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Case Report

Rehabilitation of somatoparaphrenia with misoplegia: insights from a single case-pilot study

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Somatoparaphrenia lacka ownership of a paralyzed limb, i.e., the illusion that one's limbs belong to someone else. Somatoparaphrenia is one of the many forms of body misperceptions. We report a case of somatoparaphrenia with misoplegia, characterized by the absence of anosognosia for hemiplegia and personal neglect, following a surgical operation for left parietal meningioma. The patient received a novel multidisciplinary treatment, including motor rehabilitation training, traditional physiotherapy and robotic rehabilitation using the Hunova Movendo Technology and psychological counseling. At the end of the training, the patient improved in global cognitive functioning, mood, motor abilities, and the perception of herself and her body, reducing the sense of estrangement and repulsion in the lower right limb. Our result showed the importance of a specific neuropsychological assessment in patients with parietal brain lesions and the usefulness of an integrated psychological and motor approach in rehabilitating patients with somatoparaphrenia, primarily when associated with misoplegia.

Keywords

Body representation; Psychological counseling; Robotic device; Robotic rehabilitation; Anosognosia

1. Introduction

Somatoparaphrenia (SP) lacks ownership of a paralyzed limb, i.e., the illusion that one leg belongs to another [1]. SP is one of the many forms of body misperception, an uncommon, pervasive neuropsychological disorder consisting of delusional sensation in the absence of a confusional state. Body misperceptions include asomatognosia (lack of awareness of a body part), the experience of supernumerary phantom limbs, personification (naming a limb and giving it an identity of its own) and misoplegia. This latter term was coined by Critchley and coll, and it refers to the morbid dislike or hatred of paralyzed limbs [1]. Body misperceptions can result from a brain injury in the parietal, frontal or insular cortex [2], which interrupts and damages a correct body representation (BR), also leading to profound motor and somatosensory deficits [3]. BR is defined as the mental representation of one's body. It results from the integration of multisensory information (visual, sensory, vestibular, and proprioceptive) with the pre-existing knowledge of the individual [4–7]. Thus, BR consists of perception, memory, and cognition related to the body and is continuously updated by sensory inputs due to brain plasticity, reorganizing a different body perception due to internal and external feedback [5, 6, 8–11]. In recent years, some authors have highlighted that SP arises from lesions of specific cortical areas mainly located in the right hemisphere and involving the radiated crown and some subcortical gray matter structures [12–14]. In particular, the frontotemporal-parietal regions are connected to the integrity of one's body and the BR [7, 15].

Recent studies have hypothesized that the repudiation of the patients' body parts with SP is due to deficits in the ongoing dynamic representation of the body involved in perception and action. Thus, the incorrect spatial representation of the contralateral limb to the lesion would result from an erroneous delusional attribution of one's limb [3]. SP presupposes a failure of the brain multisensory areas with an odd sense of possessing a body: this is the reason why Blanke [16] suggests a deficit in coherent multisensory integration that allows for adequate bodily self-awareness. More in detail, SP (like other alterations of body self-awareness) could be due to integration disturbances between proprioceptive, interoceptive, and vestibular signals on body position and movement with visual signals from the body.

SP and the primary body representation disorders have no specific and standardized rehabilitation approach [2]. SP and misoplegia tend to regress spontaneously within a few days or weeks. In some cases, however, the disorder tends to persist, negatively affecting the rehabilitation of motor and sensory functions. Hence, an increase in awareness of the limbs is necessary to facilitate the patient's recovery. Due to the clinical complexity of these cases, Feinberg & Venneri [17] underlined that an approach including cognitive, neuroanatomical, psychological, and motor components could help promote a patient's recovery. Indeed, there is insufficient evidence of positive outcomes for such patients in the literature. Rehabilitation consists mainly of behavioral methods that allow the patient to explore his/her body (use of suggestions, mirror, and verbal commands) [18–21]. Indeed, some authors have observed that the modulation of sensory signals by caloric vestibular stimulation [18–20] and visual inspection of the renegade limb in a mirror [21] could reduce the delusions due to SP.

SP and the other form of body misperceptions, such as misoplegia are complex conditions, which affects the patient's clinical recovery and his/her quality of life. Then, such disorders should be rehabilitated using a holistic approach, as both socio-psychological and biological factors contribute to their complex etiopathogenesis. The aim of the study was the development of a new protocol to rehabilitate body misperception. We managed a patient who complained of right hemiparesis with SP and misoplegia after a neurosurgical intervention, using a novel multidisciplinary approach.

