard as possible the index and middle fingers of the researcher who compared the strength between hands. Motor weakness for the lower limb was either subjectively assessed (e.g. scored as being present if a participant was not able to stand on their feet without aid) or assessed using a muscle strength grading system (i.e. moving against gravity, resist moderate force, resist large force). In all studies, we used a goniometer to quantify inversion, eversion, flexion, and extension of the ankle; or radial, ulnar, flexion, and extension of the wrist; or a visual assessment of the same movements. In addition to confirming the presence or absence of CRPS, we computed a CRPS severity score by counting how many of 16 symptoms and signs were present.[21,22]

Neurobehavioral questionnaire

The neurobehavioral questionnaire was assessed in two out of eight studies. It is a 5-item self-report measure developed by Galer and Jensen,[14] which was further developed by Frettlöh et al.[13] to include the addition of a Likert-scale rather than dichotomous responses. The neurobehavioral questionnaire measures inattention for and disengagement from the most painful limb (i.e. "motor neglect-like symptoms"), including items such as "My painful limb feels as though it is not part of the rest of my body". Detachment from and unawareness of the most painful limb are aspects of body perception disturbances that are also measured with the B-CRPS-BPDS.

To record the severity of the symptoms, the studies included here used either a five-option or six-option Likert-scale ranging from 1 ('never') to 5 or 6 ('always'), respectively. To be able to compare scores with different scales, we rescaled the step size of 1 into 0.80 for the

six-option Likert scale, creating steps of 1, 1.8, 2.6, 3.4, 4.2, and 5. As the item on involuntary movements measures a different construct than the other items, we computed the average score of the four items on cognitive and motor neglect-like symptoms (items 1, 2, 3, and 5), and separately assessed the score of the involuntary movements (item 4).

Fear of movement

The Tampa Scale for Kinesiophobia (TSK) was administered to assess fear of movement.[38] Participants judge whether they agree with 17 statements, such as "I'm afraid that I might injure myself if I exercise" on a scale from 1 ('strongly disagree') to 4 ('strongly agree'). The total score ranges from 17 to 68, with higher scores reflecting greater fear of movement. In a population of chronic low back pain patients, the TSK is a reliable instrument with good internal consistency, substantial test-retest reliability, and good construct validity.[9,52,61]

Upper limb disability

In one out of eight studies, upper limb disability was measured. Only people with upper limb CRPS were included in this study. Therefore, we did not have a measure for lower limb disability. Upper limb disability was measured with the Quick Disabilities of the Arm, Shoulder and Hand (DASH) questionnaire.[17] The Quick DASH consists of 11 items assessing whether participants have difficulty with, or are limited in, daily activities and sleep, and whether they experienced symptoms such as pain or tingling sensations. Participants score each item ranging from 1 ('no difficulty/not limited/no symptoms') to 5 ('unable/extreme symptoms'). The Quick DASH total score is computed with the following formula: ([the sum of all responses / the number of completed responses] – 1) * 25. This results in a total score ranging from 0 to 100, with higher scores indicating more severe disability. The validity, test-retest reliability, and responsiveness of the Quick DASH have shown to be good.[2,17]

State of mood

Two out of eight studies included the Profile of Mood States questionnaire[35] to assess the short-term mood state. Participants indicate for 65 words/statements how they have been feeling in the past week, including the day of testing. Participants score each item on a 5-point Likert Scale, ranging from 1 ('not at all') to 5 ('extremely'). Items are grouped into six mood profiles with different score ranges: anger (0-48), confusion (0-28), depression (0-60), fatigue (0-28), tension (0-36), and vigour (0-32). For all profiles, higher scores indicate greater mood disturbance. The construct validity and internal consistency of the mood profiles have shown

to be good.[3,50] The factorial validity of the Profile of Mood States questionnaire remains in doubt, mainly caused by the confusion profile which is not always a factor.[3,50] Therefore, results based upon this profile should be interpreted with caution.

Statistical analyses

The level of alpha was set at 0.05. Analyses were conducted in IBM SPSS Statistics version 27.[23] Effect sizes were quantified as Pearson correlation coefficients. Pearson correlation coefficients of >0.10 were considered to reflect a small, >0.30 a medium, and >0.50 a large effect.[11]

Demographic and pain-related characteristics

We provided descriptive data on demographic and pain-related characteristics of the participants, on the day that they filled out the B-CRPS-BPDS for the first time. We conducted *t*-tests and Chi-square tests to compare the CRPS and pain-free group regarding demographic characteristics.

Internal consistency of the B-CRPS-BPDS

We computed the internal consistency for the B-CRPS-BPDS per group (i.e. CRPS and pain-free) and for the total group. For participants who had participated in multiple research sessions, we used the first B-CRPS-BPDS administration. A Cronbach's alpha of ≥0.70 was considered acceptable. Per item, we computed the item-total correlations, for which values of ≥0.30 are recommended.[11] We only kept items with a corrected item-total correlation of ≥0.30 in the total group, and included these items in the subsequent analyses. The scale without items that were dropped was called the revised B-CRPS-BPDS (r-B-CRPS-BPDS).

Known group validity of the r-B-CRPS-BPDS

We assessed the discriminative value per item, and for the entire scale. We expected people with CRPS to have higher body perception disturbance scores than pain-free controls. For participants who had taken part in multiple research sessions, we used the first B-CRPS-BPDS administration. As several assumptions for parametric testing were violated, we used non-parametric tests. We performed Mann Whitney tests to compare groups per item and regarding the total score, and a Chi-square test to compare groups regarding item 6a (i.e. "Have you ever had a desire to amputate the limb").

