LETTERS TO THE EDITOR

Right ipsilateral hypersensation in a case of anosognosia for hemiplegia and personal neglect with the patient's subjective experience

Recently, there have been some reports regarding hyperkinetic motor behaviours contralateral to hemiplegia in acute stroke.[1 2] These behaviours are probably the reflection of early plastic changes of brain maps and circuits after an acute lesion and an active process induced by disinhibition to establish new compensatory pathways.[1] I encountered a peculiar case of a patient with right ipsilateral "hypersensation" after a right hemispheric infarction in the acute period who also presented severe left sensorimotor disturbance, hyperkinetic motor behaviours in the right upper limb, anosognosia for hemiplegia, and personal neglect. It was possible to record the patient's subjective experience of the acute phase, which was helpful for understanding the mechanism of anosognosia.

A 76 year old right handed woman was admitted to hospital soon after the onset of left hemiparesis and hemisensory disturbance. She had undergone implantation of a cardiac pacemaker because of sick sinus syndrome. On neurological examination, she was awake and oriented to time and place, but showed inattention and motor impersistence. There was no aphasia or apraxia, but mild left hemispatial neglect was detected. Left hemiparesis was noticed (upper limb 0/5; lower limb 2/5, and face 3/5). Sensory loss was complete in all modalities in the upper limb and severe in the face and lower limb, being slightly preserved for pain and coldness. She denied the existence of left hemiparesis and had completely lost the sensation of ownership of her left hemibody. When I asked her the owner of her left hand and leg while showing them to her, she remarked that these belonged to her grandmother. Brain CT (figure) showed a fresh infarction in the right precentral and postcentral gyrus, extensively extending to the right medial aspect of the frontal lobe (supplementary motor area).

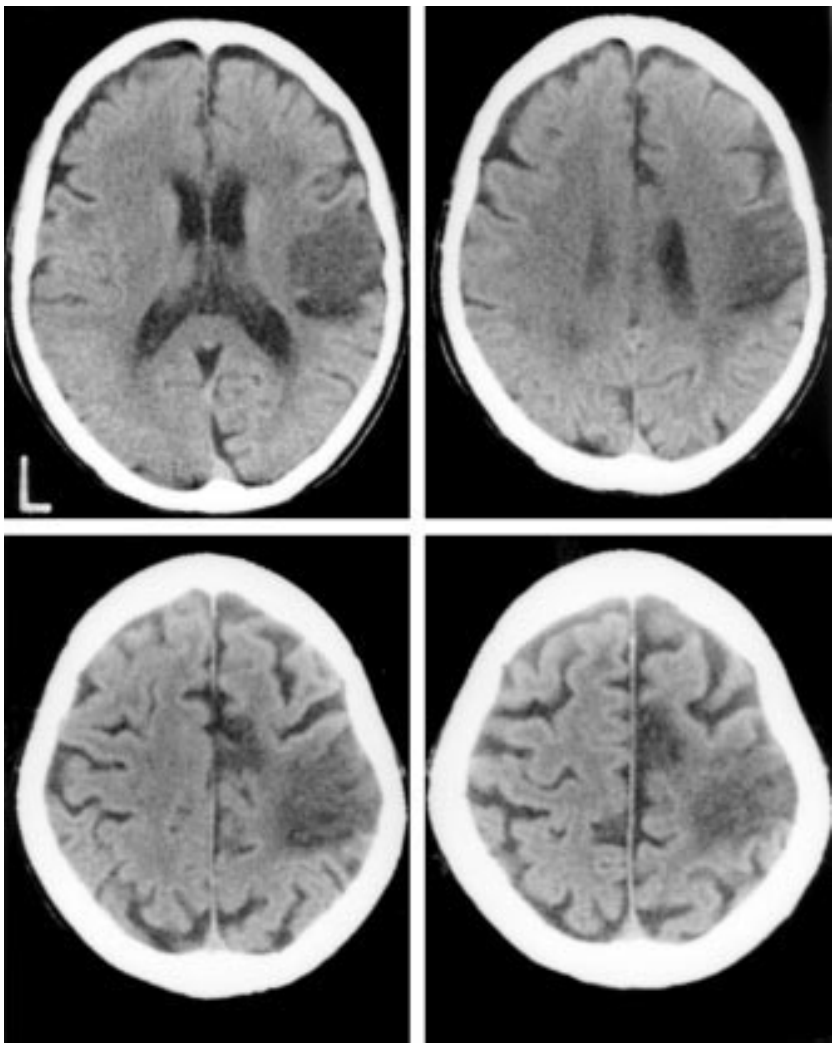
From the second hospital day she complained that she felt very cold in the right half of her body and even sometimes felt pain because the wind from the air conditioner was too strong. I told her that the air conditioning system worked but it was not set at a low temperature because it was winter. She understood my explanation but she continued to complain of spontaneous, abnormal sensation in her right hemibody. The sensation was most severe in the upper limb followed by the face and lower limb, whereas it was not triggered or worsened by any sensory stimulation, and objective sensory deficits were not present in the right hemibody. She usually wrapped herself tightly in a blanket to avoid coldness. She did not complain of any other delusional or illusional feelings. There were also hyperkinetic motor behaviours in the right upper limb such as patting the head with the right arm, manipulations of sheets and blanket, and rhythmic finger movements. The result of a mini mental state examination performed on the fourth hospital day was 25/30.

