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Case resport

Dystonia after a bone fracture of the arm in a patient with a history of striato-pallidal ischemic stroke: a case report

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Abstract

We report on a female with segmental dystonia of the upper limb after an anatomically related ischemic stroke. Dystonia developed almost 3 years after the onset of the stroke and immediately following the removal of the cast because of a bone fracture in the same limb. We discuss the case considering issues such as: delay-onset, lesion topography and pathophysiology, peripheral input and their possible contribution to the development of secondary dystonia.

Keywords: Dystonia; Basal ganglia; Stroke; Peripheral injury

1. Introduction

Dystonia is a syndrome characterized by sustained and forceful muscle contractions, frequently causing twisting and repetitive movements, or abnormal postures. According to the etiological classification, dystonia can be divided into two major categories: idiopathic or primary and symptomatic or secondary [1]. Delayed-onset focal or segmental dystonia secondary to focal cerebral vascular injury, such as infarction of the basal ganglia is a rare but well-described entity [2–6]. Moreover selectivity of the lesions within the basal ganglia that result in dystonia is a matter of great interest because it may give clues to the functional organization of the lesions. Finally, there is a growing body of evidence that defective sensory inputs could play a crucial role in the development of dystonic dyskinesia [7]. Our case describes a patient with a history of right striato-pallidal ischemic stroke, who developed segmental dystonia of the left upper limb 2 months after a peripheral injury (bone fracture) of the left elbow and immediately following the removal of the plaster cast. Our purpose is to consider among others issues the following crucial question; was the onset of the movement disorder related to the peripheral trauma or could it have occurred by chance?

1.1. Case history

A 58-year-old right handed female was referred to a movement disorder outpatient clinic complaining of ‘pain and discomfort’ as well as marked involuntary movements restricted to the left hand. Three years before she had a stroke of the right basal ganglia territory (Brain CT scan: hypodense area at the anatomical position of the right putamen). resulting in a left hemiparesis involving mainly the upper limb, together with mild paralysis of the left lower face (asymmetry of the nasolabial fold). At the time the patient had no sensory complaints or defects. An NIH stroke scale performed immediately after the insult showed only minor motor impairment [score 1 for ‘facial facial palsy’ (item 4) and score 1 for ‘motor arm and leg’ (item 5 and 6)] and the neuropsychological evaluation revealed some degree of apathy and loss of drive. Apart from mild hypertension for the previous 5 years, medical history was unremarkable and she had no family or personal history of psychiatric or movement disorders. Following the stroke she was seen every 6 months in the outpatient clinic, and by the end of the first year the patient had no residual weakness of the left hand.

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About two and a half years after the stroke she fractured left elbow and she was in a plaster cast for the next 2 months in order to immobilize the joint. Immediately after the removal of the cast at the end of 2 months, the patient experienced marked discomfort of the hand, which was not helped by physiotherapy. The discomfort 'forced' the patient to make continual 'irregular, mainly twisting, movements' of the hand mostly characterized by finger extension and abduction, or flexion of fingers III and IV in the metacarpophalangeal joints with extension of the other finger joints, pronation of the forearm, and to a lesser degree by internal rotation, as well as adduction of the arm. She also stated that when walking she had less discomfort when the limb 'goes' at behind her. Muscle strength in the hand was almost normal and the clinical examination did not reveal additional signs. The continuous writhing character of the movements added an athetotic component to them. There were no signs of autonomic dysfunction of the limb as seen in cases of complex regional pain syndrome (CRPS). There was no history of toxic exposure or treatment with neuroleptic drugs. The patient underwent a brain MRI (Fig. 1), which showed the old lesion of the right putamen and the globus pallidus with additional involvement to a minor degree of the right internal capsule and gliosis at the border zone around the lesion. The pattern of the dystonic movements did not change over a 2-year follow-up period.