2. Case description

A 50-year-old, right-handed, Italian woman attended our Institute (in day hospital regimen) from June to September 2019, owing to a right hemiparesis after a neurosurgical intervention for a left parietal meningioma (occurred in March 2019). She was an English/French teacher and an unmarried translator and lived with her 80-year-old mother. Her family and personal history were negative for neurological and psychiatric disorders.

At the admission, the patient presented with difficulty standing in the upright position, abnormal gait and right moderate hemiparesis, mainly involving the lower limb. No alteration of proprioception or sense of vibration was observed. No cognitive deficits were reported after the surgical intervention and during the rehabilitation period in another Hospital. However, when reviewing her past rehab history, we found that the patient systematically refused the exercises focused on her lower limb. No one investigated the reason why she behaved this way. The patient also reported that this was the first time she talked about this problem. A brain MRI scan revealed the results of the parietal vertex craniotomy, with the surgical cavity surrounded by peripheral gliotic reaction and Wallerian degeneration of the left corticospinal tract. No Gadolinium enhancement was appreciable (Fig. 1).

2.1 Outcome measures

We provided the patient with an extensive neuropsychological evaluation focused on BR, cognitive and linguistic functions, as the patient presented with a left-brain lesion (Tables 1,2,3). The neuropsychological assessment was performed by a neuropsychologist who was blind to the conditions of the treatment. The cognitive and linguistic functioning was measured through a test battery consisting of Montreal Cognitive Assessment (MoCA), Frontal Assessment Battery (FAB), Beck Depression Inventory-II (BDI II), Hamilton Rating Scale for Anxiety (HRS-A), Token Test, Phonological Fluency, Semantic Fluency, Rey Auditory Verbal Test-Immediate, Rey Auditory Verbal Test-Long term, Digit Span, Attentive Matrices, and Weigl Test (Table 1). The patient showed normal cognitive functioning and mood, despite the presence of a moderate anxious state. Also, she was able to comprehend and speak correctly.

The presence of extra-personal spatial neglect was investigated employing the Albert Line-cancellation [22], the Letter-cancellation tasks [23], and the Line-bisection task. The patient successfully filled all of the tests to exclude the presence of neglect. Personal neglect was assessed according to the procedure defined by Bisiach and colleagues [24]. Awareness for contralesional motor deficits was explored using the four-point scale by Bisiach and colleagues [24]. The patient scored 0, which defines full awareness of the deficit and excludes anosognosia.

The BR evaluation included the Human Figure Test (HFT) and the Body Uneasiness Test (BUT (Table 3)). Somatoparaphrenia was evaluated by interviewing the patient on the contralesional limb, as performed in other studies [25], including the following questions: "What is this? Whose limb is this? Where is your limb? What do you think about your limb?" The patient was diagnosed with SP as she firmly maintained that her foot did not belong to her. Such explanations were elaborate, strange, persistent, and refractory to correction that she had even written stories about "the foot", separated from her will or the rest of the body [26]. We considered the patient affected by pure SP, as she presented with firmness and refractoriness to corrections, delusions of disavowal of the concerned part of the body while being aware of the hemiplegia (Bisiach score <2). In addition, through an interview, we also detected misoplegia, because the patient defined the foot as an enemy, capricious, that she did not want to help her in rehabilitation and with feelings of hatred towards this part. Therefore, she correctly indicated all parts of her body and the examiner's body, without signs of astereoagnosia, finger agnosia, and visual field impairment.

However, HFT and the BUT scale highlighted problems with the BR, with a failure in recognizing the right foot, which the patient believed to be "alien and separate from her", and "an uncooperative enemy". We concluded that the patient did not show anosognosia due to hemiplegia or personal and extracorporeal neglect: SP was limited to the right lower limb, with misoplegia.

Given that misoplegia and SP frequently fluctuate from day to day (sometimes even hour to hour) with frequent spontaneous remission, we took multiple observations of SP (once a day at different times for a week) to exclude an effect of time *per se*. This procedure was taken into account also for post-intervention assessment. Average test scores were considered for statistical analysis purposes.

