

Explicit and implicit experience of own's body in painful musculoskeletal disorders and rheumatic diseases: A scoping review protocol of available quantitative and qualitative evidence

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Abstract

Introduction: Our body experience is organized at twofold levels: perceptual and cognitive-emotional. These higher-order processes are clearly different from the primary sensory processing of somatic stimuli (somatosensation). However, most of the available studies have mainly investigated the mechanisms of somatosensation. Moreover, disturbances of our body experience have been documented in some pathological conditions of interest for rehabilitative interventions, but their clinical role and relevance is yet to be clarified. Because in this field we have limited knowledge on perceptual and cognitive body experience, there emerges a need to better clarify this matter. The aim of the present scoping review is to systematically map this topic and to examine the magnitude and the nature of the available evidences.

Materials and Methods: The scoping review will be performed following the six-stage methodology suggested by Arksey and O'Malley. Ten electronic databases will be investigated since their inception. The search strategy will be peer reviewed by PRESS 2015 Evidence-Based Checklist as a quality assurance step. All records retrieved will be screened by two independent reviewers. The Population, Concept, and Context method will be adopted for eligibility criteria and Preferred Reporting Items for Systematic Reviews and Meta-Analyses will be used for results reporting. Two reviewers with different background will perform the search process independently. One author will extract data, checked by a second reviewer checking the matching with the research questions and goals. Any disagreements will be solved by a third reviewer.

Ethics and Dissemination: Ethical approval is not required for scoping reviews. Dissemination will include submission to peer-reviewed journal and presentations in conferences in the area of rehabilitation.

Keywords

Body perception, body representation, body image, chronic pain, scoping review

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Introduction

Each of us unequivocally owns a unique object, our own body. It is 'unique' because it is a multisensory object.¹ This univocal multisensoriality is provided by a complex and integrated network of cortical areas coding for different types of stimuli, the so-called 'body matrix'.² The brain houses multiple representations of the body³ and this set of representational systems makes us capable of experiencing what happens to us, viewed both from the inside and the outside. In order to better study and explain this duality, Longo et al.⁴ suggest differentiating

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the modality by which we feel our body from the inside, as an object of direct perception (*somatoception*), from the other modality by which we can cognitively reflect the body from the outside (*somatorepresentation*). With the former, we can perceive our body and objects contacting it, while with the latter we can consider our body as a physical and biological object like another in the external world, with which we establish a relationship.¹

Moreover, Longo et al.⁴ highlight another important difference between these two high-order processes (*somatoception* and *somatorepresentation*), and the basic mechanisms producing the sensations of touch, proprioception, cold, warm, nociception, and vision (called *somatosensation*). These three different mechanisms are integrated and linked,² but are independent of each other. The integrity of the peripheral receptors deputed to *somatosensation*, for example, is neither sufficient nor necessary to produce *somatoception* or *somatorepresentation*. The most striking demonstration of this phenomenon is represented by the illusory perception of owning a limb (*somatoception*) when the person knows that it has been amputated (*somatorepresentation*), and all the sensitive receptors are physically missing from that body part (*somatosensation*). The fact that this perceptual sensation is referred to as vivid and has detailed sensorial characteristics indicates that the peripheral information has been stored somewhere centrally in the brain: sensations from the 'old' limb are maintained in a body structure model of reference and continue to exist even in the absence of the limb itself.³ It follows that there may be a dissociation between these different components of mental representation of the body and the primary sensory process because each one has different underlying functional and neuro-anatomical bases (see Longo et al.⁴ for a detailed dissertation and other examples).

However, we have a less accurate knowledge of *somatoception* and *somatorepresentation* because the mechanisms underlying *somatosensation* have been the primary focus of studies conducted in experimental Psychology and Neurophysiology.⁴

Pain itself can induce alterations in body/space perception.^{5,6} In turn, modifications in the way by which we perceive our bodies (e.g. through visual manipulations)⁷⁻⁹ or the space with which our bodies interact^{10,11} may have implications in pain perception. These results highlight how the complex experiences of pain perception and body perception itself may depend on multisensorial representations stored in our brains, and how they may be potentially linked.^{2,12,13} Pain represents the conscious perception that the body is threatened; it follows that it is fundamental to understand how a body that needs protection is perceived, because these mechanisms may be related to the emergence of pain itself.¹⁴ It is noteworthy from a clinical and therapeutic perspective that this kind of 'bizarre' body perception disturbance may not be referred by patients to health care providers, if it is not directly questioned^{15,16} for fear of not being believed¹⁷ or of

being considered mentally disturbed.¹⁸ Thus, overall, how we mentally represent our bodies may influence our quality of life, social interactions, and mental health.^{17,19}

