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REVIEW ARTICLE

Assessment of body perception disturbances in complex regional pain syndrome: A systematic review using the COSMIN guideline

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Abstract

Objective: To conduct a systematic review to identify which tools are being used to assess body perception disturbances in Complex Regional Pain Syndrome (CRPS) and to provide an evidence-based recommendation in the selection of an assessment tool, based on measurement properties.

Databases and data treatment: Five electronic databases (EMBASE, Pubmed, PsycInfo, Science Direct and Web of Science) were searched for English or French written articles, with no time restrictions. All original articles using a body perception assessment tool with adult patients with CRPS were selected, regardless of their design (controlled trials, single case, qualitative study). Two investigators screened abstracts, selected full articles and extracted data independently.

Results: Thirty-eight full-text papers were obtained and three main methods to evaluate body perception disturbances were identified: The Bath Body Perception Disturbance Scale, the Neglect-like Symptoms questionnaire adapted from Galer and the patient's body perception description. No full psychometric assessments were found. The Limb Laterality Recognition Task was also used in conjunction with another method.

Conclusions: Three main assessment methods for CRPS body perception disturbances are currently used. Full psychometric evaluation has not been completed for any of the assessment methods. As a consequence, we could not fully apply the COSMIN guideline. To date, there is no agreement concerning the use of a specific questionnaire or scale. The results indicate a need for further research such as psychometric properties of these questionnaires.

Significance: This systematic review identified body perception disturbances assessment methods and their the psychometric properties in order to provide help and guidance to researchers and clinicians to investigate those clinical features.

1 | INTRODUCTION

Complex Regional Pain Syndrome (CRPS) is a chronic pain condition characterized by spontaneous and evoked pain disproportionate in magnitude or duration to the initial lesions that usually occur in a distal extremity (Bruehl, 2015). It displays a combination of pain, vascular, trophic, autonomic and motor abnormalities as well as sensory disorders such as altered body perceptions (Lotze & Moseley, 2007).

Body perception can be defined as 'the process of how we perceive our bodies' (Lewis & Schweinhardt, 2012). It involves complex interactions between proprioceptive, vestibular, somatosensory and visual inputs interrelating with motor systems(Schwoebel et al., 2001).

Body perception disturbances in CRPS are defined as 'the individual's perceived alteration of their CRPS affected body part while regarding the remainder of their body as normal' (Lewis & McCabe, 2010). Patients with CRPS report that they pay little attention to their affected limb, describing it as it was not part of their body (Lewis et al., 2007). They tend to perceive that it is distorted in shape or size (Lewis & Schweinhardt, 2012), usually larger than it actually is (Moseley, 2005). They typically report difficulties recognizing their affected limb (Moseley Moseley, 2004a, Moseley, 2004b, Moseley, 2004c) and are not aware of its position or orientation (Moseley, 2005; Peltz et al., 2011).

First described by Galer et al. (Galer et al., 1995), the term 'neglect-like' symptoms were previously attached to such clinical observations, referring to its striking similarity with post-stroke neglect. Gradually, the term 'neglect-like' was replaced by 'body perception disturbances', to clearly differentiate it from neglect after stroke (Forderreuther et al., 2004; Lewis et al., 2007; Michal et al., 2017).

BPD in CRPS has been reported in many studies (Lewis et al., 2007; Moseley, 2005; Peltz et al., 2011), together with their potential role in persistent pain phenomena. They have been increasingly acknowledged as a feature of CRPS, as the majority of patients with CRPS report disturbances in body perception (ranging from 54% (Forderreuther et al., 2004) to 72% (Michal et al., 2017) and 84% for the presence of at least one symptom (Galer & Jensen, 1999)). Several studies have confirmed that body perception disturbances are more prevalent and more severe in CRPS (Frettlöh et al., 2006; Michal et al., 2017; Wittayer et al., 2018) compared with other chronic limb pain.

A body of evidence provided by brain imaging studies suggested that cortical reorganization occurs in CRPS, in particular, in areas involved in sensorimotor functions, pain perception and body schema (Di Pietro et al., 2013; Lewis & McCabe, 2010; Maihofner et al., 2003; Pleger et al., 2014). This evidence suggests a close relationship between disrupted cortical limb representation and perception disturbances of the affected limb (Moseley, 2005; Peltz et al., 2011). These changes seem to be related to pain and correlate with body image, but there is no evidence that they cause or are caused by pain (Bruehl, 2015). Schwoebel et al. (2001) were the first to indicate a link between the degree of body schema disturbance and the intensity of pain and those results were confirmed in other studies (Lewis & Schweinhardt, 2012). They interpreted their findings as a disruption in body representation. The specificity of these symptoms for CRPS remains uncertain. However, while reorganization of the somatosensory cortex often appears to be associated with persistent CRPS, these results were recently questioned. FMRi studies did not confirm these findings (Di Pietro et al., 2015; Mancini et al., 2019), compelling us to review the mechanism hypotheses.

