

# Painful supernumerary phantom arm following motor cortex stimulation for central poststroke pain

## Case report

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✓ In this report, the authors describe a case in which the patient began to experience a supernumerary phantom arm after she received motor cortex stimulation for central pain. The patient had a history of right thalamocapsular stroke. It is speculated that the motor cortex activation triggered a response in the patient's parietal lobe, precipitating perception of the phantom limb.

To the authors' knowledge this is the first reported case of its kind.

**KEY WORDS • supernumerary phantom limb • cortical stimulation • single-photon emission computerized tomography • central pain**

**S**UPERNUMERARY phantom limbs (pseudopolymyelia) are seldom reported.<sup>1,7-10,13,14,16-23</sup> They indicate a neurological symptom of (generally) right-sided stroke, head or spinal cord injury, brain tumor, or multiple sclerosis.

We describe a woman who developed central post-stroke pain. Following motor cortex stimulation (MCS) performed to ease the pain, the patient began to experience a supernumerary arm hallucination. No similar case appears in the literature.

### Case Report

*History.* This 61-year-old woman had previously experienced hypertensive hemorrhage in the right side of her brain, which involved the thalamus, internal capsule, and deep subparietal corona radiata, and dissected into the ventricles (Fig. 1).

*Examination.* On admission 3 years later (1998), the patient's neurological examination disclosed a plegic left arm, a paretic left leg that hampered her gait, and tactile-pinprick anesthesia of the left side. The patient complained of a ripping, squeezing, and burning pain in the left half of her body, mostly occurring in the face, arm, and leg; allodynia was also present.

*Treatment.* The woman's pain was completely relieved when a subhypnotic propofol test was administered, but not when placebo was substituted (Class A/ $\gamma$ -aminobutyric acid-responsive central pain<sup>4</sup>). Her pain also was relieved when, according to our protocol,<sup>4</sup> extradural cortical

stimulation to the primary motor cortex was applied.<sup>3</sup> The ripping and squeezing pains were totally relieved and the burning pain was diminished by half. Unfortunately, the burning sensation could not be controlled further during a 5-week treatment period, despite the fact that parameter adjustments were made to the stimulator.

*Phantom Limb.* Approximately 6 weeks after implantation of the stimulator (May 1998), the patient spontaneously volunteered that she experienced the presence of a painful supernumerary left arm, which she unhesitatingly called "my phantom arm." This phantom limb was sensed once every 1 to 2 weeks. The experience lasted 10 to 15 minutes and sometimes longer; resolution of the pain was always abrupt and spontaneous. The stimulator was switched off because the patient obtained no more relief and the pain was worsened by changing the stimulator parameters into the high-voltage range. The patient's central pain syndrome relapsed. Some relief was later obtained using a course of high-dose mexiletine and gabapentin.

As part of our neurostimulation protocol, both neurometabolic and neuropsychological assessments (State Trait Anger Expression Inventory and Cognitive Behavioral Assessment [version 2.0]) were made. High-resolution single-photon emission computerized tomography, performed using hexamethylpropylenamine oxide and Iomazenil, revealed right thalamic and temporal hypoperfusion, which was normalized by the MCS. On examination, the woman appeared fully oriented, coherent, alert, and cooperative. She had no history of epilepsy or psychiatric

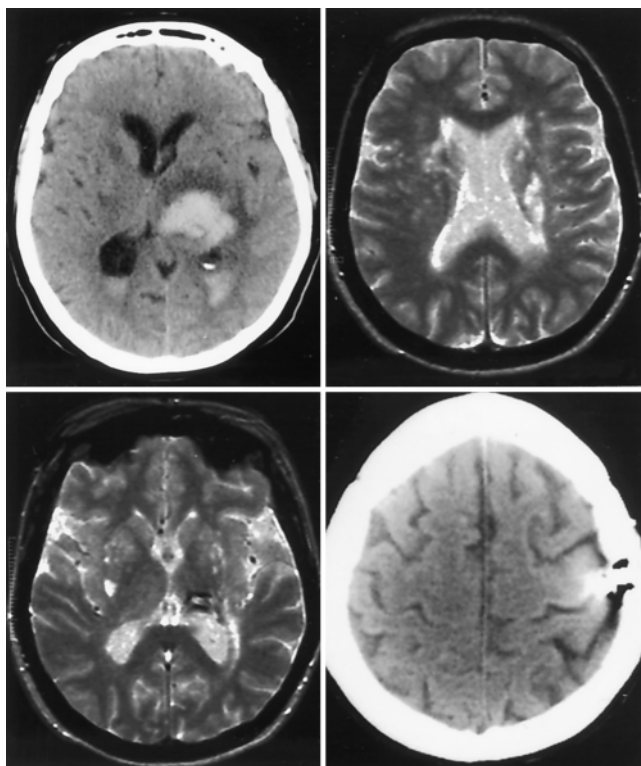


FIG. 1. Computerized tomography (*upper left*) and magnetic resonance (*upper right* and *lower left*) images demonstrating evidence of the patient's hemorrhagic stroke. Another computerized tomography scan revealing the motor cortex stimulator in place (*lower right*).