Construct validity of the r-B-CRPS-BPDS: relationship with "motor neglect-like symptoms"

We assessed the relationship between the r-B-CRPS-BPDS and the "motor neglect-like symptoms" as measured with the neurobehavioral questionnaire. We expected these scales to be strongly related as they are thought to measure a similar construct.[20,51] We included all people with CRPS for whom the neurobehavioral questionnaire was administered. For participants for whom this was the case for multiple research sessions, we used the first session. We performed Spearman correlations. Spearman's rho was interpreted as small (>0.10), moderate (>0.30), large (>0.50), or very large (>0.70).[10]

Predictors of the r-B-CRPS-BPDS

We conducted linear regression analyses with r-B-CRPS-BPDS total score as the dependent variable. We did not include all variables in one model because we did not have data for the same variables for all participants. For each model, a series of bivariate linear regression analyses were first undertaken to examine associations between each of the potential predictors and the r-B-CRPS-BPDS total score. We then performed a multiple regression analysis, in which all baseline variables with *p*-values up to 0.10 in the bivariate analysis were included at once. This selection was made to maintain statistical power in this subsample. Beforehand, independent variables were checked for collinearity. The enter method was used to restrict the risk of capitalizing on chance features of the data. We included all participants with CRPS for whom data on all of the predictor variables were available. If this was the case for multiple research sessions, we only included the first session. Note that we choose to only include one research session per participant and did not include all sessions in one model, because there was too much missing data on the relevant clinical variables on consecutive research sessions. In other words, we selected the most complete research session for eact participant and included this session in the model.

First, we assessed relationships with demographic and pain-related characteristics, including age, sex (i.e. male, female), handedness (i.e. left, right), affected side (i.e. left, right), affected limb (i.e. upper, lower), disease duration in years, pain intensity, CRPS severity score, and fear of movement (TSK) as potential predictors. Second, we assessed relationships with states of mood as measured with the Profile of Mood States (tension, depression, anger, fatigue, confusion, and vigour). Pain intensity was included again since this variable was available for all participants with the Profile of Mood States scores and in this way we could control for the effect of pain intensity. For upper limb disability (i.e. Quick DASH) we only had data for a

small number of people with CRPS (N = 17). Therefore, we only performed Spearman correlations between this factor and the r-B-CRPS-BPDS.

Changes in r-B-CRPS-BPDS scores over time

For a subset of people with CRPS, there was data on the r-B-CRPS-BPDS for multiple time points. We were interested in whether there was a consistent increase or decrease of r-B-CRPS-BPDS scores over time. To this aim, we included people with CRPS for whom more than one research session was available. A linear mixed effects model analysis was performed with the baseline r-B-CRPS-BPDS score and the variable 'time' (i.e. the first assessment was referred to as time point '0', all consecutive assessments were referred to as the number of days since the first assessment) as potential predictors, and the r-B-CRPS-BPDS total score at subsequent research sessions as dependent outcome measure. This approach is appropriate for evaluating repeated measures in a heterogeneous group and allows the variable 'time' to be treated as a continuous measure (which is necessary given differences between participants in both the number of total sessions and the time intervals between the sessions).[16]

In the same analysis, we assessed which other variables at baseline (i.e. next to the baseline r-B-CRPS-BPDS score) predicted future r-B-CRPS-BPDS scores. We used clinical and demographic data from the baseline session in order to derive clinically relevant predictors of changes of body perception disturbances.

The linear mixed effects model used a heterogeneous first-order autoregressive covariance structure and included a random intercept for each participant. The effects of theoretical interest were time (i.e. days since baseline), r-B-CRPS-BPDS score at baseline, age at baseline, sex (i.e. male, female), handedness (i.e. left, right), affected side (i.e. left, right), affected limb (i.e. upper, lower), disease duration in years at baseline, pain intensity at baseline, and TSK at baseline. Because there was 31.4% missing data for the CRPS severity score, this variable was not included as a predictor. There was no missing data for the other variables. The baseline r-B-CRPS-BPDS score was included in the basic model and the other variables were introduced as fixed effects in the basic model one by one. To statistically compare the fit of each new model with the old model, we assessed the change in -2 log-likelihood ($\chi^2_{Change} = -2LL_{Old} - -2LL_{New}$) in light of the number of additional parameters ($df_{Change} = k_{Old} - k_{New}$).[11] The coefficients of the best performing model were reported (thus, not all possible predictors were included, as this depended on their statistical significance).

Results

Demographic and pain-related characteristics

There were 114 unique participants with CRPS and 69 who were pain-free. Table 3 lists the demographic and pain-related characteristics split per group. The CRPS and pain-free group did not differ from each other regarding age, sex, and handedness

Table 3. Demographic and pain-related characteristics of all unique participants, at the time of the first Bath CRPS Body Perception Disturbance Scale (B-CRPS-BPDS) assessment. Means (SD) and frequencies (%) are depicted. Note that we did not have data on all characteristics for all participants at the first testing day.

	N	**CRPS**	***N***	**Pain-free**	**Statistical comparisons between groups**
Age, years	114	46.06 (13.52)	69	46.61 (14.37)	$t(181) = -0.26$, $p = .794$
Sex, female	114	93 (81.6%)	69	55 (77.9%)	$\chi^2(1) = 0.10$, $p = .755$
Handedness, right	112	102 (91.1%)	69	60 (87.0%)	$\chi^2(1) = 0.77$, $p = .380$
Affected side, right	114	48 (42.1%)	-	-	
Affected limb, upper	114	76 (66.7%)	-	-	
Disease duration, years	111	5.60 (6.41)	-	-	
CRPS severity score (0-16)	80	12.39 (1.59)	-	-	
Pain intensity (0-10)	107	6.04 (2.13)	-	-	

Abbreviations: CRPS, Complex Regional Pain Syndrome.

Internal consistency of the B-CRPS-BPDS

Table 4 shows that for the total group and the CRPS group, the internal consistency of the B-CRPS-BPDS was acceptable (Cronbach's alpha >0.70). For the pain-free controls, Cronbach's alpha was actually negative, due to a negative average covariance among items. Item 3 ("how much attention do you pay to your limb…") showed an item-total correlation below 0.30 in the total group, and the lowest item-total correlation in the CRPS group (0.34). In addition, this was the only item for which the pain-free controls obtained higher scores than the CRPS group (see "Known group validity of the r-B-CRPS-BPDS").

Therefore, we computed a revised total score without this item which we used in the subsequent analyses (r-B-CRPS-BPDS; see supplementary material for the revised scale). To compute this score, the sum of numerical ratings from (newly numbered) items 1 to 3 and 5b are added to items 4 and 5a (scored no = 0, yes = 1). The mental representation drawing (item 7 in the original scale, item 6 in the revised scale) also had somewhat lower item-total correlation in the CRPS group (0.35) than the other remaining items (>0.40). However, we

retained this item in the r-B-CRPS-BPDS because the item-total correlation in the total group (0.65) did not differ markedly to that for the remaining items. This item is graded on a three-point scale; no distortion = 0, distortion = 1, severe distortion = 2. The total score of the r-B-CRPS-BPDS therefore ranges from 0 to 47, with higher scores indicating more body perception disturbances. Cronbach's alpha for the revised total score (without former item 3) was 0.80 for the total group, indicating good internal consistency. For clarity, we use the numbering from the original scale in the remainder of this paper.