Phenomena of disturbed *somatoception* and *somatorepresentation* have been documented in a variety of clinical neuropathic pain conditions affecting the musculoskeletal system, and particularly in Complex Regional Pain Syndrome (CRPS)^{15,17,20,21} and phantom limb pain,^{22,23} but remain undiscovered for musculoskeletal disorders and rheumatic diseases (MDRDs). Moreover, while compelling preliminary evidence about interactions between body representation and space perception have been published in CRPS,^{11,20,24-27} this interaction remains to be clarified in MDRDs. Considering the correlation between body perception dysfunction with pain intensity^{17,26,28} and duration^{17,29} highlighted in CRPS, it seems to be clinically important to clarify if this link exists in MDRDs. Thus, in order to have a complete overview of this research area, a literature search must be conducted to guide further research and clinical practice. Figure 1 summarizes the theoretical bases and the aims of this review.

Moreover, different terminologies have been used in scientific literature to describe body perception and mental representations, and the relative meanings assigned within this terminology are sometimes ambiguous or contradictory,³⁰ making it difficult to uniformly cover this research area. Although the issue of the terminology used is still under debate,³¹ for the purpose of the present review, we arbitrarily refer to *somatosensation* and the other higher-order processes, namely, *somatoception* and *somatorepresentation*, as defined by Longo et al.⁴

Aim

The aim of this review is to map and examine the magnitude and nature of available published research about disturbances of explicit and implicit¹⁹ mechanisms of body perception, body representation, and perception of surrounding space in MDRDs. The present protocol represents a detailed, step-by-step description of how we intend to conduct the scoping review in order to limit the occurrence of reporting bias. Any deviations from the protocol that may occur will be addressed and explained in the final scoping review report.

Materials and methods

The scoping design approach was selected in order to systematically^{32,33} map and synthesize the research trends and findings on body perception and body representation that seem to be applicable in the clinical context of MDRDs. The scoping study represents the most appropriate method of over-viewing the literature about an emergent research area which is still fragmented, complex, wide, poorly understood, or not previously deeply investigated.^{33,34} Our methodology is inspired by recently published scoping studies and