BPD is thought to 'interfere with the ability to process information coming from the limb and the space around it' (Bultitude et al., 2017). Research exploring sensory modalities and changes in perception involving motor impairments (Reid et al., 2018), somatospatial inattention (Reid et al., 2016) confined to bodily representation and sensorimotor disturbances with impaired self-localisation (Bellan et al., 2021) have flourished in the last years, allowing a better understanding of CRPS and BPD. As an example, Bultitude et al. (2017) pointed out that general attention bias was predicted by BPD scores. Illusions are another way to explore body awareness and its underlying processes, such as the Disappearing Hand Trick (Bellan et al., 2021), the rubber hand illusion (Reinersmann et al., 2013) or a visuotactile illusion which only creates pain in patients with CRPS (dysinchiria) (Acerra & Lorimer Moseley, 2005; Krämer et al., 2008).

The results are still conflicting: some studies support the hypotheses of neglect similar to neglect after stroke while others do not (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021; Halicka et al., 2020; Kolb et al., 2012; Reid et al., 2016; Wittayer et al., 2018).

Despite those observations, the mechanisms underlying body perception disturbances and their implications on CRPS development and severity are not fully understood.

Body perception disturbances are acknowledged as an important clinical feature in patients with CPRS. In clinical practice, patients tended to keep to themselves those perceptions and feelings, out of fear of being considered mentally ill (Galer & Jensen, 1999; Lewis & McCabe, 2010). Since then, awareness has risen and clinicians have increasingly been encouraged to explore body perception disturbances among the signs and symptoms of CRPS and

to inform patients about them, as in the recent European Pain Federation task force standards (Goebel et al., 2019). The standards state that patients 'must receive adequate information soon after diagnosis on [...] (iv) signs and symptoms, including body perception abnormalities' (Goebel et al., 2019). Gillespie et al. (2016) in an audit aiming at identifying factors for the development of CRPS after distal radius fracture, recommend 'picking up verbal and visual clues from the patient earlier, [...] such as neglect of limb'.

Several authors now recommend considering body perception disturbances in the diagnostic (Forderreuther et al., 2004; Osumi et al., 2015) or as a rehabilitation target (Boesch et al., 2016; Moseley Moseley, 2004a, Moseley, 2004b, Moseley, 2004c).

Questionnaires are used to assess BPD in CRPS, but there is a lack of a comprehensive summary of available assessment methods and critical appraisal including psychometric properties.

Thus, this systematic review was designed to identify body perception disturbances assessment tools used for CRPS and summarize their measurement properties with potential gaps in knowledge. It aims to provide an evidence-based recommendation to select an assessment method to be used by clinicians and researchers.

2 | LITERATURE SEARCH METHODS

The study protocol was registered on PROPERO (International Prospective Register of Systematic review, CRD42018089652). The review was reported in accordance with the PRISMA statement (Moher et al., 2015) (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) (see checklist attached in additional file). The COSMIN (Consensus-based Standards for the selection of health Measurement Instruments) guideline for a systematic review of patient-reported outcome measures (PROM) (Mokkink et al., 2016; Prinsen et al., 2016) was used to assess measurement properties, report findings and draw a conclusion.

2.1 | Data sources and searches

In order to identify all body perception disturbances assessment tools, a systematic search was performed in PubMed, EMBASE, Science Direct, PsycInfo and Web of Science (no limitation for publication period, up to November 2020). References were hand-searched for all relevant articles to identify studies that could have been missed by the initial search. The Cochrane Database of Systematic Reviews and PROSPERO were checked for other systematic reviews.

All search terms are listed in Box 1.

2.2 | Study selection

Studies were eligible for inclusion if they were available in English or French if they recruited adult participants with CRPS type I or II (Budapest criteria or alternate diagnostic criteria before 2000, no restrictions regarding disease duration), and if they assessed body perception disturbances.

No restrictions on experimental study design were applied, as the intention was to capture all body perception disturbances measures used for CRPS.

