

Four Legs

Illusory Reduplication of the Lower Limbs After Bilateral Parietal Lobe Damage

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Objective: To report an unusual disorder of body schema and its neurologic and neuropsychological correlates.

Design and Methods: We describe a patient with a reduplicative phantom illusion of her lower limbs. Motor and sensory functions, as well as mental representation of body and space, were studied during the reduplication experience until its resolution.

Setting: Clinical neurology department in a primary care hospital.

Patient: A 64-year-old, left-handed woman who experienced the uncontrollable and distressing feeling of having 4 legs, without delusional belief, after surgical removal of a right-predominant parasagittal parietal meningioma. This phenomenon spontaneously resolved after 2 weeks.

Intervention: None.

Main Outcome Measures: Clinical neurologic

examinations and standardized neuropsychological tests, with emphasis on tests assessing orientation to body parts, right-left discrimination, and mental orientation in space.

Results: The patient had severe weakness and proprioceptive sensory loss in both lower limbs. She had no disturbances of body schema knowledge but a striking impairment in tasks requiring mental orientation in space, particularly for right-left laterality discrimination. Resolution of the reduplication experience correlated with improvement in the affected spatial abilities, while motor, sensory, and other cognitive functioning did not significantly change.

Conclusion: This patient's reduplicative phantom illusion might be related to the combination of the severe somatosensory loss with an underlying impaired mental representation of relative positions in space.

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AMONG DISORDERS of body awareness in patients with brain damage, anosognosia (or denial) of hemiplegia is most common. Other disturbances with abnormal positive elaborations regarding the affected limbs are more rare. These phenomena may include somatesthetic or kinesthetic hallucinations,^{1,2} somatoparaphrenia,³ delusional reduplication of body parts⁴ (also called pseudopolymelia),⁵ or supernumerary phantom limb sensations.⁶ The hypotheses for these phenomena have been various, referring either to impaired sensation,³ disruption in a putative body schema,⁶ perceptual completion,⁷ partial or implicit anosognosia,⁸ unconscious resolution of cognitive dissonances regarding beliefs about the body and its actual perception,⁸ or symbolic psychodynamic elaboration.^{4,9}

We report a case of imagined reduplication of both lower limbs after surgical removal of a parasagittal parietal meningioma. To our knowledge, this is the first reported case of a bilateral, symmetrical phantom reduplication of limbs following brain damage. We studied both the neurologic sensorimotor and neuropsychological correlates of this phenomenon, as well as their temporal course until recovery.

REPORT OF A CASE

A healthy, 64-year-old, left-handed woman underwent 3 extirpations of a right parietal parasagittal meningioma. The diagnosis had been made 26 months before her hospital admission after weakness and paresthesia had developed in her left leg. A first extirpation was followed by persistent left foot-drop. A second extirpation

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A brain magnetic resonance imaging scan obtained before surgery: T₁-weighted sagittal (top) and axial (bottom) gadolinium-enhanced contrast images. A parasagittal meningioma with a moderate bilateral, right-predominant, mass effect on the mesial parietal lobes can be seen, mainly involving Brodmann areas 5 and 7 in the precuneus and paracentral lobule, as well as compromising the superior sagittal venous sinus.

was performed 15 months later because of a recurring tumor and sensorimotor seizures of the left leg. Three weeks before admission, other partial seizures occurred. Magnetic resonance imaging scans revealed a new recurrence of the meningioma with a moderate bilateral, right-predominant, mass effect on the parietal lobes and extension to the superior sagittal venous sinus (**Figure**). On admission to our hospital, findings of a neurologic examination were unremarkable except for the known paresis of her left foot. Surgical removal of the meningioma was again performed, the procedure being uncomplicated, but the superior sagittal venous sinus had to be sacrificed.

