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Research paper

Structured movement representations of a phantom limb associated with phantom limb pain

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HIGHLIGHTS

- We investigated the relationship between the phantom limb pain (PLP) and its movement representation.
- We used a bimanual coordination task to evaluate the movement representation.
- Negative correlation was observed between PLP and the bimanual coupling effect.
- Structured movement representations of a phantom limb is related with PLP.

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ABSTRACT

The relation between phantom limb pain (PLP) and the movement representation of a phantom limb remains controversial in several areas of neurorehabilitation, although there are a few studies in which the representation of phantom limb movement was precisely evaluated. We evaluated the structured movement representation of a phantom limb objectively using a bimanual circle–line coordination task. We then investigated the relation between PLP and the structured movement representation. Nine patients with a brachial plexus avulsion injury were enrolled who perceived a phantom limb and had neuropathic pain. While blindfolded, the participants repeatedly drew vertical lines using the intact hand and intended to draw circles using the phantom limb simultaneously. “Drawing of circles” by the phantom limb resulted in an oval transfiguration of the vertical lines (“bimanual coupling” effect). We used an arbitrary ovalization index (OI) to quantify the oval transfiguration. When the OI neared 100%, the trajectory changed toward becoming more circular. A significant negative correlation was observed between the intensity of PLP and the OI ($r = -0.66$, $p < 0.05$). Our findings directly suggest that structured movement representations of the phantom limb are necessary for alleviating PLP.

1. Introduction

Movement representations of our body are systemically structured through the cognitive process of sensorimotor integration interacting with the surrounding environment [1]. Deafferentation of a limb frequently leads to phantom limb awareness, and patients perceive vivid kinesthesia [2]. The majority of patients perceiving a phantom limb tend to experience decreased awareness of its kinesthesia, but the phantom limb is “recognized” as fixed in one or more peculiar positions [2]. Accompanying phantom limb awareness, patients with a deafferented limb frequently suffer from phantom limb pain (PLP) with maladaptation of central nervous system plasticity [3]. PLP is often resistant to pharmacotherapy, but it responds to some kinds of neurorehabilitation techniques such as mirror visual feedback in association with plastic change of brain [4–7]. Previous studies demonstrated that PLP patients who restored voluntary movement representation of their phantom limb described PLP alleviation after neurorehabilitation or use of a functional prosthesis [4,5,8,9]. One line of thinking about PLP neurorehabilitation that uses precise visual feedback of phantom limb movements is

Abbreviations: PLP, phantom limb pain; BCT, bimanual circle–line coordination task.

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based on a working hypothesis that incoordination of movement representation of a limb causes pathological pain. However, few reports exist where the representation of a phantom limb's precision of movement was evaluated in behavioral analysis. In the present study, we assessed structured movement representation of a phantom limb objectively using the bimanual circle–line coordination task (BCT) and validated the relation between phantom limb pain and structured movement representation.

2. Methods

2.1. Participants

Nine patients, who suffered from a brachial plexus avulsion injury and perceived a phantom limb and its pathological pain, participated in this study (Table 1). All participants were outpatients at our institute with a chief complaint of phantom limb pain. The Ethical Review Board of the Faculty of Medicine, The University of Tokyo approved this study. We explained the content of this study and the purpose to all subjects and obtained their written informed consent.

2.2. Quantitative evaluation of the movement representation

The bimanual circles–lines coordination task (BCT) used in the present study to assess movement representation of their phantom limb quantitatively has been used in previous studies of various neurological conditions [10–12]. In the BCT, spatial error occurs when drawing the vertical lines repeatedly by the intact hand with intending to draw circles by the affected side (termed the "bimanual coupling effect"). Take the case of phantom limb patients: a coupling effect on the intact hand (drawing straight lines) can be evaluated objectively and quantitatively during "non-visualized" but structured movement representations of the affected hand (drawing circles), even though the affected limb is missing. In a previous case report, Franz and Ramachandran demonstrated that such a bimanual coupling effect during the BCT was observed in an amputee patient with a vivid subjective experience of moving their phantom limb, but was not observed in another patient without the experience [12]. Conversely, straight lines drawn vertically by the intact hand can remain straight when drawing circles using the affected hand by patients with the motor neglect or chronic hemiplegia who have lost movement representations of their affected hand [10]. Based on these observations, we considered the BCT is a promising assessment tool for quantifying movement representations with high validity. The oval-shaped transfiguration when drawing straight lines using one limb indicates that the intermanual interference is induced by the movement representation of the other hand when drawing circles [13]. In addition, converging neuroimaging evidence has revealed that increased activity is observed in motor-related areas, such as the premotor cortex and supplementary motor area, during the BCT [14–16]. On the basis of this neuroimaging evidence, the internal movement representation itself should be sufficient to physically produce the bimanual coupling effect.