2.2.1 | Identifying outcome measures: a definition of body perception disturbances

Body perception disturbances referred to sensory disturbances that are reported regarding the affected limb such as the feeling of foreignness, strangeness, distortion

BOX 1 Search terms and Pubmed equation

Complex regional pain syndrome	Complex regional pain syndrome* Regional pain syndrome* Reflex sympathetic dystrophy Algodystrophy Causalgia Sudeck* atrophy Sudeck* Shoulder hand syndrome* Post-traumatic pain syndrome Algoneurodystrophy Reflex neurovascular dystrophy Post-traumatic angiospasm CRPS CRPS type 1 CRPS type 2 CRPS type I CRPS type II CRPS 1 CRPS 2 CRPS I CRPS II
Body perception disturbances	Body perception disturbances Somatosensory disorders Body image Neglect-like syndrome Perceptual disorders

Note: ((‘Complex regional Pain Syndromes’[MesH]) AND (‘Somatosensory Disorders’[MesH] OR ‘Perceptual Disorders’[MesH] OR ‘Body Image’[MesH] OR ‘neglect like syndrome’)).

or differences in shape, size, length, heaviness, etc. They can also be directly mentioned as neglect-like symptoms or body perception disturbances.

2.3 | Study inclusion

Two investigators independently screened the titles and abstracts from the search results and included studies meeting the inclusion criteria. When disagreement was not resolved through discussion, a third investigator was involved to reach a consensus. The same process was then used for full-text selection.

2.4 | Data extraction

Data extraction forms were used to extract the following data: author, date, study design, demographics of participants (age, gender), CRPS diagnostic criteria, CRPS duration, objectives of the study, type of intervention and type of BPD assessment.

2.5 | Identifying and assessing of quality of the PROM(s)

The 10-step procedure for conducting a systematic review of PROM was followed (Prinsen et al., 2016).

Studies were selected for this part after full-text reading. Studies reporting the development of a PROM, the evaluation of one or more measurement properties or of the interpretability of the PROM were included. A hand search was performed to identify missing studies, without any language restriction.

Two authors independently assessed the selected studies through the COSMIN Risk of Bias checklist, using available spreadsheets. Based on the COSMIN taxonomy, the measurement properties were divided into three domains: reliability, validity and responsiveness. Then, the results of each study were summarized, and the quality of the evidence was graded using the GRADE approach.

3 | RESULTS

3.1 | Identifying assessment methods: Study selection and characteristics

A total of 863 studies were identified by the databases search. After the selection process, 38 studies were included in the final overview. See Figure 1 for details (flow chart).

As expected, the selected studies incorporated a broad range of study designs, including cohorts, case–control studies, case reports, qualitative studies, cross-sectional studies, observational studies, mixed method studies and translation/cross-cultural adaptation studies. See Appendix S1 for a list of the assessment methods identified in selected studies and Appendix S2 for the complete list of included studies.

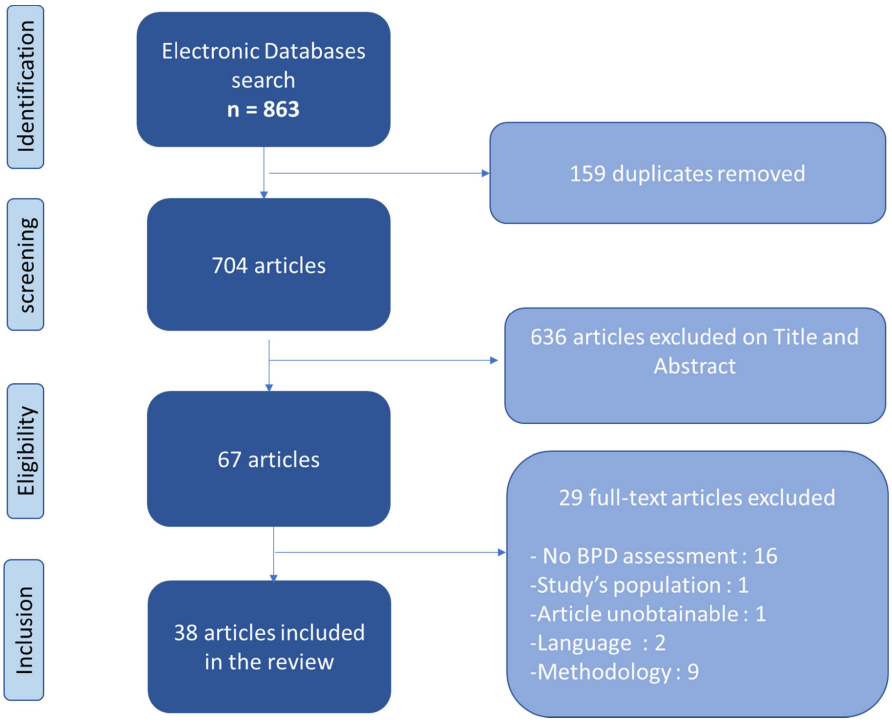

FIGURE 1 Flow chart.

