



## Clinical Postcard

# A case of somatoparaphrenia characterized by very mild somatosensory disturbance and absence of anosognosia for hemiplegia and personal neglect

Kazutaka Sakamoto <sup>a,b,\*</sup>, Kayoko Yokoi <sup>b</sup>, Kazumi Hirayama <sup>b</sup>,  
Jun Yamaguchi <sup>a</sup> and Atsuo Shinoda <sup>c</sup>

<sup>a</sup> Department of Rehabilitation, Shinoda General Hospital, Yamagata City, Yamagata, Japan

<sup>b</sup> Department of Occupational Therapy, Graduate School of Health Sciences, Yamagata Prefectural University of Health Sciences, Yamagata City, Yamagata, Japan

<sup>c</sup> Department of Neurosurgery, Shinoda General Hospital, Japan

## ARTICLE INFO

## Article history:

Received 6 March 2019

Reviewed 7 March 2019

Revised 11 March 2019

Accepted 12 March 2019

Action editor Sergio Della Sala

Published online 21 March 2019

## Keywords:

Somatoparaphrenia

Somatosensory disturbance

Anosognosia for hemiplegia

Personal neglect

Medial frontal lobe

Somatoparaphrenia is a condition in which an individual has a delusional belief that a paralyzed body part contralateral to a lesion belongs to someone else (Gerstmann, 1942). Typically, the condition is characterized by anosognosia for hemiplegia, personal neglect, and severe somatosensory disturbance (Vallar & Ronchi, 2009). Here we report a case of somatoparaphrenia characterized by very mild somatosensory

disturbance and the absence of anosognosia for hemiplegia and personal neglect.

A 75-year-old right-handed woman sought medical attention following an acute disturbance of consciousness, left hemiplegia, and conjugate deviation of the eyes toward the right side. She was a housewife with 12 years of education, and was diagnosed with a subcortical hemorrhage and underwent surgical removal of the hematoma. One and a half months after the onset of the hemorrhage, she was transferred to our hospital for rehabilitation. She reported the inability to move her left upper and lower limbs; when asked whose hand it was, the woman answered that it was not her hand because she could not move it.

Magnetic resonance imaging revealed hematomas in the superior frontal gyrus and precentral gyrus in the superior part of the right medial frontal lobe (Fig. 1). Neurologically, her left upper and lower limbs were completely paralyzed, but there was no facial paralysis. She exhibited mild spasticity in the left upper and lower limbs and an increased tendon reflex in the left upper limb. Her positional sense of the right side of her body was slightly decreased, and is described later. Further, she showed an ipsilateral instinctive grasp reaction (Mori & Yamadori, 1985) of the right hand, and exhibited utilization behaviors (Lhermitte, Pillon, & Serdaru, 1986), and imitation behaviors (Lhermitte et al., 1986). No other abnormalities were found. Table 1A presents the results of her

\* Corresponding author. Department of Occupational Therapy, Graduate School of Health Sciences, Yamagata Prefectural University of Health Sciences, 260 Kamiyanagi, Yamagata City, Yamagata, 990-2212, Japan.

E-mail addresses: [d2167007@yachts.ac.jp](mailto:d2167007@yachts.ac.jp) (K. Sakamoto), [kyokoi@yachts.ac.jp](mailto:kyokoi@yachts.ac.jp) (K. Yokoi), [khirayama@yachts.ac.jp](mailto:khirayama@yachts.ac.jp) (K. Hirayama), [yamasann1015@yahoo.co.jp](mailto:yamasann1015@yahoo.co.jp) (J. Yamaguchi), [a-shino-win@shinoda-hp.or.jp](mailto:a-shino-win@shinoda-hp.or.jp) (A. Shinoda).

<https://doi.org/10.1016/j.cortex.2019.03.008>

0010-9452/© 2019 The Authors. Published by Elsevier Ltd. This is an open access article under the CC BY license (<http://creativecommons.org/licenses/by/4.0/>).

neuropsychological assessment. To summarize, her general attention, general cognition, and episodic memory were normal and she showed moderate hemispatial neglect.

