Movement Disorders
CLINICAL PRACTICE
CASE REPORT

Re-emergent Tremor Without Rest Tremor After Lenticular Infarctions

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Moyamoya disease is characterized by progressive occlusion of the internal carotid artery and the main branches of the circle of Willis. Up to 6% of patients with moyamoya disease may develop chorea, dystonia, or dyskinesia.[1] We present a patient with moyamoya disease who developed re-emergent action tremors after infarctions in the lenticular nucleus.

Case Report

A 43-yr-old man suddenly developed action tremor and mild motor weakness of the right arm. Motor weakness began to improve several hours later, but tremor persisted. He reported that he had never noticed rest tremor. On neurological examination, performed 2 days after onset, he had mild weakness in extension and abduction of the right fingers. He had no sensory or cerebellar deficits. His right finger tapping was slow, but there was no progressive decrement in amplitude or speed. There was no rigidity in the limbs, even on the Froment's maneuver. He showed no resting tremor while walking and counting backward from 100. When he stretched out his arms, a tremor of the right fingers emerged after a latency of several seconds. The tremor worsened over the ensuing several seconds and reached a plateau. It mainly involved the right thumb, with an amplitude of up to 3 cm. While he was drawing Archimedes' spiral, a similar pattern of kinetic tremor emerged. On the finger-to-nose test, there was a terminal tremor. Surface electromyography (EMG) studies at rest showed no muscle acitivities, but while he was holding his arms in front of him, there were alternating bursts of 5 Hz over the right flexor and extensor carpi radialis muscles (Fig. 1A). Brain MRI studies showed infarctions involving the left pallidum and posterior putamen (Fig. 1B). Cerebral MR angiography studies showed stenosis in both proximal middle cerebral arteries. Cerebral angiography studies confirmed stenosis in the right internal carotid artery and the left middle cerebral artery with extracranial collaterals (Fig. 1C). [^{18}F] FP-CIT PET studies showed a punch-out defect in the left posterior putamen (Fig. 1D). Amplitude of tremor was reduced by half after levodopa (daily dose of 250 mg) treatment.

Focal basal ganglia lesions frequently cause various movement disorders, including chorea, dystonia, jerky dystonic tremor, and akineto-rigid syndrome, but rarely isolated parkinsonian tremors.[2] Few cases of delayed onset resting hand tremors have been reported after infarctions in the striatum.[3]

Postural and kinetic tremors with no rest component do occur in Parkinson's disease (PD). Pathogenic origins of such tremors are uncertain. They have been considered as a variant

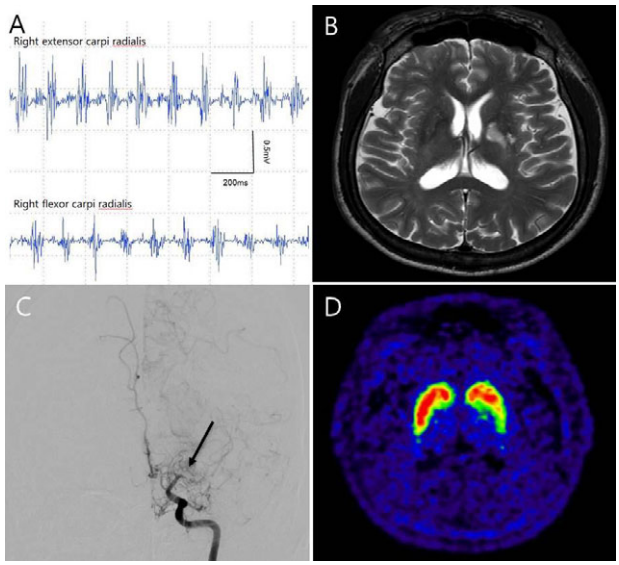

Figure 1 Surface electromyography studies, performed while arms are outstretched, reveal alternating 5-Hz bursts in the right flexor and extensor carpi radialis muscles. (B) T2-weighted axial brain MRI studies show infarctions in the left pallidum and posterior putamen. (C) Cerebral angiography studies reveal stenosis in the left middle cerebral artery (arrow) with well-developed collaterals, compatible with moyamoya disease. (D) [^{18}F]-FP-CIT PET studies show a punch-out defect in the left posterior putamen, corresponding to the area involved by the infarction.

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of essential tremor (ET), enhanced physiological tremor, or in cases of re-emergent tremor, as a form of parkinsonian rest tremor.[4,5]

An idiopathic PD (iPD) patient was reported on who had very similar tremors with our patient. The patient had 5.6- to 6.5-Hz postural and kinetic tremors with alternating antagonist contractions. The tremors emerged after a latency of several seconds. Because there was no rest tremor, the investigators referred to "emergent tremor." However, in that case, the diagnosis of iPD was confounded by past histories of psychiatric illness and lithium therapy, ongoing tremorgenic drug treatment, and no functional imaging studies on the nigrostriatal dopaminergic system.[6]

Re-emergent tremor is one of the characteristic feature of parkinsonian tremor. However, re-emergent tremor can be observed in ET[5] and scans without evidence of dopaminergic deficit.[7] Also, postural and intention tremors with alternating antagonist activities have been reported in patients with ET, multiple sclerosis, cerebellar degeneration, Wilson's disease, peripheral neuropathy, and head trauma.[8]

Our patient developed sudden-onset action tremors that were clearly associated with a lenticular infarction. The tremors shared characteristic phenotypes with a parkinsonian re-emergent tremor: (1) a latency of several seconds; (2) alternating agonist-antagonist contractions; (3) 5-Hz frequency; and (4) predominant thumb involvement.[4,5]

In our patient, lenticular infarction and striatal dopamine loss seemed to have caused rhythmic oscillations of basal ganglia and cortical motor outputs generated for movements or maintenance of posture of the arm. Alternatively, abnormal integration of kinesthetic sensory information arising from the contracted arm muscles at the putamen might cause rhythmic oscillations of basal ganglia and cortical motor outputs. This case suggests that neuronal mechanisms involved in the generation of emergent tremor can be functionally independent from those producing resting tremor.

Author Roles

(1) Research Project: A. Conception, B. Organization, C. Execution; (2) Statistical Analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript Preparation: A. Writing of the First Draft, B. Review and Critique.

M.S.L.: 1A, 1C, 3A

S.H.L.: 1C, 3B

S.J.A.: 1C, 3B

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Supporting Information

A video accompanying this article is available in the supporting information here.

Video 1. This video shows a 43-yr-old Korean man with postural re-emergent tremor of the right hand, mainly involving the thumb. The tremor emerges a latency of a few seconds and becomes worse over the ensuing several seconds and reached a plateau. While he is drawing Archimedes' spiral, a similar pattern of kinetic tremor emerges.

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