

BRIEF COMMUNICATION

Birth and death of a phantomSebastian Dieguez¹, Mélanie Kaeser², Camille Roux², Jérôme Cottet², Jean-Marie Annoni¹ & Eric Schmidlin²¹Laboratory for cognitive and neurological sciences, Department of medicine, University and Hospital of Fribourg, Fribourg, Switzerland²Laboratory of neurophysiology of action and hearing, Department of medicine, University and Hospital of Fribourg, Fribourg, Switzerland**Correspondence**

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Abstract

Patients with supernumerary phantom limb report experiencing an additional limb duplicating its physical counterpart, usually following a stroke with sensorimotor disturbances. Here, we report a short-lasting case of a right upper supernumerary phantom limb with unusual visuomotor features in a healthy participant during a pure Jacksonian motor seizure unexpectedly induced by continuous Theta-Burst Stimulation over the left primary motor cortex. Electromyographic correlates of the event followed the phenomenological pattern of sudden appearance and brutal dissolution of the phantom, adding credit to the hypothesis that supernumerary phantom limb results from a dynamic resolution of conflictual multimodal information.

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Introduction

Recent experimental advances have allowed a clearer understanding of the neural mechanisms underlying our sense of the bodily self, namely the integrated, unified, and dynamic experience of having a body.¹ Yet much of the accumulated knowledge on that topic has relied on clinical descriptions of neurological alterations of bodily awareness, which provided a rich and varied historical record of body misrepresentations such as illusory or delusional feelings of fragmentation, disappearance, distortion, duplication, or alienation of body parts.^{2–4} Collectively, such conditions entail the potential to unravel the neural and cognitive mechanisms underlying normal bodily awareness, but because they can be quite rare and unique, it remains important to pursue the careful cataloguing of their phenomenological presentations.

Supernumerary phantom limb (SPL) – “the awareness of having an « extra limb » in addition to the regular set of two arms and two legs”⁵ – is one striking example of such neurological altered states of bodily awareness. Contrary to phantom limbs of the amputee or movement and

postural illusions during sensorimotor impairment or cortical stimulation,^{6–8} SPLs entail a *concomitant* awareness of having an “extra” (illusory) limb and its physical counterpart (usually a plegic limb). The phenomenon has been observed following parietal strokes (predominantly in the right hemisphere)⁹ with severe sensorimotor impairment,¹⁰ as well as subcortical lesions,¹¹ spinal cord injury,¹² callosal and premotor damage,¹³ and has also been associated with seizures.¹⁴ Further, it can occur as a delusional belief¹⁵ or in clear-minded patients,¹¹ and cases vary widely along phenomenological characteristics such as motor and sensory features, sensitivity to visual and tactile feedback, or their experienced duration.⁴ It has been suggested that SPL phenomena result from multimodal conflict resolution at the phenomenological level.¹¹

Here, we report a new case of SPL, the striking features of which complement and shed new light on the diversity of the phenomenon. It presented as a short-lasting but vivid and terrifying experience of suddenly having, moving, feeling, and seeing an additional right arm, and of witnessing its brutal dislocation right before loss of consciousness, all part of a seizure starting in the left motor

cortex induced by continuous theta-burst stimulation (cTBS).

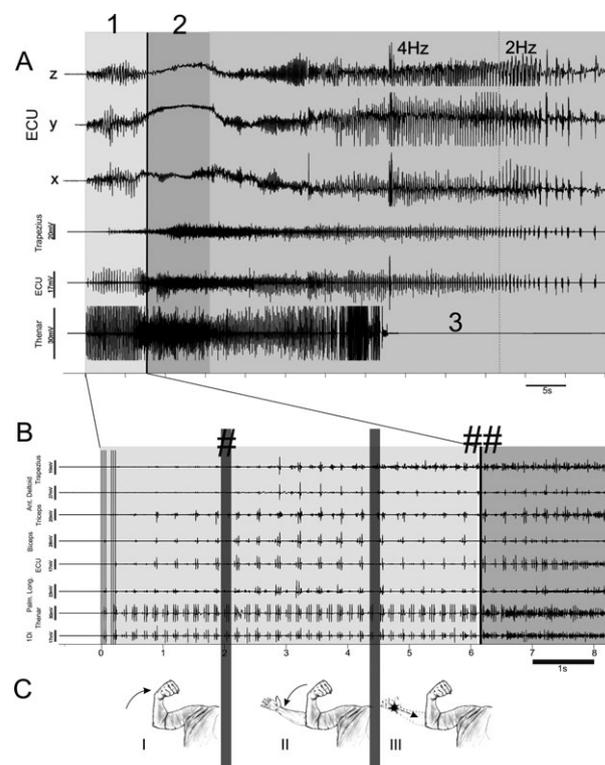
Case Report

The subject of this experience was a 24-year-old right-handed Caucasian male, a master student in neuroscience who volunteered, as a healthy control participant, in an ongoing study of upper-limb motor control after cortical inhibition through cTBS over the motor hand area in