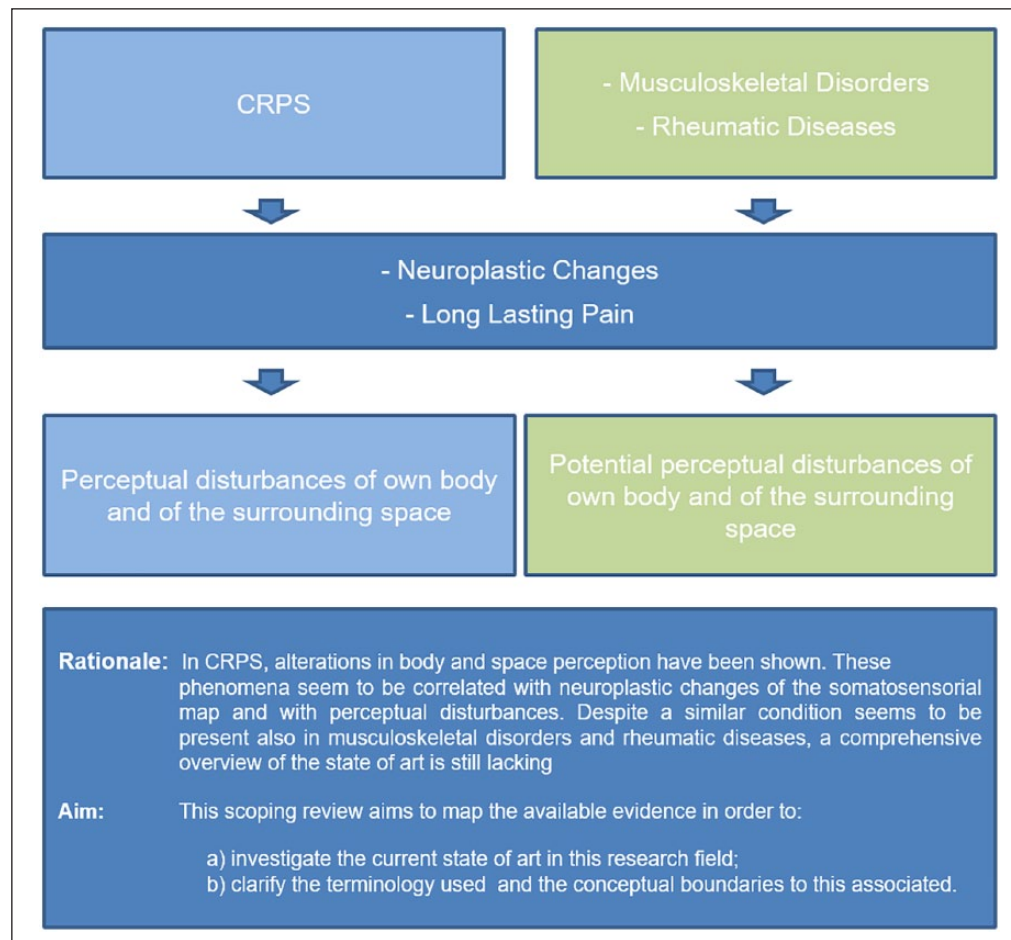


Figure 1. Summary of theoretical bases and aims of the scoping review.

protocols.^{34–36} Our research team will use the six-step framework for scoping review developed by Arksey and O'Malley³⁷ and supplemented by Peters et al.,³⁸ as a guide. Moreover, it will follow the recommendations of Daudt et al.³⁹ to enhance the six framework stages of Arksey and O'Malley. The six steps (five mandatory and one optional) are detailed below:

Step 1. Identifying research questions

- Are there any documented modifications of the implicit and explicit cognitive and perceptual representations of one's own body in MDRDs affected patients?
- Do these phenomena show different features between MDRDs patients and those with other disorders, or are there different features between MDRD sub-groups?
- What are the methodologies used to explore these phenomena? Are they easily applicable in clinical practice?
- When related to the body, the terms *perception*, *representation*, and their synonyms can be ambiguous and differently interpreted depending on the specific reference areas and disciplines. We aim to investigate what terminology is used in different publications, by

whom and for what purpose, including inter-article concordance.

Step 2. Identify relevant studies

We will follow the acronym PCC (Population, Concept, and Context) proposed by The Joanna Briggs Institute⁴⁰ to describe elements of the inclusion criteria.

Population

The studies included will report data about adult individuals (>16 years old) of both sexes, affected by MDRDs, for example, low back pain, neck pain, osteoarthritis, rheumatoid arthritis, and fibromyalgia.

Concept

The main concept of this review will be the potential alterations involved in how patients perceive their body and/or surrounding space. Another core concept of this review refers to the characteristics (entity, typology, theoretical bases, and practical implications) of the potential disturbances involved.

Context

Literature specifically concerning MDRDs will be reviewed. Both experimental and clinical settings will be considered. However, we will focus mainly on the studies which relied on measures, methodologies, and/or tests that are administrable in clinical practice.

Types of studies

Quantitative and qualitative studies will be included in this review in order to better overview the current state of the art about the topic. Details on criteria for including and excluding studies are shown in Table 1. Studies that are included must be published on humans in English or, if not in English, then containing an English abstract, with additional information of an earlier or ongoing trial study report. Data from these last sources will be extracted based on the information available in the abstract. However, we will keep a record of studies published in other languages in order to provide a general overview of the amount of international literature published.

Measure of outcomes for body perception and representation

The details are summarized in Table 1.