For the assessment of the PROM quality, five studies were included (Brink, Antonia, & Bultitude, 2021; Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021; Lewis & Schweinhardt, 2012; Mibu et al., 2020; Tschopp et al., 2018).

3.2 | Characteristics of body perception disturbances assessment tools

The search identified nine different methods to assess BPD, used as a stand-alone or in the association. Three tools were specifically designed to assess BPD in CRPS: the Neglect-Like Symptoms Questionnaire (NLSQ) by Galer et al. (Galer & Jensen, 1999) (found in three articles), its adaptation by Frettlöh et al. (2006) (in 12 articles) and the Bath Body Perception Disturbance Scale (Bath BPD-S) (in 14 articles).

Other methods were used, such as the patient's body perception verbal description in 11 articles (including the use of mental imagery in four articles), or drawings in two articles. The Limb Laterality Recognition Task (LLRT) was used in complement to another method in eight articles. A few studies used a subjective visual midline bisection (four articles), a hand size self-evaluation (two articles) and a somatosensory task (finger identification or stimuli localization in five articles).

The majority of these assessment methods are Patient Reported Outcome Measures (PROM). The performance-based outcome measures, such as the LLRT, are systematically used in association with a PROM or a qualitative assessment (Table 1).

3.3 | The neglect-like symptoms questionnaire or Neurobehavioral questionnaire

(Galer & Jensen, 1999) This questionnaire was developed based on Galer's personal clinical experience with CRPS-I patients including previous research (Galer et al., 1995). It was then part of a patient's survey mailed to patients with CRPS in order to determine the frequency of neglect symptoms among them (242 patients, 10% response rate).

The questionnaire comprises five statements. Patients are asked to check each true statement about their affected limb. Two statements assess the presence of motor neglect symptoms: 'If I don't focus my attention on my painful limb, it would lie still, like dead weight' and 'I need to focus all of my attention on my painful limb to make it move the way I want it to'. Two other statements assess the presence of cognitive neglect: 'My painful limb feels as though it is not part of my body' and 'My painful limb feels dead to me'. A fifth statement assesses the presence of involuntary movements: 'My painful limb sometimes moves involuntarily, without my control'. The expected answer is dichotomous (true or false). The completion is quick and does not require extra material.

It recently has been explored in a study comparing neglect-like symptoms between people with CRPS and other chronic limb pain (online survey). Two components were identified within the questionnaire: motor and cognitive neglect-like symptoms (items 1-2-3-4) and involuntary movements (item 4) (Brink, Antonia, & Bultitude, 2021). The internal consistency of the motor and cognitive neglect-like symptoms component was found acceptable (Cronbach's alpha 0.76) (see Table 2).

3.3.1 | The neglect-like symptoms questionnaire adapted by Frettlöh et al.

Frettlöh et al. (2006) translated into German the five-neglect-like items of the Neglect-like Symptoms questionnaire by Galer and Jensen (1999) and expanded the dichotomous options to a 6-point Likert-scale (1 = never, 6 = always), to perform a quantitative assessment of the reported symptoms. The neglect-like total score is obtained by adding the five items and calculating arithmetic mean.

The authors recommend the use of the mean neglect-like score ≥5 as an additional criterion to confirm the diagnosis of CRPS (sensitivity 21.1%, specificity 90.6%).

3.3.2 | The bath CRPS body perception disturbance scale

(Lewis & McCabe, 2010) The Bath CRPS BPD Scale aims at providing a comprehensive assessment of BPD disturbances. It was developed through previous research and a series of semi-structured interviews (Lewis et al., 2007), articulated around six themes: hostile feelings, the spectrum of dissociation, the disparity between what is apparent and what is felt, the distorted mental image of the affected part, awareness of limb position and conscious attention.

