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LETTER TO THE EDITOR

Posterior spinal artery infarction initially presenting as acute bilateral lower limb dystonia

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Dear Editor:

Compared with cerebral infarction, spinal cord infarction, accounting for 0.3–1% of all strokes, is a rare condition [1]. Posterior spinal cord infarction, accounting for approximately 22% of all spinal cord infarctions, is rarer than anterior spinal cord infarction [1]. Impaired proprioception is a characteristic feature of posterior spinal cord infarction.

Dystonia, associated with impaired sensorimotor integration, is a heterogeneous group of hyperkinetic movement disorders characterized by involuntary sustained muscle contractions that lead to abnormal postures and repetitive movements [2]. The sensory aspects of dystonia include intrinsic sensory abnormalities and the effects of external sensory input on the underlying motor abnormality. The basal ganglia, cerebellum, thalamus, and their connections, coupled with altered sensory input, seem to play key roles in abnormal sensorimotor integration [3]. Both organic brain lesion and altered sensory input will result in dystonia.

Case report

A 56-year-old adult man with a past medical history of hypertension and smoking presented to the emergency room with an acute onset of rapidly progressive ascending tingling and numbness from the lower limbs to the neck (C2 dermatome) and bilateral lower limb twisting postures. When he tried to elevate his legs, the twisting postures of the bilateral lower limbs occurred. His ability to stand and walk was severely impaired due to the twisting postures. Neither herbal medication nor anti-dopamine agents were used. He was on Foley catheter due to impaired movement, but not sphincter dysfunction.

On examination, he was afebrile. The neurological examination revealed an intermittent dystonic posture of the bilateral lower limbs characterized by bilateral extension of the knee and inversion of the foot with flexion of the toes, which worsened by elevating the legs. When touching the lateral side of his right calf muscle, the twitching posture transiently disappeared. The dystonia persisted for 2 days; when it subsided, weakness (muscle strength scale of 4) appeared over the bilateral upper and lower extremities. Deep tendon reflexes over the lower limbs were pathological hyperreflexia with bilateral ankle clonus and plantar dorsiflexion. Proprioceptive hypoesthesia was found below the C2 dermatome, especially for the lower limbs. There were no concurrent cerebellar signs, such as hypotonia, dysdiadokokinesia, and dysmetria. The initial laboratory evaluation, including inflammatory markers, was all within normal limits. Magnetic resonance (MR) imaging of the cervical spine demonstrated high diffusion-weighted image (DWI) signal in the posterior aspect of the upper cervical spinal cord from the C2 to the C7 vertebral level, and normal T2- and T1-weighted signals in the corresponding location (Fig. 1). Somatosensory evoked potential (SSEP) disclosed delayed inter-peak latency between the right L1 cortex and right N15-P37, which indicated a central conduction defect between the right lumbar cord and cortex. Cerebrospinal fluid (CSF) profiles were within normal limits. Based on the clinical features and the positive

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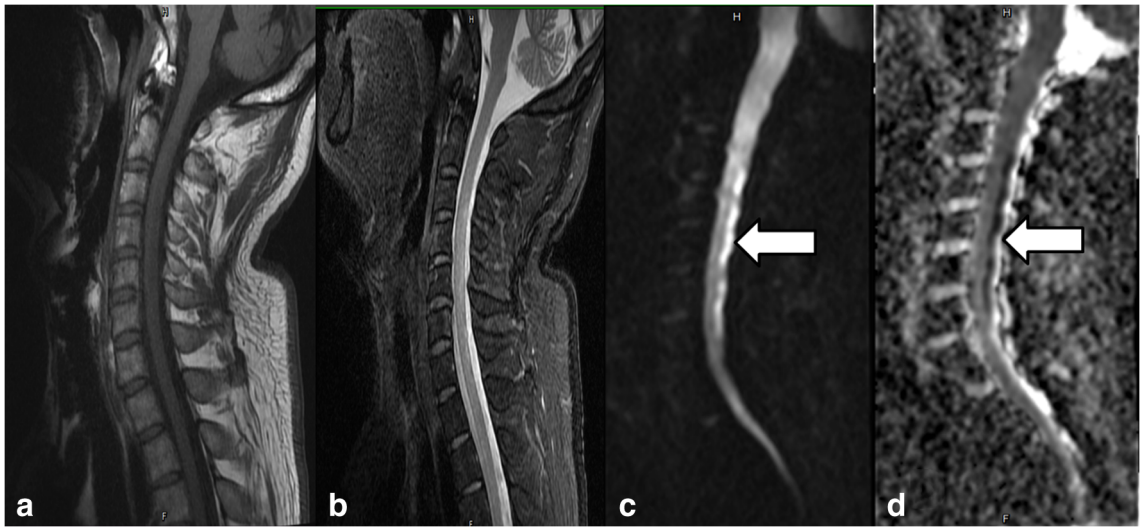