Table 4. The internal consistency of the Bath CRPS Body Perception Disturbance Scale (B-CRPS-BPDS): Cronbach's alpha and the item-total correlation per item, split per group.

	CRPS (*N* = 114)	**Pain-free (*N* = 69)**	**Total group (*N* = 183)**
Cronbach's alpha total score	0.73	-0.01	0.75
Cronbach's alpha revised total score (i.e. excluding item 3)	0.73	-0.23	0.82
Item-total correlations			
1. Detachment	0.66	-0.06	0.74
2. Awareness	0.54	0.02	0.53
3. Attention	0.34	0.11	-0.04
4. Emotional feelings	0.45	-0.10	0.57
5. Size/temp./press/weight	0.43	0.01	0.65
6a. Amputation	0.50	*	0.67
6b. Amputation degree	0.61	*	0.68
7. Mental image	0.35	-0.02	0.65

Abbreviation: Complex Regional Pain Syndrome, CRPS.

* Item 6a and item 6b were all answered with 'No' or '0' respectively, by the pain-free controls.

Known group validity of the r-B-CRPS-BPDS

Figure 3 depicts the B-CRPS-BPDS item scores and the r-B-CRPS-BPDS total score split per group. In Table 5, the group comparisons are depicted. People with CRPS had higher scores than pain-free controls on all items and the revised total score, except for item 3, for which they had lower scores (medium effect size). Effect sizes were large for items 1, 4, 5, 7, and the revised total score; and medium for items 2 and 6b.

Table 5. Statistics for the non-parametric group comparisons per item of the B-CRPS-BPDS, and the revised total score (i.e. excluding item 3). Groups are people with CRPS (N = 114) and pain-free controls (N = 69).

Item	Statistical comparisons between groups
1. Detachment	U = 769, p < .001, r = -0.70
2. Awareness	U = 2260, p < .001, r = -0.37
3. Attention	U = 2493.5, p < .001, r = -0.31
4. Emotional feelings	U = 1045, p < .001, r = -0.62
5. Size/temp./press/weight	U = 195.5, p < .001, r = -0.83
6a. Amputation	$\chi^2(1) = 75.20$, p < .001
6b. Amputation degree	U = 2001, p < .001, r = -0.50
7. Mental image	U = 249, p < .001, r = -0.85
Revised total score	U = 257.5, p < .001, r = -0.78

Group scores were compared with Mann Whitney Tests and a Chi-Square test. The odds ratio could not be computed for comparisons of item 6a with the pain-free controls, as none of the pain-free controls answered 'yes' to this item.

Abbreviations: Complex Regional Pain Syndrome, CRPS; odds ratio, OR.