The patient was also provided with a standard EEG recording to seek whether the responsibility of the so-

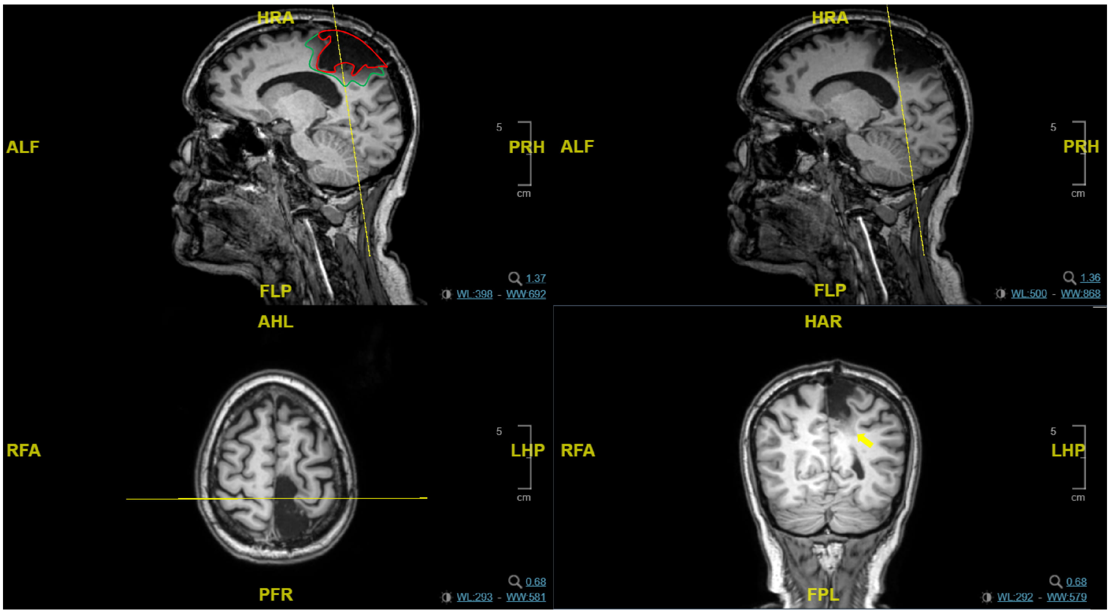

Fig. 1. Brain MRI scan showing the results of the parietal vertex craniotomy, with the surgical cavity (red line delimited area) surrounded by peripheral gliotic reaction (green line delimited area) and Wallerian degeneration of the left corticospinal tract (yellow arrow).

matosensory network to tactile stimulation was abnormal. We compared the neural activity (using eLORETA plugin into EEGLab) resulting from tactile stimulation (according to somatosensory evoked potentials standards) [27] of the left and right lower limbs (randomly performed) without making the patient aware of which leg was stimulated. We averaged together both the evoked responses. Then, we sought whether the evoked activities were different between the lower limbs by comparing the evoked neural activations, to elucidate whether an association with SP and misoplegia was present. Lastly, we sought whether an abnormal sensory network activation could have influenced motor network activation by analyzing the EEG data acquired during repeated, alternated ankle dorsiflexion-plantarflexion (1 cycle per sec) of both the lower limbs.

2.2 Procedures

Following the evaluation, as no standardized treatment for SP exists, the patient received a multidisciplinary treatment, including motor rehabilitation with traditional physiotherapy and robotic rehabilitation using the Hunova Movendo Technology and psychological counseling (PC). This novel multidisciplinary approach aims to promote a global and integrated management of the patient, allowing for better personal and body awareness.

The PC aimed at promoting an integrated vision of the body in the patient through a guided auto-observation. The therapeutic alliance has proven to be fundamental, allowing the patient to verbalize her emotional experiences [28]. Through the technique of positive connotation, it was possible to understand that splitting the foot from the body served to remove the negative experiences related to the disease and personal events. Cognitive restructuring helped the patient give meaning to the reactions of fear, panic, and anger and recognize the foot as an integrated body part. PC was performed twice a week for 8 weeks, each session lasting about 1 hour.

According to the Bobath approach (3 times a week, for 8 weeks), traditional physiotherapy was provided to improve balance, reduce spasticity, and increase muscle strength on the left side.

Lastly, the robotic rehabilitation (which was the main novelty of such an integrated approach) was performed using the Movendo Technology Hunova, a device composed of sensors that analyze movements: the position of the trunk, limbs, used power and rhythm are constantly monitored, allowing the operator to intervene, if necessary. A balance and joint mobilization therapy were implemented with a footplate. Hunova allows precise and objective assessments, and training is progressive with increasing difficulty. The device allows customization of the difficulty: 9 sessions in 3 blocks, divided into easy/medium/difficult levels. The sessions were held in monopodalic mode (right foot) considering balance, stability limits, strengthening of the lower limbs, trunk control [29]. Each treatment session lasted 60 min and was performed three times a week for 8 weeks.