Following the procedure, presumably because of venous infarction, the patient was found to have a severe paresis and anesthesia of both lower limbs. Initially, although there was no overt spatial neglect, the patient acknowledged weakness of the right leg, but not of the left

leg. On the second postoperative day, after she realized that both legs were paretic, she reported experiencing the uncontrollable feeling of having 4 legs. Alertness and orientation were normal and she was not confused. This phantom feeling was rather distressing for the patient, who claimed that it was continuous and could not be voluntarily suppressed. She spontaneously criticized its unreality and was afraid of going insane. "I know it is not possible to have 4 legs, so I know it cannot be true," she noted. However, the patient confessed that at times she could not help checking by touch or sight.

Patient: "When I touch them, I don't feel it . . . when I see there are only 2 legs, it is as if there were 2 invisible ones." Examiner: "So, the 2 legs too many are different?" Patient: "No, they are all the same." Examiner: "But there must be 2 real and 2 unreal?" Patient: "I guess it is so, but they seem all the same, all real to me."

The patient described that all 4 perceived limbs were complete and attached to her body, 2 of them being right legs lying on the right side, and the other 2 being left legs on the left side. They were all normal and equal in size, similarly stretched out on the bed, and heavy. When she was seated, she felt the hips and knees of the 4 legs to be flexed alike. All 4 legs were motionless and subjectively paralyzed. However, the patient was convinced that the 2 left legs could eventually be moved with sufficient effort, while the 2 right legs could not:

I feel that the 2 legs on each side are associated . . . not bound together, but associated . . . they are distinct legs, each a full leg, but they must work together. I think this happens because I want them to be all right, so there are 2 legs on each side, maybe to help each other . . . but it is not a phantasm, you know, it is how things are . . . there are 2 legs too many, I am not mad yet.

There was no other interpretative elaboration, delusion, or reduplication of people, places, or events.

CLINICAL AND NEUROPSYCHOLOGICAL ASSESSMENT

Findings of neurologic examinations on the fourth and sixth postoperative days showed a severe bilateral paresis of the lower limbs, predominating on the right side and including hip flexion, knee extension, and foot dorsiflexion (M2, M2, and M3, respectively), with slowed initiation of movement, bilateral hyperreflexia, and bilateral Babinski sign. Position and passive motion sense was completely absent in the toes, ankle, knee, and hip on the right side (0/10, 0/10, 0/10, and 2/10, respectively), while it was markedly impaired for the toes and hip (6/10 and 3/10, respectively) and moderately impaired for the ankle and knee (8/10 each) on the left side. Tactile, pain, temperature, pressure, and vibration sensations were also markedly decreased on both sides, while topesthesia, graphesthesia, directional sense, and 2-points discrimination were virtually lost.¹⁰ Sensation could not be precisely referred to 1 of the 4 illusory limbs: the patient either could not locate the stimuli to any of them or she located the stimuli to 1 of the 4 legs randomly, even when encouraged to guess. There was no extinction for bilateral homonymous double stimulations. Sen-

Results of Tasks of Body and Space Orientation

Task	Score		Scores (\pm SD) From Healthy Subjects*
	Days 6-7 (Reduplication Present)	Day 15 (Reduplication Resolved)	
Orientation to Body Parts			
Naming touched body parts	20/20	20/20	...
Touching named body parts	20/20	20/20	...
Touching body parts in imitation	5/10	8/10	...
Touching body parts according to schema			
Front-view figure	1/6	4/6	...
Back-view figure	6/6	6/6	...
Right-Left Orientation			
Show-Me Test	10/10	10/10	...
Laterality identification on drawings	9/20	17/20	17 (\pm 3.0)/20
Orientation in Space			
Road-Map Test	13/21	18/21	18 (\pm 1.5)/21
Blank geographic map			
Conventional version	5/5	5/5	5/5
180°-Rotated version	2/5	4/5	5/5
Mirror-inverted version	2/5	3/5	5/5
Mental reorientation	30/84	68/84	78 (\pm 5.6)/84

* Ellipses indicate tasks not performed in healthy subjects.

sory disturbances extended up to the pubic level. Results of the Diller Center Test,¹¹ which requires the patient to indicate the midline of his or her body when tapped along from side to side, were normal on the back as well as on the abdomen for right-to-left taps, but the patient failed in half of 12 trials for left-to-right taps. Sensation and stereognosis were intact in her upper limbs.