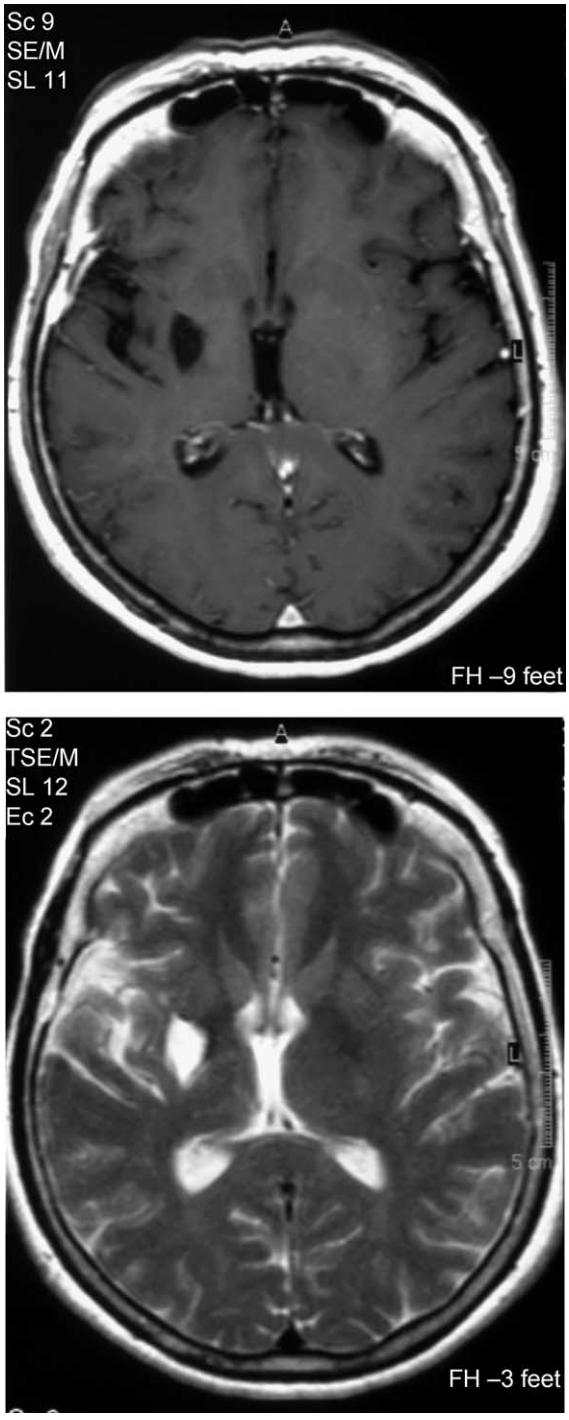

Fig. 1. Brain MRI of the patient. The lesion is visible as hypodense and hyperdense area on T1 and T2 weighted images, respectively, involving the posterior part of the right putamen and the globus pallidus.

2. Discussion

Delayed-onset dystonia is a rare sequel of stroke. Delayed-onset of a movement disorder after ischemic-hypoxic injury may reflect the time required for remyelination, inflammatory changes, ephaptic transmission, oxidation reactions, maturation or aberrant synaptic reorganization, trans-synaptic neuronal degeneration, or denervation supersensitivity [11].

The anatomical basis and pathogenesis of secondary dystonia are both uncertain. The involvement of the internal capsule together with the adjacent pallido-striatal complex in stroke patients with dystonia confined to the upper limb has been also demonstrated by others [5,6,13]. Moreover, the literature concerning the question of which structure among putamen and globus pallidus, internal (GPi) or external (GPe), is mainly responsible for the development of secondary dystonia, is conflicting and confusing. It has been suggested that putaminal lesions might affect both direct and indirect pathways [14]. This disruption may predominantly affect the indirect pathway resulting in dystonia by increased thalamocortical drive due to disinhibition of the thalamo-cortical projections [6,15]. On the other hand, there is indirect evidence in primates and humans showing that, at least in primary dystonia, both the direct and indirect pathways are overactive, and certain models of primary dystonia share this notion [15]. If this is a valid point, then secondary dystonias associated with putaminal lesions are difficult to explain based on striatal overactivity. The same difficulty exists in the case of involvement of the external globus pallidus (GPe) since lesions of GPe tend to produce parkinsonism by disinhibition of the subthalamic nucleus (STN) [17], and certain movement disorders like chorea