2.3 Statistical analysis

To assess the patient's improvement (between T0 and T1), we used the Reliable Change Index (RCI). RCI is a statistic used to determine whether a change in an individual (or group) score is statistically significant based on the test-retest reliability of the measurement [30, 31]. RCI is calculated by dividing the change in an individual's score by the standard error of the difference for the test being used. The cut-off value for statistical significance within the RCI is $\geq$1.96 (1.96

Table 1. Tools of neuropsychological and functional measure.

Test/Scale	Domain	Description
MoCA	Cognitive domains	The Montreal Cognitive Assessment is a brief cognitive screening assessing: the short-term memory, visual-spatial abilities, executive functions, attention, concentration and working memory, language and orientation in time and place.
BDI II	Depression	The BDI II is a test consisting of 21 items that evaluate cognitive, affective, motivational and somatic symptoms of depression.
HAM-A	Anxiety	The Hamilton Anxiety Rating Scale is a rating scale developed to measure the severity of anxiety symptoms; the scale consists of 14 items, which measures both psychic anxiety and somatic anxiety.
FAB	Executive functions	The Frontal Assessment Battery is a cognitive and behavioural battery to assess frontal lobe functions. The FAB consists of the following six tasks: similarities; verbal fluency; motor series; conflicting instructions; inhibitory control; prehension behavior.
Weigl test	Executive functions	The WEIGL Test assesses the abstraction process and the flexibility of thought to operate different categorizations. The test requires short administration time and reduced attention load.
Fluency phonological semantic	Executive functions language	The verbal fluency test consists of two tests: semantic and phonological. Those semantics were analyzed by asking the words belonging to 3 semantic classes "animal, colors, fruit", in 1 min. The phonological one was evaluated after the first test, by pronouncing words starting with the letter "F, P, L" in 1 min for each letter.
Token test	Comprehension	The token test is used to evaluate listening comprehension. The stimuli are made up of 36 verbal orders divided into 6 parts with increasing difficulty.
Rey auditory verbal test	Memory immediate long term	This test measures immediate memory span and a learning assessment. The test consists of 5 presentations, with recall, of a list of 15 words. In addition, it also assesses long-term memory by asking to recall the list after 15 min.
Digit span	Memory	Digit Span measures memory in two ways: Digits Forward, the storage of forwarding digits, and Digits Backward, the storage of backward digits. The examiner reads a numerical sequence (one number per second) and when the sequence is repeated correctly by the subject, the examiner continues with the next sequence, which is one number longer than the previous one.
Attentive matrices	Attention	The Attentive Matrices is composed of three different number matrices, the subject must cross the target numbers. The administration time is approximately 5 min. It allows for the evaluation of selective attention.
BUT-A	Body representation	The Body Uneasiness Test (part A) is a self-report of 34 items that assesses the discomfort related to one's body image. The sum of the scores of the 5 scales determines a total value that corresponds to the global severity index-GSI.
BUT-B	Body representation	The Body Uneasiness Test (part B) is a self-report consisting of 37 items for the assessment of specific concerns regarding various parts of the body and other aspects such as concern associated with one's own smell, blush, sweat and noises emitted by one's own body.
Human figure test	Personality	The test of the drawing of the human figure is a projective test. The figure that the subject draws is one projection of the self-image. It is possible to get indications about the presence of conflicts related to the intimate sphere of the Self and the relationship with one's own body image.
Ashworth scale	Spasticity	The Ashworth scale is a qualitative assessment tool for spasticity. This scale assesses the resistance of the muscle in passive stretching. This resistance is assessed for the upper limbs (shoulder, elbow and wrist), and the lower limbs (hip, knee and foot). Muscle tone is described by a score from 5 (maximum spasticity) to 0 (absence of spasticity).
FIM	Disabilities	The Functional Independence Measure (FIM) is a tool for assessing the functional status of patients during the rehabilitation process. FIM assesses the degree of disability based on a patient score in 18 categories, focusing on motor and cognitive functions. Each item is rated on a 7-point scale.

equates to the 95% confidence interval).