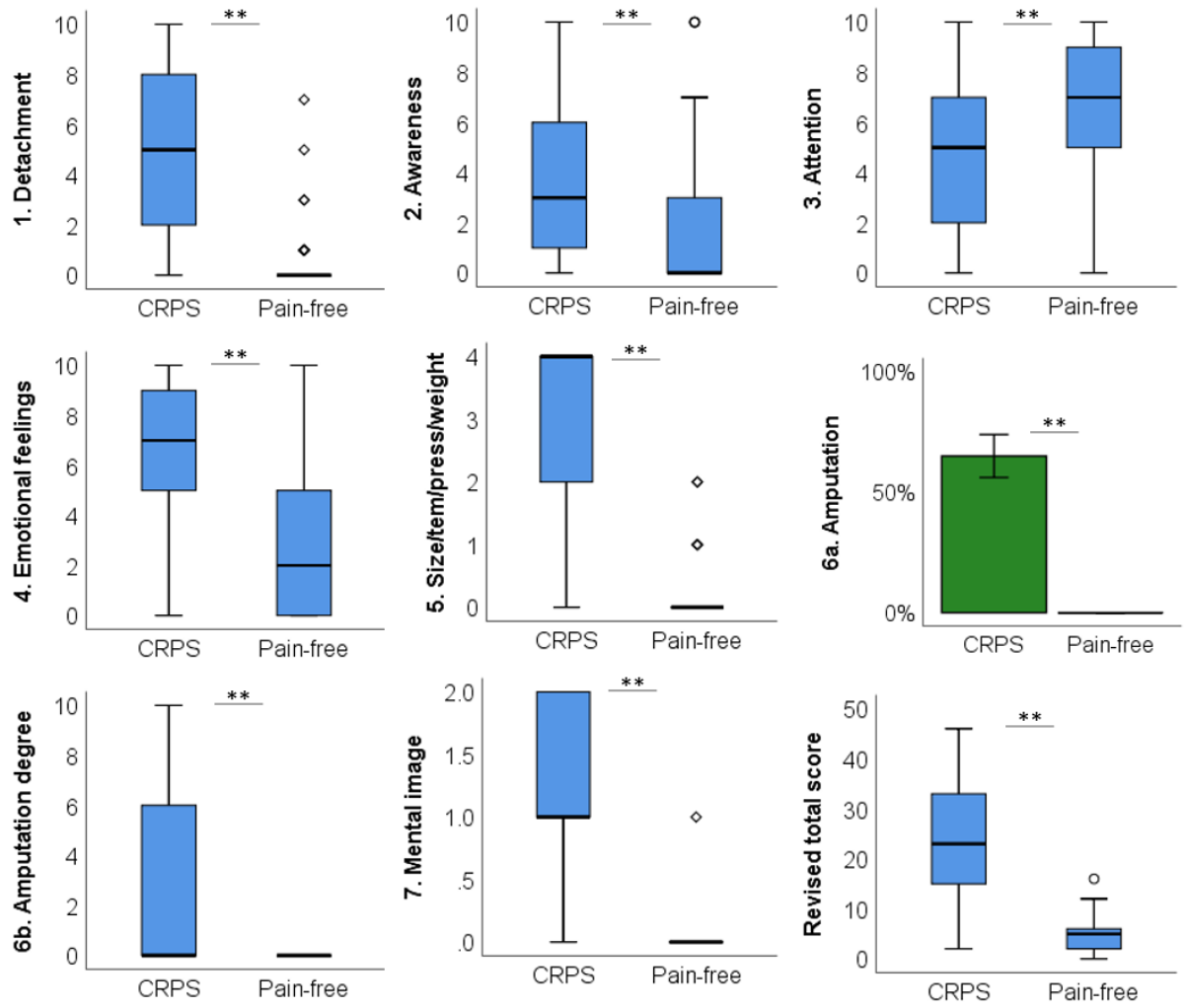

Figure 3. Scores for items 1, 2, 3, 4, 5, 6b, and 7 of the Bath CRPS Body Perception Disturbance Scale; and the total score for the revised Bath CRPS Body Perception Disturbance Scale (sum of all original items except item 3, scores ranging from 0 to 47). The thick line in the middle is the median. The top and bottom box lines show the first and third quartiles. The whiskers show the maximum and minimum values, with the exceptions of outliers (circles) and extremes (diamonds). The percentage of participants who answered 'yes' on item 6a is depicted in bar graphs; error bars depict 95% confidence intervals. ** Asterisks indicate that groups differed with a significance level of $p < .001$. CRPS = Complex Regional Pain Syndrome.

Construct validity of the r-B-CRPS-BPDS: relationship with "motor neglect-like symptoms"

The sample of people with CRPS and data on the neurobehavioral questionnaire consisted of 37 people. Their average score on the r-B-CRPS-BPDS was 22.45 (*SD* = 10.42). Their average score on cognitive and motor neglect-like symptoms was 3.15 (*SD* = 1.08), and on the item on involuntary movements 2.86 (*SD* = 1.41).

The r-B-CRPS-BPDS showed a very large positive relationship with motor and cognitive neglect-like symptoms (r = 0.73, p < .001); and a large positive relationship with involuntary movements (r = 0.51, p < .001). This supports the idea that the r-B-CRPS-BPDS and the neurobehavioral questionnaire assess a similar construct.

Predictors of the r-B-CRPS-BPDS

Relationship between the r-B-CRPS-BPDS and demographic and pain-related characteristics

The sample consisted of 79 people with CRPS. Table 6 shows the outcome of the bivariate and multiple regression analyses. Based on the bivariate regression outcomes, sex, affected limb, CRPS severity score, pain intensity, and fear of movement (TSK) were considered for the multiple regression model. The VIF values were all ≤1.17, indicating lack of collinearity.

The multiple regression model explained 32% of the variance. Pain intensity and fear of movement (TSK) were positively associated with the r-B-CRPS-BPDS total score and explained 8% and 9% of the variance, respectively.

Table 6. Outcome of the bivariate and multiple regression model of revised Bath CRPS Body Perception Disturbance Scale (r-B-CRPS-BPDS) total scores in people with CRPS, with **demographic and pain-related characteristics** as independent variables (N = 79). Significant predictors of the multiple regression are depicted in bold.

	Bivariate			Multiple		
Predictors	*B* (95% CI)	*p*	R^2	*B* (95% CI)	*p*	R^2
Sex (female)	-6.20 (-12.31 to -0.10)	.047	.05	-2.42 (-8.01 to 3.18)	.392	.01
Age	-0.06 (-0.24 to 0.12)	.540	.01			
Handedness (right)	2.63 (-5.77 to 11.03)	.535	.01			
Affected limb (lower)	5.29 (-0.28 to 10.87)	.062	.04	1.72 (-3.35 to 6.80)	.501	.01
Affected side (right)	-0.97 (-5.84 to 3.90)	.692	0			
Disease duration	-0.09 (-0.43 to 0.25)	.602	0			
CRPS severity score	1.79 (0.59 to 3.00)	.004	.10	1.00 (-0.13 to 2.13)	.082	.04
Pain intensity	**2.01 (1.00 to 3.01)**	**< .001**	**.17**	**1.30 (0.29 to 2.31)**	**.012**	**.08**
TSK	**0.52 (0.27 to 0.77)**	**< .001**	**.18**	**0.34 (0.09 to 0.60)**	**.009**	**.09**

Abbreviations: Complex Regional Pain Syndrome, CRPS; Tampa Scale for Kinesiophobia, TSK.

Relationship between the r-B-CRPS-BPDS and state of mood

We included 75 people with CRPS for whom data on the Profile of Mood States was available. Table 7 shows the outcome of the bivariate and multiple regression analyses. Based on the

bivariate regression outcomes, all mood profiles except vigour were considered for the multiple regression model. The VIF values were all ≤4.78, indicating lack of collinearity.

The multiple regression model explained 50% of the variance. Pain intensity and depression were positively associated with the r-B-CRPS-BPDS total score and explained 19% and 28% of the variance, respectively.

Table 7. Outcome of the bivariate and multiple regression model of revised Bath CRPS Body Perception Disturbance Scale (r-B-CRPS-BPDS) total scores in people with CRPS, with **pain intensity and mood profiles (Profile of Mood States)** as independent variables (N = 75). Significant predictors of the multiple regression are depicted in bold.

	Bivariate			Multiple		
Predictors	*B* (95% CI)	*p*	R^2	*B* (95% CI)	*p*	R^2
Pain intensity	**2.55 (1.43 to 3.67)**	**< .001**	**.22**	**1.97 (-1.83 to 11.99)**	**< .001**	**.19**
POMS Tension	0.54 (0.25 to 0.83)	< .001	.16	-0.24 (-0.74 to 0.26)	.343	.01
POMS Depression	**0.46 (0.31 to 0.61)**	**< .001**	**.33**	**0.73 (0.45 to 1.02)**	**< .001**	**.28**
POMS Anger	0.41 (0.17 to 0.65)	.001	.14	-0.28 (-0.64 to 0.09)	.133	.03
POMS Fatigue	0.48 (0.19 to 0.78)	.002	.13	-0.07 (-0.40 to 0.27)	.696	0
POMS Confusion	0.29 (-0.02 to 0.60)	.070	.04	-0.11 (-0.42 to 0.21)	.502	.01
POMS Vigour	-0.22 (-0.64 to 0.21)	.310	.01			

Abbreviations: Complex Regional Pain Syndrome, CRPS; Profile of Mood States, POMS.

Relationship between the r-B-CRPS-BPDS and upper limb disability

We included 17 people with CRPS affecting an upper limb for whom data on the Quick DASH was available. There was a very strong positive relationship between the r-B-CRPS-BPDS and upper limb disability as measured with the Quick DASH (r = .85, p < .001).