Fig. 1 There are hyper-signal intensity on DW imaging (**c**, arrow), hypo-signal intensity on ADC imaging (**d**, arrow), and normal signal in the corresponding area on T1 MR (**a**) and T2 MR (**b**) imaging

DWI MR image, spinal cord infarction was highly suspected. The patient was administered aspirin 100 mg once per day. The bilateral lower limb dystonia subsided gradually 2 days after the onset, but the impaired proprioception remained. He reported that the muscle strength of his legs returned to normal, but he could not control his legs appropriately and correctly. It is hard to completely exclude the possibility of transverse myelitis totally on clinical grounds, although the CSF profile was within normal limits. He was treated with intravenous (IV) methylprednisolone (1 g daily) for 5 days starting on the seventh day after the onset symptoms, without a notable improvement. After 1 month of rehabilitation, he could walk independently and freely.

Discussion

For adults, idiopathic focal dystonia usually affects the upper limbs or craniocervical regions, but not the lower limbs. The occurrence of lower limb dystonia in adults is often associated with a secondary etiology, such as parkinsonism, trauma, and cerebral stroke [4]. Posterior spinal cord infarction is rarely reported. Sometimes, upper limb dystonia can be due to cervical spinal cord disease, such as syringomyelia, or tumors, with impaired proprioception [5]. The pathophysiology is explained by a reduction in ipsilateral reciprocal inhibition due to interneuron dysfunctions responsible for the inhibition at the spinal level, which is regulated by the cortical control and the peripheral input from Ia afferents [6, 7]; this could also explain the pathophysiology of dystonia in our case.

Although dystonia was traditionally classified as a basal ganglia disease, dystonia is now considered a network disorder, including not only cortico-basal ganglia-thalamo-cortical, but also cortico-ponto-cerebello-thalamo-cortical circuits. The role of the cerebellum is perceived as more and more important [8, 9]. Even in the absence of traditional "cerebellar signs" in most dystonia patients, there are more subtle indications of cerebellar dysfunction and imaging correlates [9]. One possibility is that the dystonia of our case, although without obvious cerebellar dysfunction, may subsequently result from dysfunction of spinocerebellar and then the cerebello-thalamo-striatal pathway.

Unlike those of cerebral infarction, the exact causes and risk factors associated with spinal cord infarction remain unclear because of its rarity [10]. Most spinal cord infarction (88%) occurs spontaneously, but some occurs in the special clinical setting, such as aortic surgery, aneurysm, and dissection [11]. Patients with posterior spinal cord infarction classically present unilateral loss of proprioception below the level of injury. Weakness has been described in some cases, but is typically mild and transient. Compared with lower spinal cord (mid-thoracic cord and conus) infarction, upper spinal cord (cervical) infarction is rare but with better prognosis because of its better collateral circulation [11]; this can explain the transient and benign course of acute lower limb dystonia in our case.

In conclusion, acute bilateral lower limb dystonia can be the initial presentation of posterior spinal cord infarction at the cervical segment. Bilateral lower limb dystonia is a novel phenomenon of the posterior spinal cord infarction. Diffusion-weighted MRI is essential for the diagnosis of acute spinal cord syndrome.

Compliance with ethical standards

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

Ethical compliance statement The authors confirm that the approval of an institutional review board was not required for this work. We confirm that we have read the journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

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