A detailed standardized neuropsychological examination was performed on the sixth and seventh postoperative days. The laterality index on the Edinburgh Handedness Inventory¹² was -9% (ambidextrous). Oral and written language functions, oral and written arithmetics, ideomotor, ideational, and orofacial praxis, digit and visuospatial spans, verbal learning, and frontal lobe functions were all intact. Recognition of famous faces, objects, overlapping figures, and illusory contours was normal. There was a slight left neglect on copies of line drawings and the Rey-Osterreith Complex Figure test, but line bisection was correct. Results of the Hooper Visual Organization Test¹³ were moderately abnormal.

INVESTIGATION OF MENTAL REPRESENTATION OF BODY AND SPACE

All testing was performed on the sixth and seventh postoperative days. The patient's performance on the draw-a-person task¹⁴ was unremarkable. Her orientation to body parts was assessed according to the following procedures adapted from Semmes et al¹⁵ and Lezak.¹¹ The patient correctly named all body parts pointed to by the examiner on her own body and on the examiner's body (10/10 each). She also correctly indicated all parts of her own body and the examiner's body by name (10/10 each). On touching her own body parts in imitation of the examiner while facing each other, the patient made errors in distinguishing right and left sides on half of the trials,

although body parts were correctly identified (**Table**). Similarly, on touching her own body parts according to numbered schematic diagrams of a human figure, the patient typically made mistakes concerning the side (left or right) on the front-view figure (1/6 correct), but she made no mistakes with the back-view figure. There was no difference in accuracy between lower and upper limbs. Right-left identification of body parts on verbal command using the Smith Show-Me Test¹¹ (ie, "Show me your left ear, my right foot," etc) was unimpaired. However, on the Culver Laterality Discrimination test,¹⁶ which consists of judging whether line drawings of body parts are from the body's left or right side, the patient's ability was severely impaired (**Table**). Finger recognition and naming of her own body parts, examiner's body parts, and those in schematic diagrams¹⁷ was normal.

These results suggested a failure in right-left reversal and mental reorientation of relative positions in space, which was further explored by the following tests. On the Road-Map Test,¹⁸ the patient was presented with a traced pathway on a schematic street map and asked to tell the direction taken at each turn (right or left) without moving the map; 4 maps were used with 7 turns on each. As shown in the **Table**, the patient's performance was poor. In another task, a blank geographic map of Switzerland was presented in 3 different versions: the conventional way (ie, north pointing up and east to the right), in a 180°-rotated way (ie, south pointing up and west to the right), and in a mirror-reversed way (ie, north pointing up and west to the right). When asked to point to well-known topographic landmarks, the patient performed flawlessly on the conventional map but poorly on both the rotated and mirror-reversed maps (**Table**). A final test of mental reorientation¹⁹ was administered. Different line drawings of objects were displayed as targets along with other drawings of the same object, the

latter being either rotated (half of the trials) or both rotated and mirror-inverted (half of the trials). The patient had to indicate "same" for rotated objects or "different" for mirror-inverted objects. Again, her performance was severely impaired in both conditions (Table).

RESOLUTION

The feeling of supernumerary legs lasted continuously for 14 days. On the last 2 days the patient reported that the feeling had decreased in intensity: it could be experienced only with her eyes closed and was suppressed by vision. The feeling had totally disappeared on awakening on the 15th postoperative day. The same day, a detailed neurologic and neuropsychological examination was repeated as before. Paresis of both lower limbs had improved little and still predominated on the right side (M3 for hip flexion and knees extension, M4- for feet dorsiflexion). Her position sense had improved a little also, mostly in the proximal left lower limb (9/10 and 10/10 for hip and knee, respectively), but it was not significantly changed in the distal left lower limb (4/10 for both ankle and toes) nor in the right lower limb (0/10, 0/10, 0/10, and 2/10 for toes, ankle, knee, and hip, respectively). Except for a moderate improvement in tactile, pain, and temperature discrimination, all other sensation modalities were still impaired as on the previous days. On the other hand, the neuropsychological assessment showed a considerable improvement on all tasks of mental representation of positions in space and right-left orientation (Table). As before, the rest of her cognitive functioning was normal, except for a persisting left spatial neglect on copying tasks. After 9 months of follow-up, somatosensory loss remained severe in both legs; the patient was still unable to walk but she never experienced a reduplication sensation again. Neuropsychological test findings were then normal.