Changes in r-B-CRPS-BPDS scores over time

We included 73 people with CRPS, for whom 2 to 10 research sessions were available (median = 5), resulting in a total of 258 research sessions. The time between the first and last research session ranged from 25 days to 5.7 years.

The final model included baseline r-B-CRPS-BPDS score, $F(1, 67.98) = 114.61$, p < .001, with a Beta coefficient of 0.74 (SE = 0.07, 95%CI = 0.60 to 0.87, p < .001). This was the only significant predictor of consecutive r-B-CRPS-BPDS scores. Thus, time was not a significant predictor of r-B-CRPS-BPDS scores, indicating that there was no consistent trend of decreasing or increasing r-B-CRPS-BPDS scores over time.

Discussion

This study aimed to examine the psychometric properties of the B-CRPS-BPDS. First, we assessed the internal consistency of the individual items and found that all item-total correlations were acceptable, except for the item on attention. Second, we evaluated the discriminative value of the individual items of the B-CRPS-BPDS. For all items, except that on attention, people with CRPS obtained higher scores than pain-free controls.

In our experience, the item on attention is not sufficiently specific. People with CRPS can exhibit guarding of their CRPS-affected limb[40] and/or hypervigilance to any object that might approach it. However, many patients simultaneously report a neglect-like disregard of their CRPS-affected limb. Thus, some participants expressed indecisiveness about this item. In the German version of the B-CRPS-BPDS, this item is worded in terms of how much attention someone pays to their affected limb *when* they look at it or think about it (instead of *in terms of* looking at it or thinking about it). This subtle difference in phrasing might be the reason why the internal consistency of the German version (including the item on attention) is much higher (Cronbach's alpha = 0.92) than seen in previous studies.[57] We propose a revised B-CRPS-BPDS (r-B-CRPS-BPDS) excluding the item on attention (see supplementary material for a downloadable version with scoring instructions). Alternative approaches would be to investigate whether rewording this item in the English B-CRPS-BPDS improves its validity, or to create two separate items that independently capture hypervigilance towards, and inattention for the CRPS-affected limb(s). If a new scale is validated, other items could also be rephrased with clearer wording for patients to understand (e.g. item 4). However, even with the current wording, the r-B-CRPS-BPDS has excellent utility. The internal consistency of the revised version was good in people with CRPS. Given that the scale was developed specifically for CRPS, it is not surprising that we found internal consistency was insufficient in pain-free controls. This can be explained by the lack of variability and reinforces the importance of validating measurement tools in condition-specific groups. Consistent with a previous study,[51] the r-B-CRPS-BPDS showed large positive relationships with the neurobehavioral questionnaire,[14] indicating good construct validity.

Another item with relatively low internal consistency involved the limb drawing based on participant description. Such finding may reflect researcher scoring subjectivity and/or a small possible range (0-2), resulting in reduced ability to capture variability. Because this item met our validity criteria and it directly assesses body representation distortions, we advise retaining it. The drawing itself might also provide qualitative insights not captured by the quantitative score. In future studies, more detailed instructions for making and scoring the

drawing could be beneficial. For example, use of standard participant instructions/prompts as they describe the mental image, a check-list of different experiences (e.g. feels distorted, missing), and/or having participants modify a digital limb avatar.[58] Body representation distortion is partly covered by the item exploring differences in how the affected limb looks compared to how it feels; this item was superior (even relative to the total score) at dissociating people with and without CRPS. Thus, this item alone may be useful to identify body representation distortion when time constrains are present, particularly as it allows comments and thus potentially useful qualitative insights.

Next, we evaluated which demographic or clinical variables are related to body perception disturbances as measured with the r-B-CRPS-BPDS. Across two analyses, the r-B-CRPS-BPDS showed positive relationships with pain intensity, fear of movement, and depression, consistent with previous studies.[32,36,51] In a third analysis we found a positive relation with upper limb disability (including sensations of pain). We are unable to comment on the relative importance of these factors, because we did not have sufficient data to include all variables in a single analysis. We did not evaluate whether body perception disturbances are specific for CRPS, they could relate to chronic pain in general. Indeed, previous studies have described body perception disturbance in other limb pain conditions, as measured with e.g. the neurobehavioral questionnaire[13,26,37,48] and limb laterality recognition task.[45] In our analysis the predictor variables explained 32% to 50% of the variances of r-B-CRPS-BPDS scores , leaving a large part of variance unexplained. This emphasizes the importance of separately assessing body perception disturbances, in addition to, for example, pain intensity. The relevance is furthermore stressed by evidence that greater body perception disturbances are associated with poorer outcome (e.g. higher pain intensity, disability, longer disease duration) in CRPS,[39,62] and other pain conditions such as knee osteoarthritis,[42,53] although this is not always found.[13,37]

We also looked at predictors of body perception disturbances over time. The only predictor of future r-B-CRPS-BPDS scores was the baseline score. There was no consistent change (increase or decrease) of r-B-CRPS-BPDS scores over time. This finding should be interpreted with caution, since our dataset was retrospective and thus recruited a convenience sample of people with CRPS of varying duration, rather than a prospective sample. Despite these limitations, our results show that in people with long-standing CRPS (mean duration = 5.6 years), there is no evidence for improvement or worsening of body perception disturbances over time. Evaluation of an inception cohort, where people with CRPS are enrolled at the time of diagnosis (or within the first year of diagnosis) may be warranted to more fully explore the trajectory of body perception disturbances over time.

