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Original article

Complex regional pain syndrome associated with hyperattention rather than neglect for the healthy side: A comprehensive case study

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ABSTRACT

Complex regional pain syndrome (CRPS) is a dehabilitating chronic condition occurring with peripheral lesions. There is growing consensus for a central contribution to CRPS. Although the nature of this central body representation disorder is increasingly debated, it has been repeatedly argued that CRPS results in motor neglect of the affected side. The present article describes a comprehensive and quantitative case report demonstrating that: (1) not all patients with chronic CRPS exhibit decreased spatial attention for the affected side and (2) patients may actually exhibit a substantial, broad and reliable attentional bias toward the painful side, akin to spatial neglect for the healthy side. This unexpected result agrees with the idea that patients can be hyper-attentive toward their pathological side as a manifestation of lowered pain threshold, allodynia and kinesiophobia.

1. Introduction

Complex regional pain syndrome (CRPS) is a lateralized chronic pain condition that usually appears after a traumatic and noxious event such as fracture or surgery and is characterized by severe and disproportionate pain concerning a joint and its neighborhood. CRPS patients show body schema abnormalities [1–3]. Galer et al. observed underuse of the pathological limb, which they related to a kind of motor extinction: the arm can move satisfyingly only if the patient is paying a lot of attention to it. The pathological limb movements were also described as being hypokinetic, bradykinetic and hypometric [4], which had been described in spatial neglect [5]. Twenty years ago, Galer et al. chose the term "neglect-like" to qualify the motor symptoms they observed in CRPS patients: poor motor function and motor neglect complaints expressed by patients. The authors explicitly did not intend to "suggest that the symptoms and signs seen in our patients are analogous to the classic hemispatial neglect that develops following stroke" [4]. Nevertheless, this terminology paved the way for a long series of publications focused on the question of spatial neglect [2,3,6–10].

A considerable amount of theoretical elaborations on this issue have been published [11–18], but relatively few experimental studies are available. Among these contributions, the very notion that CRPS involves spatial neglect had been discussed or challenged. For example, it was proposed that these symptoms could be regarded more like a learned underuse than neglect-like symptoms [16]. Legrain et al. [12] proposed that a top-down attentional bias could be responsible for greater weight given to somatic or nociceptive input, thereby leading to an amplified perception of pain.

Keeping in mind the prototypical picture of spatial neglect [19], CRPS patients obviously do not present such a profound attentional bias regarding all sensory and motor modalities. The most commonly evoked feature of CRPS that has been associated with CRPS is motor neglect (e.g., [16,18]). Several aspects of motor neglect have been described: arm underuse, movement reductions and motor extinction. Arm underuse corresponds to patients' total

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or partial lack of spontaneous movement in the absence of actual motor deficit. Movement reduction can be viewed as a minor form of underuse. Decreased movement amplitude (hypometria), increased latency (hypokinesia) or duration (bradykinesia) have been described in neglect patients acting with their healthy arm toward the left [5]. One yet milder variant of this symptom is motor extinction: although movements can be performed normally in unimanual condition for both hands, bimanual movements unveil a progressive decrease of quality, amplitude and frequency on the pathological side [20]. Clinically, this symptom is commonly explored by using a simple finger-tapping task.

As for neglect patients, CRPS patients show modifications of spatial reference frames. The Sumitani et al. [7] study showed a visual straight-ahead deviation for the 36 CRPS patients they examined. However, surprisingly, this deviation was found toward the painful side, which is the opposite of the implicit hypothesis of "neglect-like" behavior of the pathological side, which would have implied a deviation toward the healthy side, as found in neglect. However, a bias in the perception of the visual subjective body midline has also been found to be independent of the side of pain. Reinersmann et al. [10] observed that in the dark, CRPS patients perceived their visual body midline as shifted toward the left, independent of the actual side of pain, whereas Kolb et al. [9] found no difference between CRPS patients and pain control patients.

Another similarity between neglect after stroke and the spatial cognition disorder in CRPS is the therapeutic effect of prism adaptation. Indeed, prism adaptation is one of the most widely used rehabilitation methods for neglect and also one of the most effective [13,22,23]. It is also an effective method for CRPS rehabilitation [24,25,44]. Prism adaptation for CRPS patients alleviates pain and restores motor ability and range of motion. However, determining the direction of the prismatic displacement is not straightforward, because whether CRPS is associated with neglect or over-representation of the painful limb is unclear.