COMMENT

After she postoperatively became paraparetic, our patient experienced for 2 weeks an uncontrollable subjective feeling of reduplication of both lower limbs. The reduplication experience appeared as a continuous somatosthetic-proprioceptive illusion or hallucinosis, rather than as a delusion, because the patient did not elaborate any abnormal belief about it and spontaneously criticized its unreality. Furthermore, since she was rather perplexed and distressed, and there was no evidence of a failure to monitor reality, this feeling cannot be simply considered a form of confabulation. She had no disturbances in her body schema knowledge, autotopagnosia, or right-left confusion in direct, uncrossed testing conditions. To our knowledge, there is no other reported case of a similar reduplication phenomenon of the lower limbs following brain damage without any degree of delusion or confusion. Davis²⁰ observed one patient with head trauma and cervical spinal cord injury who had quadriplegia and felt that he had 2 additional legs tied up with the 2 others. Similarly, Ohry et al²¹ described a patient with traumatic quadriplegia and a duplication sensation of all 4 limbs, the extra limbs being

perceived as parallel to the real ones. However, there are only scant details about the neurologic and cognitive status of both of those patients. Delusional belief in 2 pairs of legs (front and back) was expressed by one patient of Weinstein et al⁴ who had a left hemiparesis after a severe closed head injury and showed many other reduplicative delusions about people, places, and events. Belief in a unilateral illusory supernumerary limb on one affected side (usually a third arm) also has been reported in a few cases following stroke in the right hemisphere,^{6,8,22} head trauma with right frontal contusion and extensive nerve damage in the upper limb,⁹ and in multiple sclerosis with hemisensory disturbances.²³

In our patient, illusory reduplication was associated with a profound proprioceptive sensory loss in both lower limbs caused by bilateral parietal damage. A severe sensory loss in their affected limbs was also noted in several other patients,^{8,9,22} and it was likely to be present as well in the patients with quadriplegia of Davis²⁰ and Ohry et al.²¹ Complete sensory deafferentation, such as may occur with lesions of the peripheral nerves, brachial plexus, or spinal cord, commonly leads to phantom limb sensation.²⁴⁻²⁶ This phenomenon has been used to support the existence of a specific neural network in the brain (neuromatrix) subserving the conscious awareness of body parts (body image), which may persist and construct the phenomenological experience of a body part phantom when deprived of the normally modulating sensory inputs.²⁴ Such phantom limbs may take various positions in space, dissociated from that of the real limbs, but are not reduplicated.^{24,25} Similar completion of the body image in conscious awareness also occurs with severe sensory loss following cerebral damage.²⁷ Thus, in our patient, dense loss of sensation (and motor strength) in the lower limbs could have resulted in the perception of phantom limbs. However, while this would account for one pair of phantom legs,⁸ how can we explain their reduplication?

One hypothesis accounting for the reduplicative experience would be to consider that the patient's history of a slowly growing parietal meningioma and previous surgery had induced a reorganization in the cortical sensory somatotopic representation of the lower limbs.²⁷ A former somatosensory representation, repressed until then, could have been released because of the new postoperative sensorimotor deficit and generated the somatosthetic illusion of duplicated legs. However, we found that the patient's sensory and motor functions did not significantly improve as the reduplication phenomenon disappeared. Therefore, the proprioceptive deafferentation might be a necessary, but not sufficient, condition for supernumerary phantom limbs to occur.