3. Results

At the end of the multidisciplinary approach, the neuropsychological evaluation showed improved cognitive functioning and reduced anxiety (Table 2). In addition, the hu-

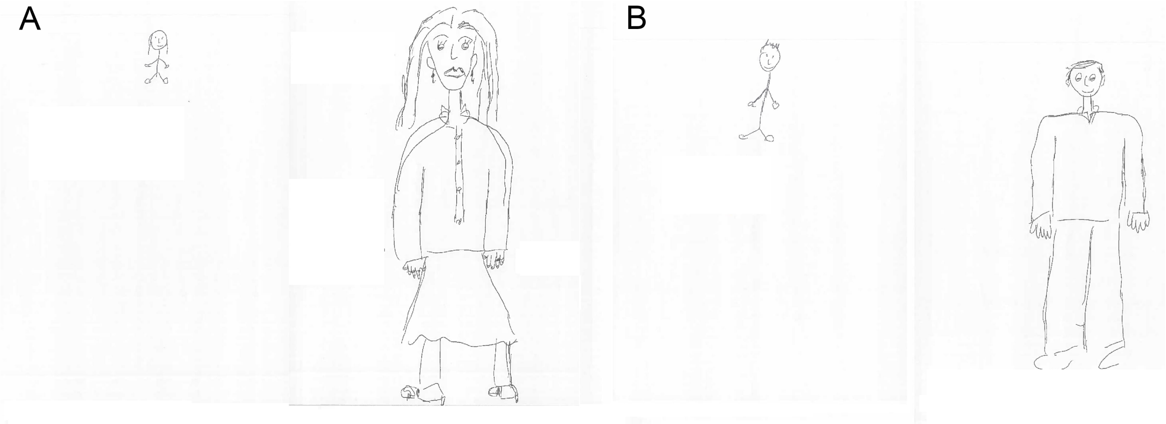

Fig. 2. Female (A) and male (B) Human Figure Test the baseline (left picture) and at the end of the training (right picture).

man figure test and BUT showed that the patient changed her BR and the perception of herself and her body, without any sense of estrangement and repulsion for the right lower limb (Table 3). In particular, we observed a drastic change in the human figure drawn in the HFT (Fig. 2). Initially, the figure was stylized, with few details, other than the extremities of the limbs (hands and feet) represented by circles. We also notice a difference in representing the two feet, and the right one appears larger and more angular than the left foot in both figures (male and female). At the end of the training, the figure was represented with different details, including clothing, feet with shoes, and well-defined hands. The pencil trait was safer, the drawings occupied the entire page, and the differences between the two feet were reduced (Fig. 2). The patient also noticed these aspects at the end of the HFT. From a motor point of view, the articular excursion of the right tibiotarsal joint improved and the patient's stability and endurance during walking. However, it was performed with the aid of a cane. Moreover, at the end of the training, we observed improving all cognitive domains (Table 2).

At baseline, the first analysis showed increased neural responsiveness in a large cluster covering the right frontoparietal areas, including premotor, motor, and somatosensory cortex. Following the rehabilitation training, we found a more focused activation over the sensory areas, particularly those of the lower limbs (Fig. 3).

Concerning the second analysis, we did not find any significant differences in brain activation following the stimulation of each limb at either baseline or post-intervention.

Last, there was right hemisphere lateralization of cortical activations within the motor areas at baseline, modified in a bilateral motor activation post-intervention (Fig. 3).

Table 2. Cognitive evaluation data, with the related RCI (whether significant).

Psychological evaluation clinical scale	T0	T1	RCI
MoCA	27	29	
FAB	17	18	
BDI II	10	7	
HRS-A	20	11	2.2
Token test	32	33	
Phonological fluency	34	38	
Semantic fluency	35.75	38.75	
Rey auditory verbal test-immediate	46.7	48.7	
Rey auditory verbal test-long term	11.5	12.5	
Digit span	6	8	
Attentive matrices	45.75	49.75	
Weigl test	12	14	

Legend: MoCA, Montreal Cognitive Assessment; FAB, Frontal Assessment Battery; BDI II, Beck Depression Inventory-II; HRS-A, Hamilton Rating Scale for Anxiety; RCI, Reliable change index.

4. Discussion

The patient we described complained of a rare condition, i.e., SP as a surgical outcome of benign tumor lesions. SP following brain lesions usually tends to recover spontaneously in a few days or a few weeks, whereas the symptoms persisted for months and were underrated in our patient. Furthermore, the patient had a left-hemisphere lesion, adding another piece of rarity. Most patients with SP have indeed a lesion in the right hemisphere [32]. Notwithstanding, the assessment and management of bodily misperception, including SP and misoplegia, remains challenging regardless of the pathological basis of the bodily misperception [32]. Therefore, to find new approaches to improve bodily perception should be welcomed [33].