An investigator pointed at various parts of the patient's body and asked whose body it belonged to. For anywhere on the right side of her body, she answered that it was hers; while this was also the case on the left side of her body for parts of the face, trunk, lower limb, and shoulder, when her left upper arm, forearm, and hand were pointed to, she responded that it was not hers and belonged to another person. Thus, her somatoparaphrenia was limited to the left upper limb. When the investigator held her left hand, showed it to her, and asked, "Whose hand is this?" she replied, "I do not know, heavy, heavy. Whose hand is it really? Who are you?," personalizing her left upper limb. When the investigator asked, "Don't you think it is yours?" she replied, "Absolutely not." After she sat in a wheelchair to leave the room, she said, "I should bring that hand with me, shouldn't I? That may serve as a hot-water bottle." When she was asked where her hand was now, she said, "It is where I was, isn't it? I put it on a towel (pointing her finger at a bed). It should be there. Oh, it isn't there."

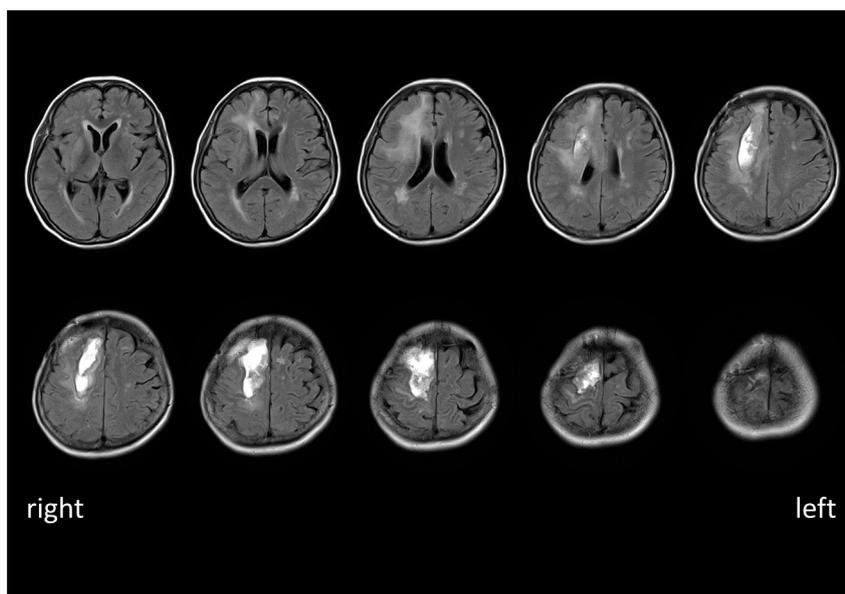
As indicated in [Table 1](#), the patient showed no anosognosia for hemiplegia (1B) or personal neglect (1C). Also, no abnormalities were found in the somatosensory tests ([Table 1D](#)), except for a slight degradation of joint position sense of her left arm. During the examination, the investigator asked the patient, "How could you feel a sensation if it was in the hand of someone else?" and she answered, "I do not know. Why? I would rather ask an expert."

The lesions responsible for somatoparaphrenia have been previously mapped using lesion overlay mapping to various

sites, including the insula ([Baier & Karnath, 2008](#)), the thalamus and surrounding white matter ([Gandola, et al., 2012](#)), and the medial frontal lobe ([Feinberg, Venneri, Simone, Fan, & Nothoff, 2009](#)). In the present case, the lesion was in the medial frontal lobe, which was consistent with the findings of the report from [Feinberg, Venneri, Simone, Fan, and Nothoff \(2009\)](#).

Traditionally, somatoparaphrenia has been considered to be a variation or severe form of anosognosia for hemiplegia ([Critchley, 1953](#)). However, a recent study ([Invernizzi et al., 2012](#)) noted the possibility of double dissociation of these two conditions and suggested that they have different neurological bases. The present case did not exhibit anosognosia for hemiplegia, thereby supporting this notion. [Vallar and Ronchi \(2009\)](#) reviewed 56 reported cases of somatoparaphrenia and found that anosognosia for hemiplegia, personal neglect, and somatosensory disturbance (except for joint position sense) were absent in seven, five, and six of the cases, respectively. However, they found no cases in which all three were missing. Thus, the present case is the first to suggest that none of these three symptoms is a requirement for somatoparaphrenia.