The r-B-CRPS-BPDS can be complemented with more objective measures of body perception disturbances, such as the sense of limb position, limb movement, and limb size;[4,30,39] and localizing or detecting tactile stimuli.[7,12,20,56] A more implicit measure is that of limb laterality recognition, thought to rely on intact body representation.[24,40,41,46,47] Relationships between such measures and the subjective reports of body perception disturbances as measured with the B-CRPS-BPDS have been found in some studies,[32,36] but not in others.[4] In the evaluation of body perception disturbances it is therefore important to use both subjective and objective measures. Furthermore, the r-B-CRPS-BPDS is a quick and easy-to-administer tool in the clinical context or as an outcome for assessing the effectiveness of treatments that target body representation alterations. For example, in two studies measuring the effect of a pain management program or mirror therapy, the B BPDS CRPS scores decreased from before to after treatment, along with pain reduction.[27,28]

Limitations

This study has several limitations. First, one of the variables in our analysis was the disease duration, based on the moment of insult, injury, or symptom onset. It is likely that, for at least a subset of patients, CRPS developed after this moment and therefore does not strictly reflect the CRPS duration. Typically, however, there is a delay between symptom onset and receiving a diagnosis of CRPS.[8,33] Therefore, the onset of symptoms might be a more accurate estimate of the start of CRPS than the moment of receiving the diagnosis. Second, participants retrospectively reported the time of their symptom onset. This was based on their memory and, therefore, prone to error. Third, our longitudinal analysis was retrospective and based on convenience sampling from numerous studies (versus prospective consecutive, random, or population sampling recruitment methods). In addition, the number of measurements and the measurement intervals differed between participants which could have added noise to the longitudinal analysis. Fourth, aside from participants involved in our own RCT,[19] whether CRPS participants received treatments between research sessions was not recorded. It is possible that significant changes over time were experienced by subsets of participants (e.g. those who underwent rehabilitation between sessions), but were undetected. Fifth, because this was a retrospective study that combined numerous datasets, some measures that may relate to body perception disturbances, such as anxiety,[51] could not be included. Future work exploring body perception disturbances and psychological wellbeing/distress appears warranted. Sixth, this study involved relatively few people with lower limb CRPS, and in general their clinical profiles were not as well characterized as those with upper limb CRPS (e.g. lower limb motor

impairment or disability was not measured). Finally, no dissociation was made between CRPS I and II since not all the participants knew their type and of those who knew, only a small number presented with CRPS II.

Conclusion

We propose a revised B-CRPS-BPDS, excluding item 3 on attention. The r-B-CRPS-BPDS has good internal consistency in people with CRPS, and dissociates between people with CRPS and pain-free controls. Higher scores on the r-B-CRPS-BPDS relate to higher scores on the neurobehavioral questionnaire, indicating good construct validity. The r-B-CRPS-BPDS showed positive relationships with pain intensity, fear of movement, depression, and upper limb disability. Finally, in people with CRPS, there was no consistent change (increase or decrease) of r-B-CRPS-BPDS scores over time. Body perception disturbances may be important for the development and maintenance of CRPS; yet its relationships to pain, other symptoms, and treatment effects remain unclear. Validly measuring body perception disturbances in CRPS, and potentially other forms of chronic pain, is important for investigating these potential relationships and could be helpful for gaining a fuller picture of individual patient experiences.

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References

1. Bean DJ, Johnson MH, Heiss-Dunlop W, Lee AC, Kydd RR: Do psychological factors influence recovery from complex regional pain syndrome type 1? A prospective study. Pain 156:2310–8, 2015.
2. Beaton DE, Katz JN, Fossel AH, Wright JG, Tarasuk V, Bombardier C: Measuring the whole or the parts? Validity, reliability, and responsiveness of the disabilities of the arm, shoulder and hand outcome measure in different regions of the upper extremity. J Hand Ther Hanley & Belfus, Inc.; 14:128–42, 2001.
3. Bourgeois A, Leunes A, Meyers M: Full-scale and short-form of the Profile of Mood States: A factor analytic comparison. J Sport Behav 33:355–76, 2010.
4. Brun C, Giorgi N, Pinard A-M, Gagné M, McCabe CS, Mercier C: Exploring the Relationships Between Altered Body Perception, Limb Position Sense, and Limb Movement Sense in Complex Regional Pain Syndrome. J Pain 20:17–27, 2019.
5. Bultitude JH, Petrini K: Altered visuomotor integration in complex regional pain syndrome. Behav Brain Res Elsevier B.V.; 397:112922, 2021.
6. Bultitude JH, Walker I, Spence C: Space-based bias of covert visual attention in complex regional pain syndrome. Brain 140:2306–21, 2017.
7. Catley MJ, O'Connell NE, Berryman C, Ayhan FF, Moseley GL: Is Tactile Acuity Altered in People With Chronic Pain? A Systematic Review and Meta-analysis. J Pain Elsevier Ltd; 15:985–1000, 2014.
8. Choi YS, Lee MG, Lee HM, Lee CJ, Jo JY, Jeon SY, Lee SC, Kim YC: Epidemiology of complex regional pain syndrome: a retrospective chart review of 150 Korean patients. J Korean Med Sci 23:772–5, 2008.
9. Crombez G, Vlaeyen JWS, Heuts PHTG, Lysens R: Pain-related fear is more disabling than pain itself: evidence on the role of pain-related fear in chronic back pain disability. Pain 80:329–39, 1999.
10. Dancey C, Reidy J: Statistics without Maths for Psychology: using SPSS for Windows. London: Prentice Hall; 2004.
11. Field A: Discovering statistics using IBM SPSS statistics. 4th ed. London, UK: Sage Publications Ltd; 2013.
12. Förderreuther S, Sailer U, Straube A: Impaired self-perception of the hand in complex regional pain syndrome (CRPS). Pain 110:756–61, 2004.