Our case report raises awareness on two main issues concerning bodily misperception. First, the assessment of bodily misperception requires an appropriate neuropsychological investigation in patients with parietal lesions, even when anosognosia and neglect are not clinically evident. Indeed, SP was misdiagnosed before the patient came to our attention, and the diagnosis was fundamental to the patient's tailored treatment. Second, we have used an advanced approach to better manage bodily misperception management.

As previously stated, there are currently no standardized treatments for body-awareness disorders, especially for SP and misoplegia. Considering that these disorders of-

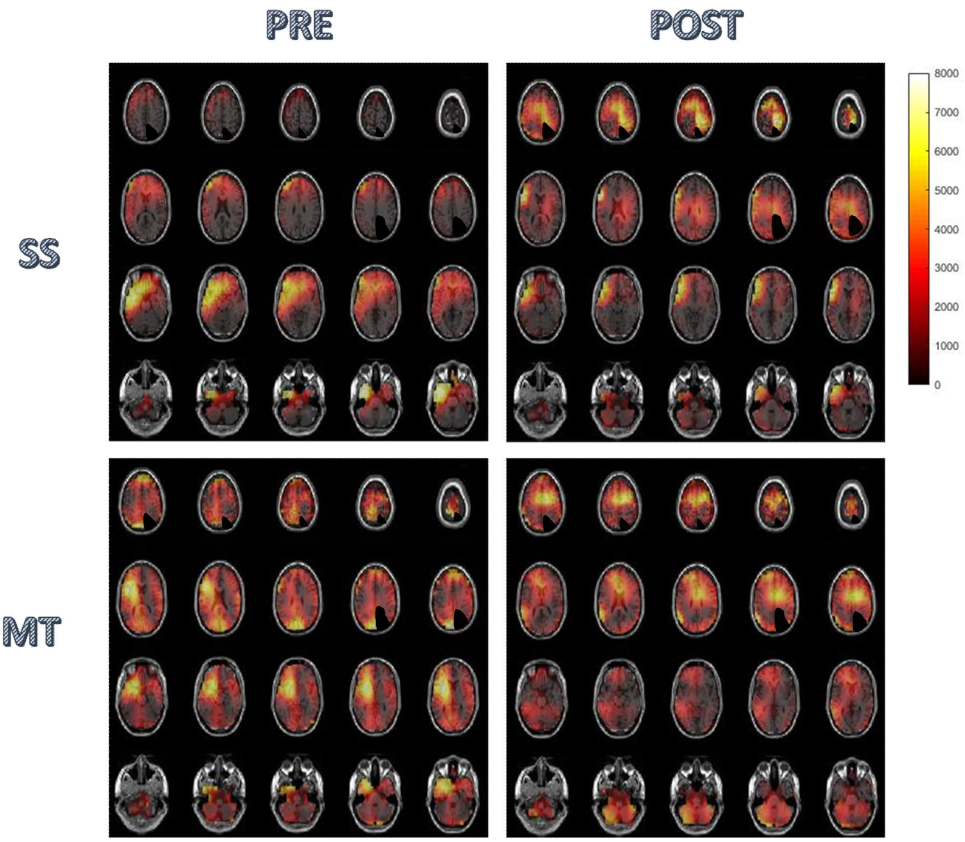

Fig. 3. Representation of the brain areas with statistically significant activations (Z scores on the color bar) on 19-channel analysis in the omega range (full band: 1–45 Hz). eLORETA was used to analyze the cortical distribution of the current source density during bilateral lower limb sensory stimulation (SS) and bilateral stepping motor task (MT) at pre- and post-intervention.

ten regress spontaneously, there is not much attention in the rehab setting. Nonetheless, some studies suggest using awareness-raising therapies, such as providing information on paretic body parts or touching the body parts involved in the neuropsychological deficit [2, 34]. Behavioral or physiological methods, such as vestibular caloric stimulation, have led to controversial results. Recently, Dieguez and Lopez have observed that to better understand this pathology, it could be helpful to use virtual reality, robotics, and neuroprosthetics to learn more about the neural mechanisms involved [35]. On this basis, we decided to combine various rehabilitative approaches: the conventional methods, such as psychological support, physiotherapy, and body exploration and innovative methods, such as robotics, to further potentiate the patient's recovery. Innovative technology has proven effective in patients with BR disorders [36, 37].

