

## Short communication

## Capsular warning syndrome mimicking a jacksonian sensory march

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## ABSTRACT

A 57-year-old man, operated eight years before for a left frontal falx meningioma, presented with short lasting, stereotyped episodes of paresthesias ascending from the right foot to the hand. A diagnosis of somatosensory seizures with jacksonian march was made. The patient was given antiepileptics but 5 days later, a few hours after another paresthetic episodes, he developed right hemiplegia, hemianesthesia and dysarthria due to an infarct of left capsular posterior limb. We deem that in this patient the paresthetic episodes were more likely an expression of a capsular warning syndrome than of parietal epilepsy because of the frontal localization of the surgical lesion, the absence of motor components in all episodes, the negativity of repeated EEG, and the lack of recurrences after stroke. In capsular warning syndrome sensory symptoms mimicking a jacksonian march can be due to ischemic depolarization progressively recruiting the somatotopically arranged sensory fibers in the posterior capsular limb.

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## 1. Introduction

Pure sensory transient ischemic attacks (TIAs) presenting with tingling episodes are rare [1,2]. On the other hand, isolated paresthesias are quite characteristic of somatosensory seizures of parietal lobe epilepsy [3]. We report an intriguing patient in whom a cluster of paresthetic episodes with a jacksonian march pattern was thought to be secondary to and operated frontal meningioma since when he developed a capsular infarct.

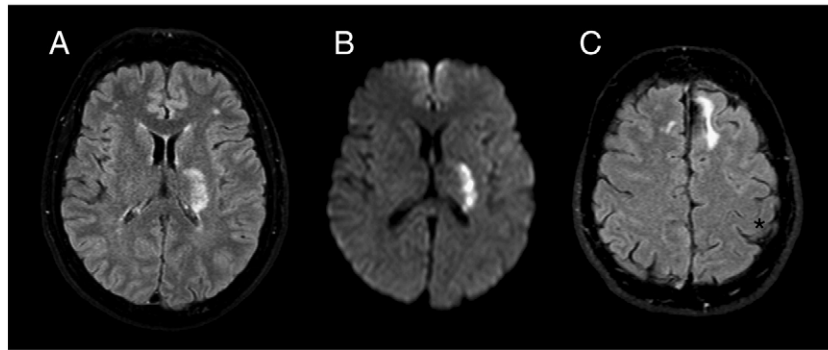
## 2. Case report

A 57-year-old, right-handed man, with no vascular risk factors, acutely developed tingling and numbness beginning in the right foot and ascending to the leg, thigh, trunk, arm and hand. The symptoms lasted few minutes and rapidly resolved with complete recovery. During the following 6 h the patient experienced four similar stereotyped episodes lasting maximum 5 min with a complete recovery between each event. At the arrival in the emergency room the neurological examination was normal. Blood pressure was 120/80 with a regular pulse of 74 beats per minute. History revealed that 8 years before the patient had 2 generalized tonic-clonic seizures secondary to a parasagittal falx meningioma compressing the left frontal lobe. He was treated with oxcarbazepine (1200 mg/day) and the meningioma was surgically removed without any sequelae. After 2 years oxcarbazepine was interrupted without the recurrence of seizures. In the emergency

room, while lying on a couch, the patient had a further episode of tingling and numbness ascending from the right foot to the hand. Speech and comprehension were normal. Symptoms lasted 3 min and rapidly resolved. Cranial CT scan revealed a left anterior frontal gliotic and proencephalic outcome of the previous surgery without signs of recurrence of the meningioma. Interictal EEG was normal. Laboratory testing, including serum triglyceride, total cholesterol, VLDL-cholesterol level, hemochrome and leukocyte count, coagulation screening, sedimentation rate, electrolytes and renal function, urine sample and gas analysis were normal. ECG, chest X-ray and color doppler ultrasonography evaluation of major vessels in the neck were normal. Somatosensory seizures with jacksonian march were hypothesized, lorazepam 4 mg i.v was given and oxcarbazepine restarted with disappearance of the episodes. The patient was discharged but five days later, in the morning, he presented again an episode of ascending paresthesias and numbness which completely recovered and in the afternoon he acutely developed weakness of right arm and leg. Examination, 6h later, showed hemiplegia of the right face, arm and leg, severe dysarthria and right hemianaesthesia for all sensory modalities. Cognitive and visual functions were normal. Blood pressure was 100/70 with regular pulse of 68 beats per minute. Two days later MRI was done with a 1.5 Tesla system. Images revealed ischemic changes involving the posterior limb of the left internal capsule and the posterior paraventricular corona radiata (Fig. 1). Source images of 3D Time of Flight MR-angiography sequences showed a very thin and irregular left anterior choroidal artery (AChA); the rest of the intracranial arterial circle was unremarkable. Repeated EEG were normal. Transthoracic echocardiography, antinuclear and antiphospholipid antibodies, lupus anticoagulant, protein C and S, antithrombin III, homocysteine, were all