This 7-item measure covers the following aspects: a sense of ownership (1); limb position awareness (2); attention paid to the limb (3); emotional feelings towards the limb (4); perceptual disparities in size, temperature, pressure and weight (compared with the unaffected limb) (5); desire to amputate the limb (6a and 6b) and mental representation of the limb (7). Items 1–4 and 6b rate individual aspects of BPD on a 0–10 scale. In item 7, the clinician asks the patient to visualize a mental picture of their

TABLE 1 Summary of the measurement properties

	Language	Population characteristics (age, gender)	Number of questions	Psychometric evaluation	Diagnostic tool/ cut-off	Ability to detect change/ MCID	Internal consistency	Reliability
The Neglect-Like Symptoms questionnaire or Neurobehavioral questionnaire (Galer & Jensen, 1999)	English	242 CRPS I	5 (true/false)	No evaluation	—	—	—	—
The Neglect-Like Questionnaire adapted by Frettlöh et al. (2006)	German	123 CPRS 117 chronic limb pain	5 (6 pt response scale)	Yes	score ≥5 (low sensitivity, but specificity 90.6%)	—	Cronbach's alpha = 0.86	—
Neurobehavioral questionnaire (Brink, Antonia, & Bultitude, 2021)	English	335 CRPS 407 other chronic pain	5 (true/false)	Yes	—	—	Cronbach's alpha = 0.76 (motor and cognitive component)	—
The Bath CRPS Body Perception Disturbance Scale (Bath CRPS BPD Scale) (Lewis & Schweinhardt, 2012, Brink, Antonia, Halicka, Vittersø, Keogh, and Bultitude (2021))	English	22 (15 W, 7 M; 50.6 yo) 22 healthy controls matched 114 (93 W; 46.06 years)/ healthy matched controls	6 + drawing (10 pt scale, true/false) *Revised version 5 + drawing*	Yes	Higher scores represent greater body perception disturbance	—	Cronbach's alpha 0.66–0.73	Cohen's kappa 0.87-

TABLE 2 Measurement properties of the included studies for the COSMIN checklist

		Language	CRPS population characteristics (gender, age [mean])	Internal consistency	Cross-cultural validity	Reliability	Remaining measurement properties	Concurrent validity
Bath CRPS body perception disturbance scale	Lewis and Schweinhardt (2012)	English	22 (15 W, 50.6 years)	Cronbach's alpha 0.66	—	Cohen's kappa 0.87	—	—
	Mibu et al. (2020)	Japanese	22 (14 W, 46 years)	Cronbach's alpha 0.73	+	—	—	TPD ratio ($r = 0.65$) TSK ($r = 0.51$)
	Tschopp et al. (2018)	German	56 (49 W; 50.9 years) (28 acute/28 stable)	Cronbach's alpha 0.92	+	ICC 0.85	SDC 10	EuroQol-5D ($\rho = 0.16$) NRS($\rho = -0.19$)
	Brink, Antonia, Halicka, Vittersø, Keogh, and Bultitude (2021)	English	114 (93 W; 46.06 years)	Cronbach's alpha 0.73 Cronbach's alpha 0.73[a]	—	—	—	Pain intensity[a] ($R^2 = 0.17$), TSK ($R^2 = 0.18$), Depression[a] ($R^2 = 0.33$), upper limb disability[a] ($r = 0.85$)
Neurobehavioral Questionnaire	Brink, Antonia, Halicka, Vittersø, Keogh, and Bultitude (2021)	English	335 (297 W; 46.64 years)	Cronbach's alpha 0.76 (Item 1-2–3-5)	—	—	—	—

Abbreviations: TPD, two-points discrimination threshold; TSK, Tampa Scale for Kinesiophobia; NRS, Numeric Rating Scale; +, sufficient (Prinsen et al., 2016).

[a]Revised version of the Bath BPD Scale (i.e. excluding item 3).

affecting limb with his eyes closed. The clinician draws a pencil line picture of both limbs as the patient describes his mental image. A textual description can be added. The drawing can be assessed and amended by the patient. The drawing is graded on a three-point scale (no distortion = 0, distortion = 1, severe distortion = 2). The rating 'severe distortion' is given if one or more parts of the limb are missing.

This scale is scored from 0 to 57, higher scores represent greater body perception disturbance. It takes longer to complete compared to the Neglect-Like Symptoms Questionnaire and requires the presence of a clinician for the drawing task.