Search strategy and information sources

The multidisciplinary team composed of physiotherapists, neurologists, and psychologists will define search strategy. As recommended by Valaitis et al.,⁴¹ we will follow a three-step search strategy. First, a preliminary search will be pilot tested in order to pre-select key words from abstracts and titles of papers considered relevant to the topic. This step will only involve two online databases, Medline (PubMed) and the Cochrane Library. Moreover, we will use PubReMiner (a free version data mining tool: <http://hgserver2.amc.nl/cgi-bin/miner/miner2.cgi>) in order to retrieve additional key words and Mesh terms to assemble in the final search query. Based on the key words and Mesh terms retrieved, a definitive search strategy will be developed using Medline (PubMed interface) (see Supplemental Appendix I). This primary search strategy will be peer-reviewed independently by two of the co-authors using the PRESS 2015 Evidence-Based Checklist⁴² and re-adjusted after suggestions. The search strategy is detailed in Supplemental Appendix I. Search history and the peer review of the search strategy will be available upon request. We will use a wide range of search terms but also manual filters for Species (Human) in order to balance the comprehensiveness and specificity of the search strategy with feasibility. No date limitations will be imposed for the search strategy.

A secondary search strategy will be performed by adjusting the primary search strategy for all the other databases

included. Finally, in order to retrieve additional sources, a third strategy will be adopted: the four journals with the highest impact factor in the field of MDRDs will be electronically searched, and the reference list of all the articles and reviews of interest identified will be scanned. Moreover, inherent articles provided by each member of the team after searching personal databases will be analysed. If necessary, authors of the original studies will be contacted for further information or missing data.

Differently from systematic reviews, scoping reviews are characterized by an iterative search process rather than a fixed and pre-established one. For this reason, our search strategy and the inclusion/exclusion criteria may be changed and updated during the development of the review, following the feedback of the research team, if required.

Data management. All references of identified records will be imported and managed with Zotero (<https://www.zotero.org/>); this will allow us to remove duplicates. A Google Drive folder will be used to share sources between team members.

Electronic databases. The search will be conducted in the following electronic databases: MEDLINE (PubMed), The Cochrane Library, The Cochrane Central Register of Controlled Trials (CENTRAL), The Campbell Library, the Physiotherapy Evidence Database (PEDro), SCOPUS, PsycINFO, Web of Science, TRIP Database, and ProQuest Central database. Theses and dissertations will be searched on PQDT Open. In addition, a search for grey literature will be conducted through correspondence with specialists and experts as well as using web search engines (i.e. Google, Google Scholar), limited to the first 100 citations. A secondary review will be performed by scanning the Grey Matters Checklist (<https://www.cadth.ca/resources/finding-evidence/grey-matters>) of the Canadian Agency for Drugs and Technology in Health and searching the conference abstracts contained therein. The search strategy will be modified and adapted, if required, in order to balance the thoroughness and relevance of the records with the total volume and the feasibility of data management. The International Prospective Register of Systematic Reviews database (PROSPERO) has been previously searched in order to check whether there are any reviews already registered on this topic.

Step 3. Study selection

The reviewing process will be carried out in two different steps: (1) preliminary identification of relevant studies based on the title and/or abstract; (2) study selection based on the eligibility criteria (see Table 1). For the first step, before starting the screening, the researchers will perform a pilot test, pre-formal screening for a random 10% of the records retrieved on PubMed as a calibration exercise, with the goal of improving reliability across reviewers. For the second step, pre-formal screening will be performed on a random 10% of overall records retrieved on all databases included. For both calibration tests, percentage inter-rater

Table 1. Eligibility criteria for included and excluded studies.

