

Alien hand syndrome and paroxysmic dystonia after right posterior cerebral artery territory infarction

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Received: 8 April 2015 / Accepted: 8 May 2015
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Alien hand syndrome (AHS) is a disorder of involuntary, yet purposeful hand movements, with the feeling that the hand is foreign to the patient or has a will of its own. Association with a focal dystonia is exceptionally reported. We report both syndromes after a right posterior cerebral artery territory infarction.

A 25-year-old right-handed man with past medical history of mood disorders was referred to our stroke unit due to the sudden onset of left-sided hemiparesis, left-sided hypoesthesia with proprioceptive dysfunction, and left homonymous hemianopsia. One day later, while the initial motor deficit was improving, he complained that his left hand was controlled by someone else. In addition, he also presented a painful mobile focal dystonia of his left side arm. He described it as a cramp-like feeling resulting in abnormal bending of his left elbow and fisting movements of his hand. These involuntary movements were aggravated by actions such as walking and writing, relieved by rest and sleep. Neuropsychological assessment failed to disclose any visuospatial dysgnosia, or left spatial or motor neglect.

There was no previous or family history of similar movement disorders, and no exposure to neuroleptic, anti-emetic or anti-depressive medications or cocaine use. AHS, dystonia and painful spasms were initially uncontrolled by clonazepam (5 mg twice daily), but cleared over 1 week under carbamazepine (200 mg twice daily).

Diffusion-weighted MRI imaging (Fig. 1) showed an acute ischemic lesion involving the right posterior cerebral artery territory including right thalamus (ventral posterior lateral nucleus) and the medial part of temporal lobe. Etiological stroke workup detected a patent foramen ovale and baseline antiphospholipid antibodies.

Focal thalamic lesions have been associated with a variety of involuntary movements such as tremor, dystonia, and chorea-ballism, but exceptionally with the occurrence of AHS. AHS is a rare condition described in anterior cerebral artery strokes, midline tumors, callosotomy and neurodegenerative disease. It is known to occur concomitantly with dystonia, particularly in cortico-basal degeneration but rarely in stroke [1].

There are three AHS variants: two anterior variants (frontal subtype, callosal subtype) [2] and one posterior variant [3]. The latter has been classically documented after thalamic infarction. Little is known about the underlying mechanisms of AHS and the relation between the anatomic substrate. The posterior AHS fits the model of a distorted body scheme, because of a disrupted integration of somatosensory input to cingulate region due to the thalamic infarct [4].

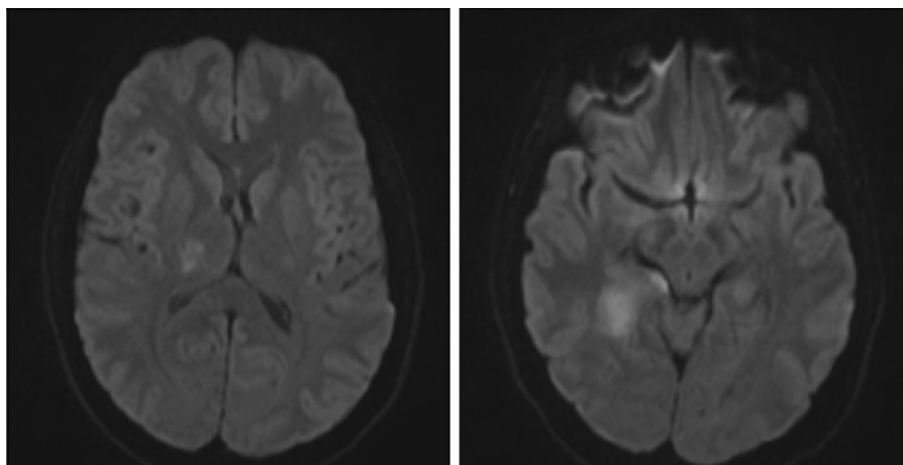
The pathological mechanisms of thalamic dystonia may involve an interruption/dysfunction of the cortico-striato-pallido-thalamo-cortical loop. The simultaneous emergence of both symptoms after a thalamic infarct reflects the role of the thalamus as a key relay in motor control [5].

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Fig. 1 Diffusion MRI shows infarction involving the right thalamus (ventral posterior lateral nucleus) and the medial part of temporal lobe



Conflict of interest The authors declare that they have no conflict of interest.

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