In contrast, as the reduplication phenomenon disappeared, we found that the patient's performance on spatial cognitive tasks also showed a substantial improvement. Indeed, when the illusory reduplication was present, a neuropsychological investigation revealed a rather selective and striking impairment in right-left orientation reversal and other tasks of mental orientation in space. All of those spatial abilities similarly require the mental representation of relative position in space and have been shown to depend on right as well as on left posterior hemi-

spheric areas,^{17,28,29} with both hemispheres most likely participating.^{30,31} Thus, the patient's severe orientation impairment was consistent with her bilateral parietal damage. Subsequently, as the illusory reduplication abated, her abilities of right-left discrimination and mental orientation in space concurrently improved. Other cognitive functions remained notably unimpaired throughout the ordeal.

Therefore, although a parallel time course does not prove a causal relationship, it raises the question of whether a disorder in the mental representation of relative positions in space, particularly so for right-left laterality discrimination, contributed to the illusory reduplicative experience. It is remarkable that in the only other reported case⁹ of limb reduplication where orientation to personal body parts was tested, there was evidence of a similar disturbance in spatial reversal for pointing to lateral body parts on a confronting examiner or diagram, in contrast with flawless performance in nonconfronting conditions. Which, if any, disorder in spatial cognition could underlie both impaired mental orientation in space and somesthetic reduplicative illusion remains open to speculation. There is at least one classic condition where a distorted representation of relative positions in space directly produces an illusory duplication of a unique perception: touching an object with crossed fingers creates the illusion of touching 2 different objects at 2 different places, a phenomenon known since Aristotle's time but still unexplained.^{32,33} Awareness of one's body parts undoubtedly implies distinction of position and orientation in space, and following Bonnier³⁴ and Schilder,³⁵ most authors^{1,36} have postulated that the conscious body image results from a supramodal integration of somatosensory and spatial information. Tentatively, the reduplicative phantom limbs phenomenon might be considered a result of the co-occurrence of 2 deficits: a severe somatosensory loss, allowing a phantom construct of the limbs, and a spatial disorder underlying an inability to represent relative (especially right-left) positions in space. Both deficits might somehow combine in conscious awareness to produce an illusory reduplication of the phantom perception.⁸