Inclusion criteria	Exclusion criteria
<p>Studies investigating alterations of the implicit and explicit body and space perception, body representation, and the characteristics of neurocognitive impairments (entity, typology, theoretical bases, and practical implications) to these associated.</p> <p><i>Language:</i> Full text in English^a</p> <p><i>Setting:</i> Experimental or clinical^b</p> <p><i>Participants:</i> Studies on humans (>16 years old), male and female Patients affected by musculoskeletal disorders or rheumatic diseases (i.e. low back pain, neck pain, osteoarthritis, rheumatoid arthritis, and fibromyalgia), including radicular syndromes (radicular pain and radiculopathies)</p> <p><i>Study design:</i> Primary research studies:</p> <ul style="list-style-type: none"> Quantitative design: <ul style="list-style-type: none"> Experimental designs (randomized controlled trials, controlled clinical trials, proceedings,^c conference abstracts^c) Observational designs (descriptive studies, surveys, cohort studies, cross-sectional studies, observer-reported or patient-reported outcome studies, case studies^c and case series,^c proceedings,^c and conference abstracts^c) Qualitative designs: phenomenological and grounded theory approaches <p>Secondary research studies:</p> <ul style="list-style-type: none"> of quantitative studies: systematic review with or without meta-analysis, observational of qualitative studies: meta-summary, meta-synthesis <p><i>Outcomes:</i></p> <ul style="list-style-type: none"> Quantitative research design: <ul style="list-style-type: none"> Primary outcomes: measures, methodologies, and tests used by authors to assess body perception, body representation, and perception of surrounding space, the association between pain (intensity and duration) and disturbances of body perception, body representation, and perception of surrounding space Secondary outcomes: the association between neuro-anatomical and/or neurophysiological correlates and measures of body perception Qualitative research design: <ul style="list-style-type: none"> Primary outcomes: the frequency and typology of words used by patients in describing the alterations of one's own body perception or representation in musculoskeletal disorders and rheumatic diseases. Themes and subthemes will be derived by the analysis of patient interviews 	<p>Studies investigating body image intended as the satisfaction about one's own bodily appearance</p> <p><i>Language:</i> Full text and abstract not in English^e</p> <p><i>Participants:</i></p> <ul style="list-style-type: none"> Patients affected by: <ul style="list-style-type: none"> neuropathic pain (e.g. complex regional pain syndrome – CRPS, phantom limb pain) or myelopathies Eating disorders (e.g. anorexia, bulimia) Psychiatric or neurological conditions (e.g. personality dissociation, somatoform disorders, body identity integrity disorders – BIID, dementia, Alzheimer and Parkinson diseases, multiple sclerosis, stroke, cerebral palsy, spinal cord diseases) Congenital, hereditary, or endocrine abnormalities and deformities (e.g. pectus carinatum, phocomelia, acromegaly, gigantism, Marfan syndrome, benign joint hypermobility syndrome) Neoplastic or post-neoplastic conditions (e.g. breast cancer) <p><i>Study design:</i></p> <ul style="list-style-type: none"> Narrative review^d Editorials, commentaries or expert opinion articles, point of view, letters to editors, correspondences or replies to letters,^e book reviews, or chapters Study protocol^f

^aOr full text not in English but with the abstract in English, only if containing additional information of an earlier or ongoing trial study report.

^bWith a particular focus on studies reporting methodologies or test/measures feasible to translate into clinical practice.

^cDescribed with a rigorous methodology.

^dA reference list of narrative review about the topic of interest will be manually scanned.

^eIncluded in the qualitative analysis only if containing additional information on an earlier or ongoing trial study report.

^fExcluded from the analysis but reported in order to provide a general overview of the amount of international literature published.

agreement will have to be >90% in order to start the formal screening.³⁴ If agreement observed is lower, the inclusion and exclusion criteria will be clarified and another round of pilot testing will be performed. Feedback from this test will also be used to refine inclusion/exclusion criteria. Another pilot trial charting exercise will be performed prior to data abstraction in order to improve the adherence of extracted information with research questions and objectives.^{37–39,41,43} Study features coming from a convenience sample of five of the included articles will be extracted separately by two

reviewers. A pilot form of the spreadsheet developed a priori will be used and the inter-rater agreement will be verified. If necessary, the spreadsheet will be modified, based on the research team feedback, until it reaches 90% or more inter-reviewer agreement.⁴⁴

In the first step, records screening will be conducted to determine the eligibility of the studies. Each record will be considered as included, excluded, or uncertain. At this stage, uncertain studies will not be eliminated; these records will be analysed in the second stage, undergoing a full-text review

best fitting the inclusion and exclusion criteria. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow chart⁴⁵ will be used to report the search process and the final numbers of articles included in the qualitative analysis. An excel spreadsheet will be used to report all results from the two searching steps and to assign a univocal identification number to each record.

Two reviewers with different backgrounds, for example, a neuropsychologist and a physiotherapist, will perform all screening steps independently. The multidisciplinary nature of our research team, which includes physiotherapists, neurologists, neuropsychologists, and methodologists, along with its inter-professionality, that is, members trained in clinical practice and academic researchers,^{46,47} is highly appreciable, especially in this area of research, which has historically been addressed within psychological disciplines. By contrast, in this review, the area of research is discussed from the rehabilitation perspective. Discrepancies during the screening phase will be resolved via discussion between reviewers until a consensus is reached; in case of persistent disagreement, a third reviewer will be introduced.