M1. In what follows, we describe the participant's report of a SPL during an unexpected seizure shortly after stimulation began (Fig. 1A and B; see Data S1 for details on the procedure and clinical investigations). Full consent for the experiment and the current report has been obtained according to the guidelines of our local ethics committee. At the time of the event, the participant knew only about phantom pain in the amputee and never heard of SPL phenomena, having taken a biological- rather than clinical-oriented cursus.

Early during the course of clinical investigations, about one hour after the seizure and loss of consciousness, the fully recovered participant mentioned clear memories of a paroxysmal experience of having an additional arm, right prior to loss of consciousness. In a complementary recorded interview (see Data S1), he explained that shortly after stimulation onset, his right arm involuntarily started shaking and flexing toward his face, after which he immediately felt and saw an additional "virtual arm" (his term) emerging and extending (as he put it, "excorporating") from the elbow of his physical right arm toward its original position (Fig. 1C). As far as he could tell, the right "virtual arm" looked no different than his right physical arm. Then, he saw it "peeling like a banana" at the level of fingers and subsequently "crushing" along the arm toward the right shoulder, at which point he had the feeling that he was "losing his arm" and "about to die", and then lost consciousness. Asked for

Figure 1. Electromyographic and phenomenological phantom correlates of an unexpected cTBS-induced seizure in a 24-year-old experimental participant. (A) Recordings of tri-dimensional acceleration (z , y , and x) acquired from the electrode located at the surface of the extensor carpi ulnaris muscle (ECU) during the entire event, starting at the elicitation of the seizure, showing a clonic phase (1) followed by a tonic phase (2) and of the electromyographic activity of three hand and arm muscles: ECU, Trapezius and Thenar (3 corresponds to the loss of contact of the electrode due to excessive movement). (B) Detailed electromyographic recordings from eight arm and hand muscles (surface skin recordings: Trapezius, Anterior part of Deltoid, Triceps, Biceps, Extensor carpi ulnaris, Palmaris longus, Thenar and one dorsal interosseus (1DI)), obtained during the onset of epileptic seizure, showing a disto-proximal spread of EMG clonic activity with rapid shaking movements related to the theta-burst stimulation (sets of three vertical bars) from the intrinsic hand muscles, (1DI and Thenar), to extrinsic hand muscles (ECU, Palmaris Longus) to proximal muscles of arm and shoulder. Retrospective visual inspection of the continuous EMG recordings revealed that the tonic seizure started 6 sec after cTBS onset and that the clonic seizure (as defined by rhythmic, involuntary contractions of the 8 muscles) began 22 sec after cTBS onset. The seizure was characterized by an initial partial clonic spread of Bravais Jacksonian type during the first 6 sec after cTBS onset, followed by a tonic phase between 6 and 22 sec, followed by a clonic phase which self-terminated after around 60–70 sec. At the end of the clonic phase, frequency of myoclonic movements decreased from 4 to 2 Hz. Temporal unfolding analysis of EMG activity showed a spread of recruitment of muscles starting a distal level (intrinsic hand muscles) heading to more proximal muscles, with a pattern of activity closely related to the cTBS. The spread of transformation from clonic to tonic activity involved the muscles in the same order. EMG acquisitions (5000 Hz, low- and high-pass band filtered) were obtained and analyzed using the Spike 2 software (CED). #: probable time when the coil is moved away from the scalp, stopping external stimulation of the primary motor cortex. Note the identical pattern of EMG recordings matching the pattern of theta-burst stimulation even after the coil was removed from the scalp. ##: probable time of loss of consciousness. Amplitude of EMG activity: scale bar: 1mv; latency: scale bar: 1 sec. (C) Participant's depiction and approximate timing of the experienced supernumerary phantom limb (SPL). Three phases of the phenomenon are schematic represented: (1) initial flexion at the elbow of the physical right arm, (2) "apparition" of a right SPL; (3) "disintegration" of the SPL along the disto-proximal axis. Star corresponds to the time when the subject felt a sensation of oncoming "arm loss" and imminent death, just before passing out.