In [Vallar and Ronchi \(2009\)](#)'s study, two of 56 cases did not exhibit position sense disturbance and both patients described their hand as being their own; therefore, the authors concluded that position sense disturbance was a requirement for somatoparaphrenia. The present case involved position sense disturbance, albeit very mild. Furthermore, the majority of patients with position sense disturbance do not develop somatoparaphrenia. Thus, position sense disturbance is likely to contribute only slightly to somatoparaphrenia, if it is indeed necessary at all.



**Fig. 1** – Magnetic resonance fluid-attenuated inversion recovery scan of the patient. Hematomas were present in the superior frontal gyrus and precentral gyrus in the superior part of the right medial frontal lobe. In addition, ischemic changes around the hematomas were noted in the superior frontal gyrus, middle frontal gyrus, precentral gyrus, and the anterior part of the centrum semiovale. However, lesions were not observed in other areas, including the postcentral gyrus, insula, thalamus, and surrounding white matter.

**Table 1 – Results of the neuropsychological and somatosensory examinations.**

Test	Performance	
<b>A. Neuropsychological tests</b>		
<b>Handedness</b>		
Edinburgh Handedness Inventory (max: 100)		100
<b>General attention</b>		
Digit span		7
<b>General Cognition</b>		
Mini-Mental State Examination (max: 30) [higher than 24]		26
<b>Episodic memory</b>		
Recall of three words (max: 3)		
Immediate		3
Post-interference		3
She was able to give accurate oral descriptions of the contents of her previous day's training		
<b>Hemispatial neglect</b>		
Catherine Bergego Scale (max: 30)		0
Behavioral Inattention Test (Ishiai S, 1999)		
Conventional Subtests (max: 146) [higher than 131]		105
Behavioral Subtests (max: 81) [higher than 68]		60
<b>B. Tests of anosognosia for hemiplegia</b>		
Anosognosia questionnaire by Bisiach, Vallar, Perani, Papagno, and Berti (1986) <sup>a</sup> (max: 3) [lower than 1]		1
Estimates of ability on bilateral tasks (Marcel, Tegner, & Nimmo-Smith, 2004)		
Tasks involving both hands (max: 8) [lower than 4]		3
Tasks involving both legs (max: 5) [lower than 2]		1
<b>C. Tests of personal neglect</b>		
Touch the body parts <sup>a</sup> (max: 11)		11
Fluff Test (Cocchini, Beschin, & Jehkonen, 2001) (max: 24)		24
Comb and Compact Test (McIntosh, Brodie, Beschin, & Robertson, 2000) (% bias) [higher than -.11]		
Comb		+29
Powder compact		+26
<b>D. Somatosensory tests</b>		
Thermal nociception <sup>b</sup>	Left arm	Right arm
Pain threshold (the thumb pulp, g)	No left–right differences	
Light touch <sup>b</sup>	10	10
Vibratory sense	No left–right differences	
Two-point discrimination (the tip of the middle finger, mm)	2	2
Graphesthesia (numbers from 1 to 10. max: 10)	10	10
Position sense <sup>c</sup>		
Distal interphalangeal joint of the index finger (max: 7)	6	7
Wrist joint (max: 9)	7	9
Elbow joint (max: 14)	13	14
<sup>a</sup> The patient was asked to touch 11 parts of her body, left and right eyes, eyebrows, ears, cheeks, shoulders, breasts, flanks, thighs, left elbow, wrist, and fingers.		
<sup>b</sup> The thumb pulp, the palm, anterior surface of the forearm, and anterior surface of the upper arm were stimulated.		
<sup>c</sup> Each joint was moved by 1/10th of the normal range of motion. Number of correct responses were counted. Normal ranges of the tests are indicated in the square brackets. max., maximum.		

## Conflict of interest

The authors declare no conflicts of interest.

## Funding source

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

## CRedit authorship contribution statement

**Kazutaka Sakamoto:** Data curation, Formal analysis, Project administration, Writing - original draft, Writing - review &

editing. **Kayoko Yokoi:** Supervision, Writing - review & editing. **Kazumi Hirayama:** Formal analysis, Supervision, Writing - original draft. **Jun Yamaguchi:** Data curation. **Atsuo Shinda:** Data curation, Supervision.