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**Fig. 1.** MRI study. A) Axial Spectral Attenuated Inversion Recovery images, demonstrating hyperintense ischemic lesion involving the posterior limb of the left internal capsule and posterior paraventricular corona radiata; B) corresponding regions of restricted diffusivity on diffusion weighted images demonstrating the acute stage of the ischemia; C) left frontal gliotic and porencephalic surgical outcome distant from the somatosensory strip (asterisk).

within normal limits. ASA at 300 mg die was started and the patient was discharged 3 weeks after admission with minimal right hand weakness and complete recovery of the lower limb. Oxcarbazepine was discontinued and the patient did not have additional neurologic events during the following 12 months.

### 3. Discussion

The patient we report, with a history of generalized seizures which stopped after removal of a left parasagittal frontal meningioma, presented a cluster of short lasting, stereotyped episodes of paresthesias ascending from the right foot to the hand with complete interictal recovery. Initial involvement of lower limb has been reported in paresthetic seizures secondary to parasagittal lesions and the ascending pattern was compatible with a progressive cortical spread of an epileptic discharge along the primary sensory cortex as in jacksonian sensory march [3]. On the basis of the history and the characteristics of the episodes a diagnosis of somatosensory seizures was made. The absence of epileptic features at the interictal EEG was not considered to rule out the diagnosis, as even ictal EEG may be normal in 85% of patients with parietal lobe epilepsy [4]. The patient was treated with antiepileptics with disappearance of the episodes but five days later he developed an infarction of the posterior limb of the internal capsule and posterior paraventricular corona radiata in the territory of the AchA [5]. Retrospectively an alternative diagnosis of capsular warning syndrome (CWS) was considered for the paresthetic episodes preceding the stroke. CWS is a burst of stereotyped TIAs, clustered within few days, with unilateral motor and/or sensory deficit involving at least 2 of 3 body parts (face, arm or leg) without cortical symptoms. CWS is associated with high early risk of lacunar infarction in the territory of single penetrating vessel although occasionally striatocapsular or AchA territories are involved [6].

The point is whether the patient we report had paresthetic seizures as a delayed sequela of the operated meningioma and additionally developed a capsular infarction or whether the paresthetic episodes represented the CWS of the infarct. Against the epileptic origin of the paresthetic episodes is that at MRI reappraisal the surgical outcome of the meningioma is far anterior from the primary sensory cortex (Fig. 1) and that all the episodes developed without motor components which should be expected from the spread of excitation from a silent frontal focus to the hypothesized eloquent postcentral gyrus. Moreover, repeated EEG resulted normal and the paresthetic episodes did not recur after the capsular infarct and discontinuation of antiepileptic treatment. On the other hand two out of 50 patients described by Donnan and colleagues with CWS syndrome had exclusively sensory symptoms before developing a capsular infarction although a jacksonian sensory march has never been described in CWS [6].

The posterior limb of internal capsule is vascularized by the distal branches AchA which usually originates from the internal carotid distal to the origin of the posterior communicating artery [7]. Using source images of 3D TOF sequences it is possible to recognize the AchA, but not its branches, in 95% of normal subjects at MR-angiography[8]. In its complete form the infarct of AchA includes hemiplegia, hemianaesthesia and homonymous hemianopsia contralateral to the lesion [5,9]. Pure sensory stroke due to infarction in AchA territory has also been reported [9,10]. The hypothesized pathophysiologic mechanism of capsular warning syndrome include artery-to-artery microembolism, hemodynamic events or vasospasm [6]. We deem that in the patient we report the most likely mechanism of the stereotyped short lasting episodes is intermittent hypoperfusion, on a hemodynamic basis, in the terminal territory of the stenotic AchA. Anoxia-ischemia it is known to induce, by inhibition of the electrogenic  $\text{Na}^+/\text{K}^+$  pump, a depolarization of myelinated fibers and in the peripheral nervous system this depolarization is responsible of ischemic paresthesias [11,12]. The jacksonian-like march may be explained by the somatotopic arrangement of sensory fibers in the posterior limb of capsule with from back to forth the foot, leg, thigh, trunk, shoulder, arm, forearm, hand, fingers, the face, tongue and pharynx [13]. An intermittent ischemia progressing from back to forth in the capsular posterior limb may recruit and depolarize the somatotopically arranged fibers accounting for the ascending progression of paresthesias mimicking a jacksonian march. When the distal branch of AchA eventually occluded the cascade of molecular events, triggered by ischemia, progressed to calcium-mediated injury and the infarct of all posterior limb of the capsule developed [11].

In light of all the above observations and according to the Occam's razor we deem that in the patient we report the paresthetic episodes with jacksonian sensory march represented a CWS of the capsular infarction.

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