The abnormal sensation persisted for almost 1 month and gradually subsided, whereas the left hemiparesis and sensory disturbance improved. Touch, pain, and temperature were intact in the face and lower limb, pain and temperature were intact in the upper limb, but there was no improvement in position and vibration in the entire left hemibody. In the meantime, she began to recognise the left hemiparesis and regained the sensation of ownership of her left hemibody. The following are her recollections from the time of onset on the 60th hospital day.

"One morning, I woke up and found that there was a strange hand and foot close to the left side of my body, as though my dead grandmother lay aside me. I tried to throw them off but they were too heavy to move. I glanced at them and felt that they looked flabby and all wrinkled, so I was convinced that they belonged to my grandmother. I had no idea that the left side of my body was disabled or even ill.

After hospitalisation, I felt very cold in the right half of my body and sometimes felt pain because of the powerful wind from the air conditioner. I understood that the hospital did not use cold air conditioning in winter, but that powerful, cold wind could not have come from anything other than an air conditioner. Anyway, this unpleasant feeling gradually subsided, and at the same time, I realised that the disabled left side of my body belonged to me and that I had suffered a brain disorder."

Ghika *et al*[1] described 20 patients with hyperkinetic motor behaviours contralateral to hemiplegia in acute stroke who were found only with large infarcts in the territory of the internal carotid artery, middle cerebral artery, or the anterior cerebral artery and which correlated significantly with the severity of motor deficit and the presence of aphasia, neglect, or sensory loss. These characteristics are similar to those in the present patient. However, "hypersensation" as found in this case was not described. Regarding the mechanism of these behaviours, Ghika *et al* speculated that they represent the clinical expression of early plastic changes of brain maps and circuits after an acute lesion and probably an active process induced by disinhibition to establish new compensatory pathways.[1] Such ipsilateral symptoms might occur not only in the motor system, but in the sensory system as well.[3] In the present patient, the degree of right hypersensation



Brain CT showing an infarction in the right precentral and postcentral gyrus extensively extending to the medial aspect of the frontal lobe.

was parallel with the degree of the disturbance of sensory deficits of the homologous left side, and hypersensation subsided as the sensory disturbance of the left side improved. This suggests that the disinhibition or hyperexcitability to facilitate functional reorganisation may have been the main cause of hypersensation in this case.