more details, he insisted that he was able during the short episode to feel and see both the physical and “virtual” arms and hands at the same time, without confusion between them. Whereas he could see the “virtual arm” disaggregating in the most gruesome way – skin, flesh, and bones “peeling”, “crushing”, and “exploding” as though the limb was passed through a “mechanical press” or a “harvester” – the physical limb, it was clear to him, remained intact during the whole sequence. Although terrifying and vivid, the experience was not accompanied by pain. Rather, it involved a strong sense of impending death and the sensation that he was “losing” his arm. Yet, the participant is not sure he felt the experience as “real”. In his recollection of the event, he reports some perplexity about what he was seeing and a general anxiety as to what was going on, but not specifically that he *believed* he had three arms and that one of them was *really* “peeling” and “exploding” in front of him. Of note, he remembers having had no sense of agency whatsoever regarding this experience, which was thus passively experienced, and he had no doubt as to the ownership of both physical and virtual limbs, which were felt to belong to him at all times. No other body parts were duplicated or otherwise involved in the incident. Because the experience was of short duration and strongly emotional, however, he was unable to report whether he could have voluntarily moved the physical and “virtual” limb independently or at all, and whether other body parts or surrounding objects and people were visually distorted.

Discussion

To our knowledge, this is the first report of SPL due to cTBS-induced tonico-clonic seizure (and only the second account of cTBS-induced seizure in a healthy participant¹⁶). As argued by Khateb et al.,¹¹ the sheer diversity of SPL phenomena discourages the search for a single explanation. It seems rather that several types of cortical or subcortical disturbances, accompanied by varying degrees of sensorimotor symptoms, will lead to idiosyncratic SPL features. Our account of the present case can only be based on a comparative assessment of the EMG data and the retrospective subjective report of the experience. We suggest that excessive external activation of the motor arm area induced a conflict between visuomotor and proprioceptive information that was dynamically “resolved” by an illusory visual correlate of arm position. As motor arm area activation abnormally and rapidly spread, it provoked an involuntary and unpredicted flexing of the physical arm which was resisted with a voluntary extending movement. This created a duplicate visual arm-template reflecting the willed and expected (initial) arm position, a dramatic and transient “solution” for

conflicting and confusing motor commands. Thus, this conflicting experience not only survived immediate visual feedback, but it created a visual correlate congruent with the experience (namely, of having the same arm doing two antagonistic movements at the same time and at two different places). The participant’s account indeed clearly indicated that the physical arm *first* flexed toward his face, and that only *then* the SPL appeared and seemed to extend toward the supine position. Nonetheless, with increasing spreading seizure-activity, this impossible “solution” gave way to the illusory, yet vivid, sense that the arm-template was suffering too much pressure to remain in the initial posture and thus began to (distally to proximally) “crush” as a direct somatotopic visual correlate of the uncontrollable motor activation.

Motor, proprioceptive and visual information of arm position normally converge and are supported by a common coding, explaining for instance that one’s active arm movements seem to be “visible” in complete darkness,¹⁷ that damage to the premotor cortex can lead to the visual disappearance of a hand,¹⁸ and that experimentally induced multimodal conflicts can create illusory ownership of virtual or artificial limbs.¹⁹ Moreover, illusions induced by tendon vibration at the biceps, especially when a feeling of contraction is willfully resisted, can induce the feeling that the arm is breaking, bending, curving or, indeed, create “double or multiple images” of the forearm.²⁰ We suggest that a similar but much stronger and dynamic conflict was unexpectedly induced in our participant, although its exact mechanisms remain unknown.

The present case uniquely combines several features rarely reported in the literature. It is the fifth reported case of SPL with a paroxysmic etiology,¹⁴ and the first in an experimental TMS context with concomitant EMG recordings in a healthy person. As such, it provides a unique view on the genesis of SPL phenomena: for the first time, the very moment of SPL emergence could be pinpointed, revealing that SPL awareness can be almost sudden, unrelated to structural plasticity, very short-lasting (about 4 sec from birth to disappearance) and yet extraordinarily vivid. Left cortical lateralization is also etiologically rare for SPL phenomena.⁹ Visible SPLs are likewise unusual, this is the tenth such case¹¹ and the first as part of a seizure. The illusory and dramatic visual destruction of a SPL had also never been reported before, although a sense of impending death is sometimes associated with seizures.²¹ Finally, a sufficient causal involvement of the primary motor cortex in SPL is here demonstrated for the first time.