disorders, and she had never experienced hallucinations, including phantom sensations, prior to stimulation. Her cognition appeared normal. The autopathognosia was normal, and there was no anosognosia of her impaired limbs. In addition there was no anosodiaphoria, disorder of calculation, interpretative elaboration, delusion, or reduplication of people, places, events, confabulation, or right-left confusion. Instead, there was reactive depression, introversion, difficulty with social interaction, and high levels of inhibited aggression. She would shy away from people, being hypersensitive to criticism and external judgment. However, questions concerning her phantom limb did not cause her discomfort.

The phantom limb was described as originating from the shoulder. This phantom arm reportedly was flexed and externally rotated; it never moved spontaneously. The sensation of the phantom limb was always accompanied by pain having a crushing quality. When present, it made the patient's left arm pain worse. Touch or other stimulation could not evoke the phantom sensations. Instead, when the left arm was passively moved, the phantom limb moved also. Interestingly, on these occasions, the patient's left arm pain was made more tolerable. During the follow-up period (6 months), the patient could also feel her phantom arm stretched straight forward in front of her.

### Discussion

Supernumerary phantom appendages can be understood

in the general context of a central brain mechanism.<sup>2</sup> Ad hoc hypotheses range from psychological (resolution of cognitive dissonance, symbolic elaboration, delusional misidentification) to organic (sensory deprivation and perceptual neuronal completion). However, data do show that phantom pain is not a function of emotional adjustment.<sup>12</sup>

All cases of supernumerary phantom limb hallucinations are accompanied, to a variable degree, by both sensory and motor impairment. Although the importance of denervation is well recognized as far as phantom percepts are involved, this phenomenon alone cannot explain pseudopolyomyelia.<sup>22</sup> A recent study found that resolution of this reduplication syndrome was paired with an improvement in a patient's right-left laterality discrimination and other tasks of mental orientation in space, whereas the patient's sensory and motor functions did not significantly improve.<sup>22</sup> The fact that supernumerary phantom limbs are experienced more frequently after right-sided brain stroke<sup>14</sup> is closely akin to the higher proportion of central pain after right thalamic stroke<sup>5</sup>—the right hemisphere appears to have a role in monitoring somatic states and a dominant role in processing somatic representation (see references cited in recent correspondence<sup>9</sup>). Attentional networks may play an additional role.<sup>9</sup>

Halligan and colleagues<sup>13</sup> maintain that the emergence of supernumerary phantom limbs is determined more by motor loss than by sensory loss. The role of the motor system in this context is supported by the present case, in which MCS appears to have triggered the phantom sensations. We observed no effect of MCS on the patient's plegic arm, but her left foot seemed to "keep ahead of" her. In a functional magnetic resonance imaging study, Erslund and associates<sup>11</sup> reported activation of the primary motor cortex by phantom limb fingertapping. Kew and colleagues<sup>15</sup> found positron emission tomography evidence of an increased metabolism in both primary motor and sensory cortices in patients with postamputation phantom percepts, but not in congenitally limbless patients with no complaints of phantom sensations.<sup>15</sup> Those patients who can control the movements of their phantom limbs can sometimes reduce the associated pain.<sup>25</sup>

We speculate that MCS set off the oscillatory corticothalamic mechanism responsible for our patient's subjective awareness of her arm (see *Discussion* in another study<sup>2</sup>). This speculation hinges on the parietal lobe, including that portion responsible for sensorimotor integration and maintenance of the internal representation of the body state.<sup>24</sup> To generate stable percepts, the brain weighs evidence from many different sources. If a conflict is generated, a loop is activated and a perception of a supernumerary limb ensues. In this schema, illusory movements are explained by the cooperative action of both sensory and motor cortical areas. In theory, incorporation of this schema into the idea of Crick and Koch<sup>6</sup> that awareness is subtended by a link between primary sensory areas and motor areas may be possible.

### Conclusions

Supernumerary phantom limbs highlight brain mechanisms of consciousness. Their occurrence in the context of cortical stimulation for central pain supports the notion

## Phantom limb after cortical stimulation

that these anomalous phenomena are the result of an unbalanced information flow between the thalamus and cortex (dynamic reverberation<sup>2</sup>).

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