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REFERENCES

- Hécaen H, Ajuriaguerra J. *Méconnaissances et Hallucinations Corporelles*. Paris, France: Masson Publishing; 1952.
- Frederiks JAM. Disorders of the body schema. In: Frederiks JAM, ed. *Handbook of Clinical Neurology*. Amsterdam, the Netherlands: Elsevier Science Publishers; 1985;45:373-404.
- Gerstmann J. Problems of imperception of disease and of impaired body territories with organic lesions: relations with body schema and its disorders. *Arch Neurol Psychiatr*. 1942;48:890-913.
- Weinstein EA, Kahn RL, Malitz S, Rozanski J. Delusional reduplication of parts of the body. *Brain*. 1954;77:45-60.
- Bechterev VM. Partial cortical and subcortical paralyses of psycho-reflexive functions. *Obozr Psykhiatr*. 1926;31-41.
- Critchley M. *The Parietal Lobes*. New York, NY: Hafner; 1953.
- Levine DN. Unawareness of visual and sensorimotor defects: a hypothesis. *Brain Cogn*. 1990;13:233-281.
- Halligan PW, Marshall JC, Wade DT. Three arms: a case study of supernumerary phantom limb after right hemisphere stroke. *J Neurol Neurosurg Psychiatry*. 1993;56:159-166.
- Rogers MJC, Franzen MD. Delusional reduplication following closed-head injury. *Brain Inj*. 1992;6:469-476.
- Haerer AF. *De Jong's Neurologic Examination*. 5th ed. Philadelphia, Pa: JB Lippincott; 1992.
- Lezak MD. *Neuropsychological Assessment*. 2nd ed. New York, NY: Oxford University Press Inc; 1983.
- Oldfield RC. The assessment and analysis of handedness: The Edinburgh Inventory. *Neuropsychologia*. 1971;9:97-113.
- Hooper HE. *The Hooper Visual Organization Test Manual*. Beverly Hills, Calif: Western Psychological Service; 1958.
- McCrea CW, Summerfield AB, Rosen B. Body image: a selective review of existing measurement techniques. *Br J Med Psychol*. 1982;55:225-233.
- Semmes J, Weinstein S, Ghent L, Teuber HL. Correlates of impaired orientation in personal and extrapersonal space. *Brain*. 1963;86:747-772.
- Culver CM. Test of right-left discrimination. *Percept Mot Skills*. 1969;29:863-867.
- Benton AL, Sivan AB, Hamsher KDS, Varney NR, Spreen O. *Contributions to Neuropsychological Assessment*. 2nd ed. New York, NY: Oxford University Press Inc; 1994.
- Money JA. *A Standardized Road-Map Test of Direction Sense (Manual)*. San Rafael, Calif: Academic Therapy Publications; 1976.
- Hauert CA, Sevino O. Rotation mentale. In: *Evaluation des Fonctions Cognitives*. Geneva, Switzerland: Faculté de Psychologie et des Sciences de l'Education, University of Geneva; 1995.
- Davis A. Pain and suffering following spinal cord injury. *Clin Orthop*. 1975;112:76-80.
- Ohry A, Gur S, Zeilig G. 'Duplicate limbs' sensation in acute traumatic quadriplegia. *Paraplegia*. 1989;27:257-260.
- Ehrenwald H. Verändertes Erleben des Körperbildes mit konsekutiver Wahnbildung bei linksseitiger Hemiplegie. *Monatsschr Psychiatr Neurol*. 1930;75:89-97.
- Mayeux R, Benson DF. Phantom limb and multiple sclerosis. *Neurology*. 1979;29:724-726.
- Melzack R. Phantom limbs and the concept of a neuromatrix. *Trends Neurosci*. 1990;13:88-92.
- Ettlin TM, Seiler W, Kaeser HE. Phantom and amputation illusions in paraplegic patients. *Eur Neurol*. 1980;19:12-19.
- Laplante D, Degos JD. Troubles inhabituels du schéma corporel par désafférentation périphérique. *Rev Neurol*. 1984;140:45-48.
- Ramachandran VS. Behavioral and magnetoencephalographic correlates of plasticity in the adult human brain. *Proc Natl Acad Sci U S A*. 1993;90:10413-10420.
- Ratcliff G. Spatial thought, mental rotation and the right cerebral hemisphere. *Neuropsychologia*. 1979;17:49-54.
- Mehta Z, Newcombe F, Damasio H. A left hemisphere contribution to visuospatial processing. *Cortex*. 1987;23:447-461.
- Newcombe F, Ratcliff G. Disorders of visuospatial analysis. In: Boller F, Grafman J, eds. *Handbook of Neuropsychology*. Amsterdam, the Netherlands: Elsevier Science Publishers; 1989;2:333-356.
- Cook ND, Früh H, Mehr A, Regard M, Landis T. Hemispheric cooperation in visuospatial rotations: evidence for a manipulation role for the left hemisphere and a reference role for the right hemisphere. *Brain Cogn*. 1994;25:240-249.
- Aristotle; Tricot J, trans. *Parva Naturalia*. Paris, France: Vrin; 1951:103 (460b:16-22).
- Grüsser OJ, Landis T. *Visual Agnosia and Other Disturbances of Visual Perception and Cognition*. New York, NY: Macmillan Publishing Co Inc; 1991:297-303.
- Bonnier P. L'aschématie. *Rev Neurol*. 1905;13:604-609.
- Schilder P. *The Image and Appearance of the Human Body*. London, England: Kegan; 1935:4. Psychiatry Monographs.
- Denes G. Disorders of body awareness and body knowledge. In: Boller F, Grafman J, eds. *Handbook of Neuropsychology*. Amsterdam, the Netherlands: Elsevier Science Publishers; 1989;2:207-228.