Taken together, these observations highlight the diversity and complexity of SPL phenomena and reveal the manifold underlying miscalibrations that can give rise to

complex and vivid bodily illusions, however suddenly and fleetingly as in the present case.

Author Contribution

ES designed the initial study and collected the EMG data; CR and MK performed the rTMS stimulation; JMA performed the clinical exam; SD wrote the manuscript and interpreted the data; JC is the study participant and helped with manuscript and figures revisions. All the authors approved the final version of the manuscript.

Conflict of Interests

The authors declare no competing financial interests.

References

1. Blanke O. Multisensory brain mechanisms of bodily self-consciousness. *Nat Rev Neurosci* 2012;13:556–571.
2. Hécaen H, Ajuriaguerra J. *Méconnaissances et Hallucinations Corporelles*. Paris: Masson, 1952.
3. Vallar G, Ronchi R. Somatoparaphrenia: a body delusion. *Exp Brain Res* 2009;192:533–551.
4. Dieguez S, Annoni JM. Asomatognosia: disorders of the bodily self. In: Godefroy O, ed. *Vascular cognitive impairment and dementia: the behavioral and cognitive neurology of stroke*, 2nd ed. Cambridge: Cambridge University Press, 2013:170–192.
5. Brugger P. From phantom limb to phantom body. In: Knoblich G, Thornton IM, Grosjean M, Shiffrar M, eds. *Human body perception from the inside out*. Oxford: Oxford University Press, 2006:171–209.
6. Giummarra MJ, Georgiou-Karistianis N, Nicholls ME, et al. Corporeal awareness and proprioceptive sense of the phantom. *Br J Psychol* 2010;101:791–808.
7. Desmurget M, Reilly KT, Richard N, et al. Movement intention after parietal cortex stimulation in humans. *Science* 2009;324:811–813.
8. Bestmann S, Oliviero A, Voss M, et al. Cortical correlates of TMS-induced phantom hand movements revealed with concurrent TMS fMRI. *Neuropsychologia* 2006;44:2959–2971.
9. Miyazawa N, Hayashi M, Komiya K, Akiyama I. Supernumerary phantom limbs associated with left hemispheric stroke: case report and review of the literature. *Neurosurgery* 2006;54:228–231.
10. Antonello D, Kluger BM, Sahlein DH, Heilman KM. Phantom limb after stroke: an underreported phenomenon. *Cortex* 2009;46:1114–1122.
11. Khateb A, Simon SR, Dieguez S, et al. Seeing the phantom: a functional magnetic resonance imaging study of a supernumerary phantom limb. *Ann Neurol* 2009;65:698–705.
12. Curt A, Ngo Yengue C, Hilti LM, Brugger P. Supernumerary phantom limbs in spinal cord injury. *Spinal Cord* 2011;49:588–595.
13. Hari R, Hänninen R, Mäkinen T, et al. Three hands: fragmentation of human bodily awareness. *Neurosci Lett* 1998;240:131–134.
14. Millonig A, Bodner T, Donnemiller E, et al. Supernumerary phantom limb as a rare symptom of epileptic seizures – case report and literature review. *Epilepsia* 2011;52:e97–e100.
15. Weinstein EA, Kahn RL, Malitz S, Rozanski J. Delusional reduplication of parts of the body. *Brain* 1954;77:45–60.
16. Oberman LM, Pacual-Leone A. Report of seizure induced by continuous theta burst stimulation. *Brain Stimul* 2009;2:246–247.
17. Dieter KC, Hu B, Knill DC, et al. Kinesthesia can make an invisible hand visible. *Psychol Sci* 2014;25:66–75.
18. Arzy S, Overney LS, Landis T, et al. Neural mechanisms of embodiment: asomatognosia due to premotor cortex damage. *Arch Neurol* 2006;63:1022–1025.
19. Ehrsson HH. How many arms make a pair? Perceptual illusion of having an additional arm. *Perception* 2009;38:310–312.
20. Craske B. Perception of impossible limb positions induced by tendon vibration. *Science* 1977;196:71–73.
21. Greenberg D, Hochberg F, Murray G. The theme of death in complex partial seizures. *Am J Psychiatry* 1986;141:1587–1589.

Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article:

Data S1. (1) rTMS protocol; (2) seizure episode; (3) interview transcription.