In an early study, the scale was shown to have adequate internal consistency and inter-rater reliability (Cronbach's alpha 0.66, Cohen's kappa 0.87) (Lewis & Schweinhardt, 2012). A recent validation study (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021) proposed a revised version of the scale, excluding the item on attention (item 3). The internal consistency of the original and this revised version was found acceptable (Cronbach's alpha >0.7). They also explored known group validity, construct validity and associations with demographics and clinical characteristics. It was translated and adapted into Japanese and German (Mibu et al., 2020; Tschopp et al., 2018). All those studies found also acceptable internal consistency (Cronbach's alpha >0.7). Table 2 provides a summary of the measurement properties of these tools.

3.3.3 | Other methods

The LLRT is an implicit motor imagery task of recognizing the laterality of the image of a limb as either left or right (Parsons, 1987; Parsons et al., 1995; Schwoebel et al., 2001). Either a homemade software program, pictures or the Recognize® software program can be used. Pictures of limbs are randomly presented and subjects are asked to decide whether it is a left or a right limb. Reaction time (in milliseconds) and response accuracy are recorded.

Patients can be asked to describe their limb through direct questions (e.g. about limb ownership, 'how strong is your sense of ownership over your affected limb?' [Bean et al., 2015]). With their eyes closed, patients can be asked to visualize and describe how their limb appears (Bultitude & Rafal, 2010; McCabe et al., 2003; Osumi et al., 2015). The clinician can draw or write the kind of distortions that the patient describes (asymmetry in size, length, heaviness, colour, shape, missing parts, etc.). Similarly, a line drawing can be provided and the patient adds what he senses to the drawing (Hirakawa et al., 2020). Software and virtual avatars are also used (Turton et al., 2013). It is similar to item 7 of the Bath BPD-S but has been used on its own.

Patients can also report feelings towards the limb (hostile, foreignness, sense of ownership) by answering an open question or spontaneously (Bean et al., 2015; Forderreuther et al., 2004). The evaluator can also ask patients to evaluate their hand size (on a series of photographs) (Moseley, 2005; Tajadura-Jiménez et al., 2017), localize a tactile stimulus or identify which finger is being touched (Trojan et al., 2019). Those methods are considered by authors as part of body perception disturbances evaluation.

Although data are available for other populations, no psychometric evaluation was found for these methods for patients with CRPS.

3.4 | PROM selection, quality assessment and data synthesis (COSMIN)

No studies evaluating PROM development were included. The NLS was developed 'based on the senior author personal clinical experience with CRPS patients' (Galer & Jensen, 1999) and its development procedure or testing was not reported. Similarly, the Bath BPS-S was based on 'previous' qualitative research and has been 'developed clinically' (Lewis et al., 2007; Lewis & McCabe, 2010). Frettlöh et al. (2006) did not report on their translation method and the NLSQ was used as an outcome measurement. As a result, according to the COSMIN guideline, it was not included.

Four studies were selected, three of them concerning the Bath BPD scale, two of which being translations and cross-cultural validations (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021; Mibu et al., 2020; Tschopp et al., 2018). In the fourth study selected, Brink, Antonia, and Bultitude (2021) conducted an online survey in order to, among other things, identify components and assess the internal consistency of the Neurobehavioral Questionnaire.

The included articles investigated five measurement properties: internal consistency, reliability, measurement error, construct validity and cross-cultural validity. Responsiveness and structural validity still need to be completed. Studies ~~on the Bath BPD Scale and its translations~~ do not provide the multiple ratings that would have allowed us to compile evidence, as each version is considered separately (Prinsen et al., 2016). As a result, the level of evidence was rated as unknown.

Measurement properties of the included studies are summarized in Table 2 and the quality of studies according to the COSMIN Risk of Bias Checklist in Appendix S3.

4 | DISCUSSION

The scope of this review is to provide BPD assessment tools in CRPS to be used as outcome measures in clinical settings and research.

To do so, we conducted a systematic review as a way of establishing a full picture of the body perception disturbances assessment methods used in published CRPS intervention studies to date. We used the COSMIN guideline to include measurement properties evaluation. As it was our intention to capture this emerging domain of interest, we included studies with small patient numbers and case reports, because they can report on preliminary results or use of emerging new outcome measures.

Unsurprisingly, the vast majority are patient-reported outcome measures. The diversity of outcome measures identified across the studies demonstrates the challenge of synthesizing practices.

Three specific tools were described in the literature to explore body representation distortion in patients suffering from CRPS: the Bath CRPS BPD Scale, the Neglect-Like Symptom Questionnaire by Galer and its adaptation by Frettlöh. In addition to those questionnaires, the LLRT, somatosensory evaluations and verbal descriptions by patients (mental imagery, body scheme report etc.) were used. Interestingly, the LLRT and the NLS questionnaire are used as objective measures of body representation, while the Bath BPD Scale is considered subjective (Bultitude et al., 2017). The drawing item in the Bath CRPS BPD Scale, as a behavioural assessment, seems to be in a category of its own (Schulte-Goecking et al., 2020) and seems challenging to rate (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021).