Findings emerging from the review will be organized around the research questions previously mentioned. One reviewer will extract standard information about the studies' characteristics and background, as well as additional information, using a standardized excel spreadsheet (see Supplemental Appendix II), which may be modified after the pilot trial charting exercise. Another member of the team will check the correspondence of data extracted with the research questions and goals of the review by doing a full-text reading.

After this calibration exercise procedure, a single reviewer will extract data from each study included, which will be verified by another member of the team. A third reviewer will resolve any disagreement.

Step 4. Charting the data

A summary for numerical results and qualitative analysis will be reported and, if possible, data will be graphically charted.

Step 5. Collating, summarizing, and reporting the results

Results will be qualitatively and quantitatively synthesized in order to determine what has already been discovered in this research area and what requires a more in-depth investigation, in line with the research purpose. In agreement with the concept of 'literature map', results will be presented graphically as suggested by Peters et al.³⁸

Methodological quality appraisal

Quality critical appraisal of risk of bias will not be performed in line with guidance on scoping review conduct.³⁸ Since studies in this area tend to be non-homogeneous in terms of their

methodology and study designs, the present scoping review is intended as a broad overview of the evidence pertaining to a topic, irrespective of study quality. Moreover, the emerging nature of the topic in question renders it difficult to appraise the quality of the methodology used in each study. In fact, the main goal of this study, unlike that of systematic reviews, is to map and synthesize the evidence available about this topic rather than provide the best available evidence.^{38,40}

Step 6. Consultation phase (optional)

If considered potentially helpful by the research team, a consultation phase will be introduced with the goal of getting additional sources of information and perspectives related to the scope of inquiry.³⁷ More in detail, this step may be introduced if a conflicting use of the terminology emerges related to the alteration in body perception and representation, and/or if the studies included are scarce, come from different settings, or are obtained through very different research designs. This step can potentially provide insights on the meaning and applicability of findings, highlighting what the literature fails to clarify and how research in this area could be improved. Researchers involved in this project are experts in the field of body perception and representation; their publications on this topic attest to their expertise. Moreover, other professionals will be involved (such as physiotherapists, neurologists, psychologists, and neurophysiologists) that may share additional references, provide new knowledge, or improve the debate around the theme of this scoping review through their expertise. Recruitment will take place by personal email invitation. Experts will be invited to discuss the strengths and limitations of published studies, interpretation of the findings, clinical applicability, and proposal to guide future research in this area. Ways to standardize the terminology used if discrepancies emerge in the findings of the review will also be discussed. Email correspondence and face-to-face or Skype meetings will be used to discuss how to improve research methodology within this topic and what main areas could be further investigated. A semi-structured spreadsheet will be administered in order to systematically aggregate the questions raised and the suggestions provided.

Discussion

We aim to review scientific literature published on the dysfunctions of implicit and explicit body perception, body representation, and perception of surrounding space in patients with MDRDs. Our goals are (1) to delimit the state of the art, (2) to clarify the currently used terminology, and (3) to define the clinical usefulness and feasibility of the diagnostic and therapeutic methodologies studied. Through the publication of this protocol, we intend to define and outline the step-by-step procedure to conduct the scoping review in order to limit the occurrence of reporting bias. Any deviations from the protocol will be reported in the final draft of the scoping review.

The scoping design approach has been selected for its appropriateness in overviewing the literature published on an emergent complex and fragmented research area that has not been systematically studied before. We adopt the methodological framework currently recognized for conducting systematic reviews,^{33,37–39,47,48} modelling this protocol on those previously published by other authors.^{34,35}

Ethics

The present review is not a primary research study and does not require formal ethical approval.

Dissemination

In recent years, the publication of review (included scoping review) protocols has increased in order to improve the transparency process, which is considered important as a validity criterion. This research protocol aims to outline the methodological steps planned in conducting and reporting the review. The results of this overview will be of interest for clinicians and researchers involved in the area of body perception, body representation, and perception of surrounding space. It may be of particular interest for physiotherapists treating MDRDs, aiming to better understand body perception disturbances and their potential role in developing and/or perpetuating long-lasting pain.

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Supplemental material

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