Our innovative protocol effectively improved body perception and motor outcomes, thus highlighting the importance of a multidisciplinary approach to treat SP efficiently. For the first time, this case report shows that the combination of intensive motor training (both conventional and robotic) and PC could be a practical approach for SP rehabilitation. PC allowed the patient to acquire more awareness of her body as a whole, thanks to "auto-observation" work, enabling her to modify the dysfunctional thoughts about herself and the syndrome. Indeed, the changes reported in the HFT and the BUT scales might indicate a reconstruction of the process of body awareness, with the re-appropriation of oneself and a sense of bodily belonging. These changes are likely due to the opportunity to verbalize the patient's feelings and the knowledge about her negative emotional experiences, allowing to consider the foot as an integrated part of the body. In addition, traditional physiotherapy coupled with robotic training influenced the patient's outcomes. Indeed, the intensive, task-oriented, and repetitive motor practice probably boosted some neural plasticity mechanisms consistently with the principles of sensorimotor integration [38, 39]. Notably, Kalron *et al.* [40] proposed that cognitive and motor processes are based on shared circuits; therefore, an integrated rehabilitation approach may affect cognitive, sensory and motor processes, favoring the right functionality of the involved circuits.

The concomitant EEG findings also suggest this. Indeed, the patient showed an apparent deficit of activation within the sensorimotor areas within the affected hemisphere and an activation overflow within the unaffected hemisphere. This phenomenon may be consistent with a deteriorated interhemispheric inhibition; actually, a parietal lesion can negatively affect such mechanisms by deteriorating the frontoparietal connectivity related to parietal-lobe-driven cog-

Table 3. BR evaluation of the patient, with the related RCI (whether significant).

Psychological evaluation clinical scale	T0	T1	RCI
BUT-A (GSI)	10.0	7.0	1.9
BUT-A (WP)	0.0	2.0	
BUT-A (BIC)	0.0	1.0	
BUT-A (A)	0.0	0.0	
BU-A (CSM)	3.0	1.0	2.1
BUT-A (D)	7.0	3.0	2.9
BUT-B (PST)	14.0	20.0	3.1
BUT-B (PSDI)	3.14	3.9	2.1
BUT (B I)	2.0	1.0	
BUT (B II)	0.0	0.0	
BUT (B III)	16.0	24.0	2.5
BUT (B IV)	6.0	22.0	4.1
BUT (B V)	2.0	5.0	1.9
BUT (B VI)	10.0	10.0	
BUT (B VII)	2.0	2.0	
BUT (B VIII)	6.0	14.0	3.1
Ashworth scale LL right	3	1	1.9
FIM	104	105	

Legend: LL, Ashworth scale Lower Limb; FIM, Functional Independence Measure; BUT-A, Body Uneasiness Test (part A); BUT-B, Body Uneasiness Test (part B); GSI, Global Severity Index; WP, Weight Phobia; BIC, Body Image Concern; A, Avoidance; CSM, Compulsive Self-Monitoring; D, Depersonalization; PST, Positive Symptom Total; PSDI, Positive Symptom Distress Index; B I, Mouth; B II, Face Shape; B III, Thighs; B IV, Legs; B V, Harms; B VI, Moustache; B VII, Skin; B VIII, Blushing; RCI, Reliable change index.

nitive processes [41, 42]. Furthermore, the parietal damage and the consequent frontoparietal network impairment may account for an overflowing integration activity within the frontoparietal cortical networks to the detriment of the segregation processes [43, 44]. This last aspect is suggested by the lack of any significant difference in brain activation following the stimulation of each limb. Overall, the imbalance between segregation and integration within different frontoparietal networks resulting from a parietal lesion could have distorted BR. We can only hypothesize this pathophysiological scenario, given that many other structures, including the posterior visuomotor area and limbic system, come into play concerning BR [45, 46].

Notwithstanding, our results confirm that the delusions in SP may depend on an altered physiological index of perceptual analysis [10]. Our data point out an apparent impairment in the somatosensory processing of sensory stimuli directed to the contralesional limb compared to those directed to the ipsilesional limb. This is consistent with what has been observed in SP patients by using anticipatory skin conductance response. It seems different from what has been found in control individuals and patients with anosognosia for the somatosensory deficit and with pure motor deficits. These data further support that BR may be affected at different levels following brain damage [10].