The patients sat comfortably in a chair and put their intact index finger on a tablet personal computer (PC) that was on a table in front of the patients. The patients were asked to draw the vertical lines back and forth with the intact hand not intentionally but spontaneously. The intact-hand line trajectories were automatically recorded by the tablet PC. While blindfolded, the patients were asked to perform repeatedly unimanual line drawing movements (drawing vertical lines back and forth on a tablet PC monitor using their intact index finger: unimanual condition: Unimanual Condition) or bimanual drawing movements (drawing the lines using the intact index finger and simultaneously intending to draw circles with the phantom index finger: bimanual condition: Bimanual Condition) at a comfortable speed for 20 s during each trial (Fig. 1). An oval-shaped transfiguration of the repeatedly drawn vertical lines by the intact hand when simultaneously intending to draw circles with the phantom limb indicated that voluntary movement representations of the phantom limb influenced the intact hand (termed the "bimanual coupling" effect [11]). There were two trials for each condition, resulting in a total of four trials.

To quantify the extent of the distortion of the intact-hand line trajectories, we obtained an ovalization index (OI, %) of the lines drawn with the intact hand, according to previous studies [10,11]. From the recorded trajectories in each trial, respective circular figures were extracted by identifying two apical endpoints of respective back-and-forth cycle trajectories. Long and short axes were established for the respective circular figures. An arbitrary variable was calculated from each cycle trajectory according to the following formula: variable = [standard deviation of long-axis data/standard deviation of short-axis data] $\times$ 100. Then, for each patient, the OI was defined as the mean value of the variables computed on all recorded cycle trajectories under the respective conditions. If the OI value was near 0, the trajectory did not become distorted toward a circular transfiguration. If the OI value was 100, the trajectory became a precise circle.

2.3. Subjective evaluation of the movement representation

Phantom limb patients generally describe movement representation of their phantom limb, but their perceptual contents are varied. For example, some describe movement representation as perception of phantom limb to be telescoped, while others describe involuntary motor imagery, or the others describe movement representation only when they perceive a vivid reality of voluntary motor imagery. We designed this study to reveal the intimate relationship between phantom limb pain and subjectively-described movement representation of it. We employed a virtual reality (VR) system to measure specifically the perceptual content of voluntary movement of the phantom limb as homogeneously as possible. The patients wore a head-mounted display (Oculus Rift; Oculus VR, Menlo Park, CA USA) and a three-dimensional computer graphic (3D-CG) of an upper forearm and hand with five fingers presented on the display. The virtual forearm and hand appeared in the patients' correct orientation with respect to their body, and the patients perceived it as occupying the phantom limb. Motion of their intact arm and hand, which was detected by an infrared camera (Kinect; Microsoft Corp., Edmond, WA, USA) and a motion capture data glove (CyberGlove 2; CyberGlove Systems, San Jose, CA, USA), was horizontally flipped like a mirror-reversed image to create virtual limb motion. With this VR system, the patients were asked to exercise both the intact and phantom limbs symmetrically at their discretion (e.g., flexion-extension cycles, rotation of the limbs) for at least 5 min. Subsequently, using a 7-point Likert scale from 0 (none) to 6 (extremely strong), the following two statements were rated and summed: "I felt as if I could exercise my phantom limb voluntarily" and "The phantom limb was brought under control of my will and I could make the limb go where I wanted it to go." The patients conducted this test twice, and the mean score from two sessions constituted the subjective data regarding movement representation of their phantom limb.

2.4. Statistical analysis

To determine whether hand dominance affects the bimanual coupling effect, the coupling effects (bimanual OI scores minus unimanual OI scores) between patients with an impaired dominant hand (dominance group) and those with an impaired