Even if developing a questionnaire based on 'senior author experience with CRPS patients' (Galer & Jensen, 1999) or 'previous qualitative research' (Lewis et al., 2007) are quite common strategies, they were not reported. As a result, we could not complete the COSMIN standards for evaluation of the quality of PROM development, including items on PROM design, relevance, comprehensibility or comprehensiveness for any of the two original questionnaires. Frettlöh et al. translated the NLQ into German and expanded the dichotomous scale to a 6-point response scale. The translation method was not reported. The utility of this new tool in diagnostics has been investigated, but no full psychometric evaluation was performed.

Galer et al. did not conduct a psychometric study on the NLQ. Their survey identified the presence of neglect-like symptoms in a CRPS population (224 patients), with a 10% response rate.

The Bath BPD Scale recently provided more psychometric data (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021). It was also translated into German and Japanese. That two cross-cultural translation and adaptation provided new psychometrics and was proof of developing interest in the method.

Evidence gathered in this review was limited to the Bath BPD Scale. Several measurement properties were not available, such as content and structural validity, or poorly evaluated, such as reliability or responsiveness. Internal consistency (Cronbach's alpha ranging from 0.6 to 0.92) was evaluated in three studies. Evidence for validity (content and construct) was limited. The Bath BPD has been shown to have a very large relationship with the NLS questionnaire (Brink, Antonia, Halicka, Vittersø, Jones, et al., 2021), supporting the idea of a shared construct. Correlations with fear of movement (Tampa Scale for Kinesiophobia) were found, but correlations with pain intensity were not consistent (Brink, Antonia, Halicka, Vittersø, Jones, et al., 2021; Tschopp et al., 2018). As this body of evidence is incomplete, the quality of evidence was classified as unknown.

Recommendations on the most suitable assessment tool are based on the evaluation of the measurement properties, interpretability and feasibility aspects (Prinsen et al., 2016). With those aspects in mind, the Bath BPD Scale has the potential to be recommended, although further validation studies are needed. The NLS questionnaire has the advantage of being shorter and easier to administrate. Additional measurement properties studies are needed.

Of course, the structure (i.e. how the different items in a PROM are related among other definitions) of the Bath BPD Scale is a question that is still undergoing. Body perception disturbances as a construct are still under debate. Some authors focus on perceptual disturbances while others on 'motor neglect' or space. They refer to BPD as distorted body representation (Reinersmann et al., 2012), biased spatial attention (Reid et al., 2016; Torta et al., 2016), bodily representation, personal or peri-personal space and learned non-use (Punt et al., 2013).

The idea that all items in the questionnaire are manifestations of the same construct and thus correlated is not supported by strong evidence to date. Brink, Antonia, Halicka, Vittersø, Keogh, and Bultitude (2021) found that the item scoring attention (item 3) had insufficient corrected item-total correlation and pain-free controls scored higher. They computed a revised version of the questionnaire excluding this item (see TAB2 for psychometric evaluation). They proposed rewording the item or to create two separate items (hypervigilance and/or guarding and disregard). This new version may be proposed in the future, with full psychometric evaluation.

Evidently, body perception disturbances are investigated in other conditions. Interestingly, the Freemantle

Back Awareness Questionnaire (FreBAQ) was inspired by findings in CRPS. Based on Galer and Jensen (1999) (item 2, 3, 4) and a qualitative study in people with chronic low back pain (Wand et al., 2014), this questionnaire appears to be unidimensional with no redundant item (Wand et al., 2016). Extensive psychometric testing (Ehrenbrusthoff et al., 2018), multiple translations (German, Turkish, Japanese, Persian etc) and body parts adaptations (neck, knee etc.) are available. This shows the growing interest in the relationship between body perception and pain. The CRPS researchers' groundbreaking work may inspire other chronic pain fields in the future.

Some authors considered the limb laterality task as an objective measure of body schema (Bultitude et al., 2017; Di Pietro et al., 2013) as it is known to depend on an intact body schema (Bowering et al., 2013). Recognizing the laterality of a pictured limb involves confirming an initial decision by mentally moving the internal representation of one's own limb to match that of the picture (Parsons, 1987; Parsons et al., 1995). Previous chronometric findings suggested that performance on this task depends on the body schema (Schwoebel et al., 2001). Response times and accuracy are thought to reflect the degree of body schema disruption.