Lesional extent must also be considered. Studies in animals and patients with stroke with sensorimotor cortical lesion provided several insights into the basis for recovery. In the cortical region, there are three areas where increased activation has been suggested: the sensorimotor cortex of the unaffected hemisphere, the supplementary motor area (probably bilateral, ipsilateral much greater than contralateral to the lesion), and peri-infarct lesion of affected hemisphere.[4–6] In the present case, the right supplementary motor area belonged to the lesion and the right sensorimotor cortex was extensively involved. Acute onset of severe motor and sensory disturbance caused rapid disinhibition and increased activation which had to depend exclusively on the left (unaffected) sensorimotor cortex as the right supplementary motor area and right peri-infarct area could not be involved in the reorganisation process. I speculate that this provoked hyperkinetic motor behaviour as well as hypersensation in the right hemibody.

In the case of patients who recovered, there have been few reports of subjective perceptions in the acute stage of stroke.[7] Grotta *et al*[7] reported the subjective experiences of 24 patients with nonlacunar ischaemic stroke who dramatically recovered. They found that most patients did not recollect the severity of their problem and did not remember important events during the first 24 hours regardless of the side of the lesion. However, as most patients (19 of 24) could clearly recall the exact circumstances involving the onset of their stroke, they speculated that their unawareness of deficit was a form of anosognosia rather than a deficit of memory or global neurological function. The subjective experience of the patient in this study corresponds well with these findings.

HIDEAKI TEI
Department of Neurology, Toda Central General Hospital, 1–19–3 Hon-cho, Toda City 335–0023, Saitama, Japan

Correspondence to: Dr Hideaki Tei
webmaster@chuobyoin.or.jp

1 Ghika J, Bogousslavsky J, van Melle G, *et al*. Hyperkinetic motor behaviors contralateral to hemiplegia in acute stroke. *Eur Neurol* 1995;**35**:27–32.
2 Chang GY, Spokoyny E. Hyperkinetic motor behavior in bihemispheric stroke. *Eur Neurol* 1996;**36**:247.
3 Kim JS. Delayed-onset ipsilateral sensory symptoms in patients with central poststorke pain. *Eur Neurol* 1998;**40**:201–6.
4 Cramer SC, Nelles G, Benson RR, *et al*. A functional MRI study of subjects recovered from hemiparetic stroke. *Stroke* 1997;**28**:2518–27.
5 Pantano P, Formisano R, Ricci M, *et al*. Motor recovery after stroke. morphological and functional brain alterations. *Brain* 1996;**119**:1849–57.
6 Buchkremer-Ratzmann I, August M, Hagemann G, *et al*. Electrophysiological transcortical diaschisis after cortical photothrombosis in rat brain. *Stroke* 1996;**27**:1105–11.
7 Grotta J, Bratina P. Subjective experiences of 24 patients dramatically recovering from stroke. *Stroke* 1995;**26**:1285–8.

Phantom limb sensations after complete thoracic transverse myelitis

Phantom phenomena are common complications of limb amputations and may occasionally follow traumatic paraplegia and severe injuries of peripheral nerves. However, they have not been previously reported in patients with non-traumatic paraplegia. The following case history describes a patient with transverse myelitis resulting in complete paraplegia who experienced persistent movements and abnormal positions of her paralysed lower limbs. These findings suggest that disruption of the anatomical and functional integrity of the spinal cord may be the most important factor in the pathogenesis of phantom sensations.

A 61 year old woman presented with severe weakness of both legs, skin sensory loss and paraesthesia of the lower limbs, and bowel and bladder symptoms. She was well until 3 months earlier when she started to develop a tingling sensation and numbness over the outer side of her left leg. These symptoms gradually progressed and by the time she was admitted to hospital she had paraesthesia and sensory impairment of the whole of the left leg and in the distal half of the right leg. A month before admission she had become unsteady on her feet and developed urinary frequency, urgency of micturition, and constipation. There was also a rapidly progressive weakness of both legs, but no other symptoms.

Four years earlier the patient had had paraesthesia in both feet. This was thought to be due to peripheral neuropathy, but the diagnosis was not confirmed with neurophysiological tests. The symptoms resolved in a few weeks. The patient had a partial thyroidectomy for a nodular goitre 15 years ago. There was no other medical or family history of note. She was not taking any medication.