13. Frettlöh J, Hüppe M, Maier C: Severity and specificity of neglect-like symptoms in patients with complex regional pain syndrome (CRPS) compared to chronic limb pain of other origins. Pain 124:184–9, 2006.
14. Galer BS, Jensen M: Neglect-Like Symptoms in Complex Regional Pain Syndrome. J Pain Symptom Manage 18:213–7, 1999.
15. Galer BS, Jensen M, Butler S: Neglect-like signs and symptoms in CRPS. Pain International Association for the Study of Pain; 154:961–2, 2013.
16. Goedert KM, Boston RC, Barrett AM: Advancing the Science of Spatial Neglect Rehabilitation: An Improved Statistical Approach with Mixed Linear Modeling. Front Hum Neurosci 7:211, 2013.
17. Gummesson C, Atroshi I, Ekdahl C: The disabilities of the arm, shoulder and hand (DASH) outcome questionnaire: longitudinal construct validity and measuring self-rated health change after surgery. BMC Musculoskelet Disord 4:11, 2003.
18. Halicka M, Vittersø AD, McCullough H, Goebel A, Heelas L, Proulx MJ, Bultitude JH: Disputing space-based biases in unilateral complex regional pain syndrome. Cortex Elsevier Ltd; 127:248–68, 2020.
19. Halicka M, Vittersø AD, McCullough H, Goebel A, Heelas L, Proulx MJ, Bultitude JH: Prism adaptation treatment for upper-limb complex regional pain syndrome: a double-blind randomized controlled trial. Pain 162:471–89, 2021.
20. Halicka M, Vittersø AD, Proulx MJ, Bultitude JH: Neuropsychological Changes in Complex Regional Pain Syndrome (CRPS). Behav Neurol 2020:1–30, 2020.
21. Harden R, Bruehl S, Perez RSGM, Birklein F, Marinus J, Maihofner C, Lubenow T, Buvanendran A, Mackey S, Graciosa J, Mogilevski M, Ramsden C, Chont M, Vatine J-J: Validation of proposed diagnostic criteria (the "Budapest Criteria") for Complex Regional Pain Syndrome. Pain 150:268–74, 2010.
22. Harden R, Maihofner C, Abousaad E, Vatine J, Kirsling A, Perez RSGM, Kuroda M, Brunner F, Stanton-Hicks M, Marinus J, van Hilten JJ, Mackey S, Birklein F, Schlereth T, Mailis-Gagnon A, Graciosa J, Connoly SB, Dayanim D, Massey M, Frank H, Livshitz A, Bruehl S: A prospective, multisite, international validation of the Complex Regional Pain Syndrome Severity Score. Pain 158:1430–6, 2017.
23. IBM Corp.: IBM SPSS Statistics for Windows, Version 27.0. Armonk, NY: IBM Corp.; 2020.
24. Jones HG, Braithwaite FA, Edwards LM, Causby RS, Conson M, Stanton TR: The effect of handedness on mental rotation of hands: a systematic review and meta-

analysis. Psychol Res Springer Berlin Heidelberg; , 2021.
25. Karcioglu O, Topacoglu H, Dikme O, Dikme O: A systematic review of the pain scales in adults: Which to use? Am J Emerg Med Elsevier Inc.; 36:707–14, 2018.
26. Kolb L, Lang C, Seifert F, Maihöfner C: Cognitive correlates of "neglect-like syndrome" in patients with complex regional pain syndrome. Pain International Association for the Study of Pain; 153:1063–73, 2012.
27. Kotiuk V, Burianov O, Kostrub O, Khimion L, Zasadnyuk I: The impact of mirror therapy on body schema perception in patients with complex regional pain syndrome after distal radius fractures. Br J Pain 13:35–42, 2019.
28. Lewis JS, Kellett S, McCullough R, Tapper A, Tyler C, Viner M, Palmer S: Body Perception Disturbance and Pain Reduction in Longstanding Complex Regional Pain Syndrome Following a Multidisciplinary Rehabilitation Program. Pain Med 20:2213–9, 2019.
29. Lewis JS, Kersten P, McCabe CS, McPherson KM, Blake DR: Body perception disturbance: A contribution to pain in complex regional pain syndrome (CRPS). Pain 133:111–9, 2007.
30. Lewis JS, Kersten P, McPherson KM, Taylor GJ, Harris N, McCabe CS, Blake DR: Wherever is my arm? Impaired upper limb position accuracy in Complex Regional Pain Syndrome. Pain International Association for the Study of Pain; 149:463–9, 2010.
31. Lewis JS, McCabe CS: Body perception disturbance (BPD) in CRPS. Pract Pain Manag :60–6, 2010.
32. Lewis JS, Schweinhardt P: Perceptions of the painful body: The relationship between body perception disturbance, pain and tactile discrimination in complex regional pain syndrome. Eur J Pain 16:1320–30, 2012.
33. Lunden LK, Kleggetveit IP, Jørum E: Delayed diagnosis and worsening of pain following orthopedic surgery in patients with complex regional pain syndrome (CRPS). Scand J Pain 11:27–33, 2016.
34. Maihafer GC, Llewellyn MA, Pillar WJ, Scott KL, Marino DM, Bond RM: A comparison of the figure-of-eight method and water volumetry in measurement of hand and wrist size. J Hand Ther 16:305–10, 2003.
35. McNair D, Lorr M, Droppleman L: Manual for the Profile of Mood States. Educ. Ind. Test. Serv. San Diego: Educational and Industrial Testing Service; 1971.
36. Mibu A, Nishigami T, Uematsu H, Tanaka K, Shibata M, Matsuda Y, Fujino Y: Validation of the Japanese version of the Bath CRPS Body Perception Disturbance

Scale for CRPS. J Anesth Springer Singapore; :1–7, 2020.
37. Michal M, Adler J, Reiner I, Wermke A, Ackermann T, Schlereth T, Birklein F: Association of Neglect-Like Symptoms with Anxiety, Somatization, and Depersonalization in Complex Regional Pain Syndrome. Pain Med :pnw214, 2016.
38. Miller RP, Kori SH, Todd DD: The Tampa Scale: a Measure of Kinisophobia. Clin J Pain 7:51, 1991.
39. Moseley GL: Distorted body image in complex regional pain syndrome. Neurology 65:773, 2005.
40. Moseley GL: Why do people with complex regional pain syndrome take longer to recognize their affected hand? Neurology 62:2182–6, 2004.
41. Moseley GL, Sim DF, Henry ML, Souvlis T: Experimental hand pain delays recognition of the contralateral hand—Evidence that acute and chronic pain have opposite effects on information processing? Cogn Brain Res 25:188–94, 2005.
42. Nishigami T, Mibu A, Tanaka K, Yamashita Y, Yamada E, Wand BM, Catley MJ, Stanton TR, Moseley GL: Development and psychometric properties of knee-specific body-perception questionnaire in people with knee osteoarthritis: The Fremantle Knee Awareness Questionnaire. Sumitani M, editor. PLoS One 12:e0179225, 2017.
43. Pellecchia GL: Figure-of-eight method of measuring hand size: J Hand Ther 16:300–4, 2003.
44. Petersen EJ, Irish SM, Lyons CL, Miklaski SF, Bryan JM, Henderson NE, Masullo LN: Reliability of Water Volumetry and the Figure of Eight Method on Subjects With Ankle Joint Swelling. J Orthop Sport Phys Ther 29:609–15, 1999.
45. Ravat S, Olivier B, Gillion N, Lewis F: Laterality judgment performance between people with chronic pain and pain-free individuals. A systematic review and meta-analysis. Physiother Theory Pract Taylor & Francis; 00:1–21, 2019.
46. Reid E, Wallwork SB, Harvie D, Chalmers KJ, Gallace A, Spence C, Moseley GL: A New Kind of Spatial Inattention Associated with Chronic Limb Pain? Ann Neurol 79:701–4, 2016.
47. Reinersmann A, Haarmeyer GS, Blankenburg M, Frettlöh J, Krumova EK, Ocklenburg S, Maier C: Left is where the L is right. Significantly delayed reaction time in limb laterality recognition in both CRPS and phantom limb pain patients. Neurosci Lett Elsevier Ireland Ltd; 486:240–5, 2010.
48. Reinersmann A, Landwehrt J, Krumova EK, Ocklenburg S, Güntürkün O, Maier C: Impaired spatial body representation in complex regional pain syndrome type 1 (CRPS

I). Pain International Association for the Study of Pain; 153:2174–81, 2012.