We targeted several main questions in this case study. First, do CRPS patients systematically exhibit neglect? If so, do they neglect the healthy or the pathological side? Is their deficit limited to motor symptoms for the affected limb or do they expand to perceptual neglect? Do they exhibit a reliable, spatial frame of reference bias similar to spatial neglect patients? To address these questions, we assessed one CRPS patient for motor neglect with 2 kinematic tasks, for perceptual neglect with line-bisection and mental number bisection, and examined spatial reference frames with visual and manual straight-ahead tasks.

2. Material and methods

2.1. Patient

A 50-year-old woman had CRPS type 1 of the left hand due to surgery to remove a benign cyst 3 years before. Intense pain on the back and palm of the hand and the wrist rapidly developed. The patient presented the following symptoms fulfilling the Budapest criteria for CRPS diagnosis: continuing pain that was disproportionate to the inciting event, intense allodynia on the back of the hand, sudomotor changes, motor dysfunction (tremor) and trophic changes (hair loss). She went through several types of therapeutics with no sustainable effect; therapy included the usual analgesic medications step 1, 2 and 3; physiotherapy; occupational therapy; and trans-cutaneous electro-stimulation. She had stopped working; the psychological impact of her disability was huge and her sleep was severely affected. She experienced permanent pain, rated from 60 to 80/100 on a visual analog scale, and showed a pain behavior protecting her pathological hand.

2.2. Spatial frames of reference

Spatial frames of reference were collected following the Rode et al. procedure [26].

2.2.1. Visual straight-ahead

The task consisted of 10 trials for each condition: 100 cm and 200 cm away. The VSA1m involved 3 sessions of 10 trials each and the VSA2m, 2 sessions of 10 trials.

2.2.2. Manual/proprioceptive straight-ahead

The patient was asked to point 10 times with her right hand and 10 times with her left hand in darkness in the "straight-ahead" position in the direction of an imaginary line dividing her body into 2 equivalent halves. Measurement precision was estimated at $\pm$ 0.5 degrees. The test involved 3 sessions of 10 trials each.

For the 2 tasks, we calculated the mean $\pm$ SE of the 3 sessions in 2 ways: from the 3 sessions' means (SE1) and from the 30 measures (SE2). We compared the pooled 30 measures to the theoretical value zero by Student *t*-test.

2.3. Spatial cognition

2.3.1. Line-bisection task

The patient was comfortably seated in front of a table with a A4 sheet of paper laying on the table and aligned with her body axis on which a centered 200-mm long and 2-mm thick line was figured. The patient was asked to mark what she thought to be the middle of the line without making a calculation. The experimental session consisted in 10 bisections with each hand. The distance was calculated by measuring the distance in millimeters between the reported point and the objective midline. A leftward error was signed negatively and a rightward error positively. The results were analyzed by comparing the mean to zero by Student *t*-test.

2.3.2. Testing for line-bisection reference frames

The patient was comfortably seated in front of a table on which there was a higher shelf containing the sheet. The patient's hand lay on the table. The patient performed all bisections with her right hand while her left hand had various positions indicated by the examiner from extreme left to extreme right on the table, and the sheets of paper had also different position from left to right including the middle on the shelf (Figs. 1 and 3). In short, the left unseen hand could lie at the left or the right of each line, which could be at the right or the left or aligned with the patient's body axis. The test involved 6 trials for each combination, in a random order. Two-way Anova was computed to disentangle the contribution of left hand position with respect to the line (right

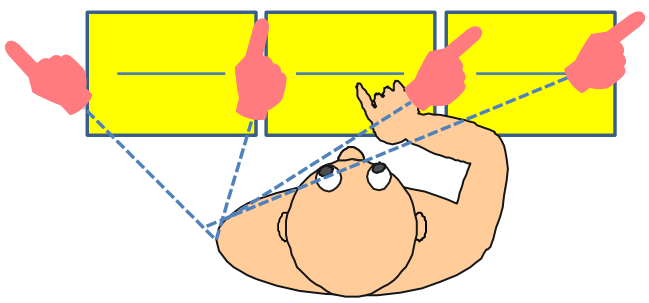

Fig. 1. The line-bisection reference frame task. The 6 combinations of the left hand position and test sheet position are presented. Three test sheet locations tested were left, centred and right to the body midline. For each sheet position, the left hand was positioned under the table in alignment with the left or right edge of the sheet. During this test, bisections were always performed with the right hand.