Physical examination confirmed the presence of complete flaccid paraplegia with skin sensory loss of all sensory modalities to the waist. The knee and ankle jerks were absent and both plantar responses were extensor. She had retention of urine and symptoms, signs, and radiological features of a paralytic ileus. The rest of the neurological and general physical examination was unremarkable. A full blood count, urea and electrolytes, and liver and thyroid function tests were within normal limits. An MRI of the cervical spine confirmed the presence of mild degenerative changes in the cervical spine at the level of C5-C7. There was no radiological evidence of an intrinsic or extrinsic cord compression or demyelination. However, the five distal segments of the thoracic cord appeared swollen and there was loss of the normal CSF rim ventral and dorsal to the cord on T1 weighted images. The T2 signal was prolonged and there was no contrast enhancement of the lesion. The appearances were considered consistent with oedema of the thoracic spinal cord. Brain MRI was normal. Visual evoked responses and brain stem auditory evoked potentials were within the normal limits. Somatosensory evoked potentials of the posterior tibial nerve could not be obtained because the patient developed severe myoclonic jerks of the entire leg at very low stimulus intensities. Her CFS protein concentration was 0.88 g/dl. No oligoclonal bands were detected on CSF protein electrophoresis. There were 2 lymphocytes/mm^3 and four polymorphs/mm^3. There was no bacterial growth on CFS culture.

Shortly after admission the patient started to experience phantom sensations in her lower limbs. At times she thought that her legs were crossed and on other occasions she felt that that she was standing on tiptoes. These symptoms were persistent and appeared to be spontaneous. Careful questioning did not disclose any specific stimuli. Their intensity remained unchanged until the patient was started on 200 mg carbamazepine three times a day. With this treatment the phantom sensations became less frequent and the images were less intense but they did not resolve completely. The paralytic ileus resolved with conservative treatment. However, the patient's neurological impairments remained unchanged until she was discharged from hospital 6 months later.

Non-painful phantom phenomena are continuous or intermittent sensations emanating from an amputated or deafferented part of the body. The missing or denervated part may be perceived in its premorbid shape, size, and other physical characteristics[1] or in a distorted form.[2] Patients often report normal functions associated with the absent organ—for example, penile erection, ejaculation, and orgasm after removal of the genitalia[3] or voluntary or involuntary movements of an amputated limb. Often, sensations such as touch, pressure, and cold are experienced in the phantom organ. Phantom sensations often occur after limb amputations[1] and have also been reported in about 15% of patients after a mastectomy.[4] Sometimes they may follow spinal cord injury.[5] However, their occurrence after transverse myelitis has not been previously reported.

Understanding the pathogenesis of phantom sensations is important for developing the appropriate treatment strategies. However, the mechanisms that underlie these phenomena are not fully understood at present. It has been suggested that they may be a manifestation of a psychological disorder or due to organic neurophysiological abnormalities.

Psychological factors such as denial or grief for the lost body part have been suggested as the cause of the postamputation phantom phenomena. However, this explanation is not supported by the currently available evidence. For example, the occurrence of phantom phenomena does not correlate with poor psychological adjustment or with the incidence of depressive symptoms in these patients.[6] Another hypothesis is that damaged peripheral somatosensory receptors fire spontaneously and give rise to the painful or abnormal experiences.[7] However, phantom sensations have been reported by patients after spinal anaesthesia in the absence of damage to the peripheral nervous system.[1] At present the neuromatrix theory[8] offers the most plausible explanation for phantom sensations and pain.

According to this theory the symptoms associated with the phantom phenomena originate from genetically predetermined sensory images (or sensory engrams) that are stored in the cerebral cortex. It was postulated that the sensory images are triggered when neural impulses from the periphery are blocked. The patient reported here had complete "functional" transection of the spinal cord. The occurrence of phantom sensations in this patient was therefore independent of the neural input from the peripheral nervous system. This case provides further evidence that phantom phenomena are due to a central neurophysiological mechanism, probably