49. Rodriguez CS: Pain measurement in the elderly: A review. Pain Manag Nurs 2:38–46, 2001.
50. Rossi V, Pourtois G: Transient state-dependent fluctuations in anxiety measured using STAI, POMS, PANAS or VAS: a comparative review. Anxiety, Stress Coping 25:603–45, 2012.
51. Schulte-Goecking H, Azqueta-Gavaldon M, Storz C, Woiczinski M, Fraenkel P, Leukert J, Azad SC, Kraft E: Psychological, social and biological correlates of body perception disturbance in complex regional pain syndrome. Curr Psychol Current Psychology; , 2020.
52. Swinkels-Meewisse EJC., Swinkels RAH., Verbeek AL., Vlaeyen JW., Oostendorp RA.: Psychometric properties of the Tampa Scale for kinesiophobia and the fear-avoidance beliefs questionnaire in acute low back pain. Man Ther 8:29–36, 2003.
53. Tanaka S, Nishigami T, Wand BM, Stanton TR, Mibu A, Tokunaga M, Yoshimoto T, Ushida T: Identifying participants with knee osteoarthritis likely to benefit from physical therapy education and exercise: A hypothesis-generating study. Eur J Pain 25:485–96, 2021.
54. Tatro-Adams D, McGann SF, Carbone W: Reliability of the Figure-of-Eight Method of Ankle Measurement. J Orthop Sport Phys Ther 22:161–3, 1995.
55. Ten Brink AF, Halicka M, Vittersø AD, Keogh E, Bultitude JH: Ignoring space around a painful limb? No evidence for a body-related visuospatial attention bias in complex regional pain syndrome. Cortex Elsevier Ltd; 136:89–108, 2021.
56. Trojan J, Speck V, Kleinböhl D, Benrath J, Flor H, Maihöfner C: Altered tactile localization and spatiotemporal integration in complex regional pain syndrome patients. Eur J Pain 23:472–82, 2019.
57. Tschopp M, Swanenburg J, Wertli MW, Langenfeld A, McCabe CS, Lewis J, Baertschi E, Brunner F: Die deutsche Version der Bath Body Perception Disturbance Scale (BBPDS-D). Der Schmerz 32:195–200, 2018.
58. Turton AJ, Palmer M, Grieve S, Moss TP, Lewis J, McCabe CS: Evaluation of a Prototype Tool for Communicating Body Perception Disturbances in Complex Regional Pain Syndrome. Front Hum Neurosci 7:1–8, 2013.
59. Vittersø AD, Buckingham G, Ten Brink AF, Halicka M, Proulx M, Bultitude JH: No evidence of impaired sensorimotor adaptation in Complex Regional Pain Syndrome. bioRxiv Prepr :1–58, 2020.

60. Vittersø AD, Buckingham G, Halicka M, Proulx MJ, Bultitude JH: Altered updating of bodily and spatial representations after tool-use in complex regional pain syndrome. Pain 161:1609–28, 2020.
61. Vlaeyen JWS, Kole-Snijders AMJ, Boeren RGB, van Eek H: Fear of movement/(re)injury in chronic low back pain and its relation to behavioral performance. Pain 62:363–72, 1995.
62. Wittayer M, Dimova V, Birklein F, Schlereth T: Correlates and importance of neglect-like symptoms in complex regional pain syndrome. Pain 159:978–86, 2018.

Revised Bath CRPS Body Perception Disturbance Scale

The following questions are about your perception of your affected body part in relation to the rest of your body.

1) On a scale of 0-10 how much a part of your body does the affected part feel?

Very much a part = 0__1__2__3__4__5__6__7__8__9__10 = Completely detached

2) On a scale of 0-10 how aware are you of the physical position of your limb?

Very aware = 0__1__2__3__4__5__6__7__8__9__10 = Completely unaware

3) On a scale of 0-10 how strong are the emotional feelings that you have about your limb?

Strongly positive = 0__1__2__3__4__5__6__7__8__9__10 = Strongly negative

4) Is there a difference between how your affected limb looks or is on touch compared to how it feels in terms of the following:

Size yes ☐ no ☐ Comment_____

Temperature yes ☐ no ☐ Comment_____

Pressure yes ☐ no ☐ Comment_____

Weight yes ☐ no ☐ Comment_____

5a) Have you ever had a desire to amputate the limb? Yes ☐ No ☐

5b) If yes, how strong is that desire now?

Not at all = 0__1__2__3__4__5__6__7__8__9__10 = Very strong

Desired amputation site_____

6) With eyes closed describe a mental image of your affected and unaffected body parts. The assessor will draw a picture which you than will verify. Alternatively, ask a friend or family member to draw your description for you if possible, otherwise draw the description yourself after one minute's consideration of the mental image.

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Once the picture is drawn, make sure it is an accurate account of your image of your affected body part.

Scoring (for the assessor): The sum of numerical ratings from items 1 to 3 and 5b are added to items 4 and 5a (scored no = 0, yes = 1). The mental representation drawing (item 6) is graded on a three-point scale; no distortion = 0, distortion = 1, severe distortion = 2. The total score of the r-CRPS BPDS ranges from 0 to 47, with higher scores indicating more body perception disturbances. This scale is similar to the scale of Lewis and McCabe (2010)[1], without the item on attention.

[1] Lewis JS, McCabe CS: Body perception disturbance (BPD) in CRPS. Pract Pain Manag :60–6, 2010.