## REFERENCES

- Baier, B., & Karnath, H. O. (2008). Tight link between our sense of limb ownership and self-awareness of actions. *Stroke*, 39, 486–488. <https://doi.org/10.1161/STROKEAHA.107.495606>.
- Bisiach, E., Vallar, G., Perani, D., Papagno, C., & Berti, A. (1986). Unawareness of disease following lesions of the right hemisphere: Anosognosia for hemiplegia and anosognosia for hemianopia. *Neuropsychologia*, 24, 471–482. [https://doi.org/10.1016/0028-3932\(86\)90092-8](https://doi.org/10.1016/0028-3932(86)90092-8).

- Cocchini, G., Beschin, N., & Jehkonen, M. (2001). The Fluff test: A simple task to assess body representation neglect. *Neuropsychological Rehabilitation*, 11, 17–31. <https://doi.org/10.1080/09602010042000132>.
- Critchley, M. (1953). *The parietal lobe*. New York: Hafner.
- Feinberg, T. E., Venneri, A., Simone, A. M., Fan, Y., & Nothoff, G. (2009). The neuroanatomy of asomatognosia and somatoparaphrenia. *Journal of Neurology, Neurosurgery, and Psychiatry*, 81, 276–281. <https://doi.org/10.1136/jnnp.2009.188946>.
- Gandola, M., Invernizzi, P., Sedda, A., Ferrè, E. R., Sterzi, R., Sberna, M., et al. (2012). An anatomical account of somatoparaphrenia. *Cortex*, 48, 1165–1178. <https://doi.org/10.1016/j.cortex.2011.06.012>.
- Gerstmann, J. (1942). Problem of imperception of disease and of impaired body territories with organic lesion. *Archives of Neurology & Psychiatry*, 48, 890–913. <https://psycnet.apa.org/doi/10.1001/archneurpsyc.1942.02290120042003>.
- Invernizzi, P., Gandola, M., Romano, D., Zapparoli, L., Bottini, G., & Paulesu, E. (2012). What is mine? Behavioral and anatomical dissociations between somatoparaphrenia and anosognosia for hemiplegia. *Behavioural Neurology*, 26, 139–150. <https://dx.doi.org/10.3233%2FBEN-2012-110226>.
- Ishiai, S. (1999). *Behavioural inattention test, Japanese edition*. Tokyo, Japan: Shinkoh Igaku Shuppan, Co Ltd.
- Lhermitte, F., Pillon, B., & Serdaru, M. (1986). Human autonomy and the frontal lobes. Part I: Imitation and utilization behavior: A neuropsychological study of 75 patients. *Annals of Neurology: Official Journal of the American Neurological Association and the Child Neurology Society*, 19, 326–334. <https://doi.org/10.1002/ana.410190404>.
- Marcel, A. J., Tegner, R., & Nimmo-Smith, I. (2004). Anosognosia for plegia: Specificity, extension, partiality and disunity of bodily unawareness. *Cortex*, 40, 19–40. [https://doi.org/10.1016/S0010-9452\(08\)70919-5](https://doi.org/10.1016/S0010-9452(08)70919-5).
- McIntosh, R. D., Brodie, E. E., Beschin, N., & Robertson, I. H. (2000). Improving the clinical diagnosis of personal neglect: A reformulated comb and razor test. *Cortex*, 36, 289–292. [https://doi.org/10.1016/S0010-9452\(08\)70530-6](https://doi.org/10.1016/S0010-9452(08)70530-6).
- Mori, E., & Yamadori, A. (1985). Unilateral hemispheric injury and ipsilateral instinctive grasp reaction. *Archives of Neurology*, 42, 485–488. <https://doi.org/10.1001/archneur.1985.04060050087014>.
- Vallar, G., & Ronchi, R. (2009). Somatoparaphrenia: A body delusion. A review of the neuropsychological literature. *Experimental Brain Research*, 192, 533–551. <https://doi.org/10.1007/s00221-008-1562-y>.