Beyond these pathophysiological aspects, the interhemispheric imbalance we found was partially recovered by the combined approach; the magnitude of this change makes the hypothesis that the patient's improvement occurred by chance unlikely. Interhemispheric imbalance recovery also favored a reduction of the right hemisphere lateralization of the cortical activations within the motor areas to a more functional bilateral activation while stepping bilaterally. This issue is of noticeable importance in gait rehabilitation, as it is a fundamental requirement for gait recovery [47]. To summarize, the multisensory stimulation carried out by multidisciplinary and integrated training could have improved BR changes, with an increase in both the motor and cognitive domains. This finding could indicate that BR is an issue that should be considered in rehabilitation programs. It can be modified through environmental stimulation and could also affect the patient's motor and psychological outcomes. Furthermore, our approach positively impacted both SP and misoplegia contemporary. This is not of negligible importance, as these disorders are distinct entities, although belonging to the same spectrum, and they have different neurophysiological bases [18, 48]. SP has been considered a variant or severe form of anosognosia for hemiplegia for many years [49].

Our patient did not show anosognosia for hemiplegia. In line with Sakamoto *et al.* [48], we found that anosognosia for hemiplegia, personal neglect, and somatosensory disorder is not required to diagnose SP.Moreover, the patient presented with SP from a left-sided lesion, which is rare [32]. These unique SP features may explain why the syndrome was not diagnosed before the patient came to our observation. Indeed, there is a predominance of the right hemisphere regarding body representation and somatic attention [50]. However, lesions of the left hemisphere may also cause neglect and related disorders, often associated with aphasia or comprehension abnormalities [51]. Some authors showed that SP could be explained by multisensory integration areas, essential for body ownership and BR sensation. Thus, the dysfunction of these regions, regardless of the hemisphere involved, could produce symptoms of limb dis-ownership [52, 53]. The neural correlates of SP involve specific brain areas, including the parietal and temporal lobe and dorsolateral frontal cortex, which are essential for the BR [3, 7, 9, 12, 13, 54–56].

Consequently, a brain injury in these areas affects the BR and the planning and motor execution skills based on these body representations [7, 57, 58]. This may result in deficits in movement planning, performance, control and imagination, mental rotation, body part position, body part naming, and tools related to body parts [57, 59]. Rehabilitation strategies aimed at targeting BR deterioration can be helpful to improve BR itself and motor outcomes [60].

We acknowledge that our findings are limited by the lack of a comparison of previous treatment strategies and a control group. However, the clinical picture we described is uncommon and challenging to compare. Moreover, given that the spontaneous recovery occurs mainly within the first months after the symptom onset, it is unlikely that this may have happened in our patient, who was treated in the late subacute stage. Therefore, any preliminary approach to managing SP and misoplegia contemporary should be welcomed as in our patient. Notwithstanding, our results suggest that an integrated psychological and motor approach may effectively rehabilitate patients with SP, even in the presence of misoplegia. Further studies with larger samples should be conducted to confirm our promising results.

Abbreviations

A, Avoidance; B I, Mouth; B II, Face Shape; B III, Thighs; B IV, Legs; B V, Harms; B VI, Moustache; B VII, Skin; B VIII, Blushing; BDI II, Beck Depression Inventory-II; BIC, Body Image Concern; BR, Body Representation; BUT-A, Body Uneasiness Test (part A); BUT-B, Body Uneasiness Test (part B); CSM, Compulsive Self-Monitoring; D, Depersonalization; FAB, Frontal Assessment Battery; FIM, Functional Independence Measure; GSI, Global Severity Index; HRS-A, Hamilton Rating Scale for Anxiety; LL, Ashworth scale Lower Limb; MoCA, Montreal Cognitive Assessment; PC, Psychological Counseling; PSDI, Positive Symptom Distress Index; PST, Positive Symptom Total; RCI, Reliable Change Index; SP, Somatoparaphrenia; WP, Weight Phobia.

Author contributions

These should be presented as follows: MGM, AN, RSC and GLR designed the research study. MGM, PC, GLR, BP, DL, PM performed the research. AN analyzed the data. MGM, PC, RSC and AN wrote the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The Institutional Review Board of IRCCS Centro Neurolesi Bonino Pulejo (Messina, Italy) approved the study. Patients provided their written informed consent to study participation and data publication.

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Conflict of interest

Given his role as the Editorial Board Member of JIN, Prof. Rocco Salvatore Calabrò had no involvement in the peer-review of this article and has no access to information regarding its peer-review.

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