The evidence regarding patients with CRPS and response time in this task is conflicting. While several authors found a delayed recognition of laterality (Moseley Moseley, 2004a, Moseley, 2004b, Moseley, 2004c; Reinersmann et al., 2010; Schwoebel et al., 2001; Wittayer et al., 2018), others failed to reproduce this result (Breimhorst et al., 2018; Reinersmann et al., 2012). Although age, duration of CRPS and insufficiently powered investigation were possible factors, the type of pictures may be of some influence. It appears that reaction time might depend on the direction of orientation of the pictured limb and that it increases with awkwardness (or difficulty) of the stimulus orientation (Parsons, 1987; Schwoebel et al., 2001). Future research might want to explore such factors to be able to determine if the Limb Laterality Recognition Task can be used on its own as an objective measure of body representation with people with CRPS. In studies selected in this review, it has only been used associated with a questionnaire, according to some authors (Brink, Antonia, Halicka, Vittersø, Keogh, & Bultitude, 2021).

New therapeutic approaches have been developed to improve cortical function by means of brain training and are indicated for BPD treatment (Bellan et al., 2021; Lewis et al., 2019). These rehabilitation strategies involve mirror therapy (McCabe, 2011), graded motor imagery (GMI) (Moseley Moseley, 2004a, Moseley, 2004b, Moseley, 2004c), motor imaging, tactile discrimination training (Moseley et al., 2008) or prism adaptation (Sumitani et al., 2007). Meta-analyses suggest that GMI and mirror therapy show encouraging results on pain and disability compared with classic rehabilitation programs (Bowering et al., 2013; O'Connell et al., 2013). Several clinical trials have investigated BPD as an outcome (Lewis et al., 2019; Osumi et al., 2015), using the Bath BPD Scale and sensory-motor rehabilitation techniques.

4.1 | Recommendations for future research and clinicians

Future research is needed to evaluate the performance of those tools in the population of patients with CRPS. Prospective and longitudinal studies using all the tools detailed in this article on a population of patients suffering from CRPS and appropriate controls (for instance other conditions with neuropathic pain) would be helpful to compare their strengths, limitations, and search for correlations/redundancies between those tests/questionnaires. Questions about the influence of CRPS type I or II can be asked, as the majority of included subjects present CRPS-I. Similarly, lower limb presentations are underrepresented. A future COMPACT (Grieve et al., 2017) may be able to incorporate body perception disturbances if such studies were to be conducted. Feasibility should be considered through the process, including digital perspectives already initiated.

4.2 | Study limitations

This review has limitations. We tried to identify all studies using a body perception disturbances measurement method, but the possibility remains that we missed some. Only papers in English and French were reviewed. The full text of one of the articles was unobtainable, despite our attempts to source it online, in academic libraries or by contacting the author.

We could not apply the COSMIN methodology in full due to the lack of information or report. As a result, we could not provide strong recommendations. Because this subject arouses interest, this will probably change with an increasing number of studies.

5 | CONCLUSIONS

This systematic review identified a variety of tools including three main methods used to assess body perception disturbances: The Bath Body Perception Disturbances Scale, the Neglect-Like Symptoms Questionnaire, and the patient's verbal description of

their body. No complete psychometric properties were found for any of these tools. Clinicians can choose between specific questionnaires for CRPS or generic tests. The use of the COSMIN guidelines did not allow us to draw firm conclusions. However, the Bath BPD Scale has received more attention and provided more psychometric data. Its revised version might also provide a more precise understanding of the CRPS experience and should be considered to use. The rewording of the item on attention might be considered in the future. The Bath BPD scale also requires the collaboration of the clinician and patient for the drawing. Even though scoring might be uncertain, qualitative data such as exploring the influence of this clinician/patient interaction may provide insight into this unusual interaction.

Thus, there is a clear need for further research in order to provide the best assessment tools that can be used. BPD assessment may be useful either for a more accurate diagnosis or prognosis of CRPS, to monitor CRPS activity, or as a treatment option.

AUTHOR CONTRIBUTIONS

SA, JN and AD designed the study. SA and TO performed all searches and data collection. TR discussed discrepancies. All authors discussed the results. SA drafted the manuscript and all authors revised it.

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CONFLICT OF INTEREST

None.

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SUPPORTING INFORMATION

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