

A Lesion of the Anterior Thalamus Producing Dystonic Tremor of the Hand

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Background: Thalamic tremor is typically characterized by resting and intention components; a postural element is often present as well. Previously reported cases of acquired thalamic tremor have demonstrated lesions in the posterior thalamus or dentatorubrothalamic tract.

Objectives: To report a case of dystonic-postural tremor of the upper extremity that occurred after a contralateral anterior thalamic infarct, and to discuss potential tremorigenic mechanisms.

Design: Case report.

Setting: Municipal hospital neurology clinic.

Patient: A 65-year-old right-handed woman suddenly developed a dystonic tremor in her left hand after undergoing coronary bypass surgery. The tremor persisted unchanged for 8 months, at which time she was evaluated by us. Cranial magnetic resonance imaging scans demonstrated a right anterior thalamic infarct.

Conclusion: To our knowledge, this is the first report of focal tremor caused by a lesion of the anterior thalamus.

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TREMORS OF thalamic origin (TTOs) typically consist of both resting and intention components; a postural element is often present as well.¹ Other features of thalamic dysfunction, such as sensory loss, dystonia, pain, and hemianopia, are commonly associated with TTOs.¹ Identifiable causes of TTOs include thalamic infarction or hemorrhage, traumatic brain injury, infection, and neoplasm.¹⁻⁴ Previously reported cases of acquired TTOs have consistently demonstrated lesions of the posterior thalamus or dentatorubrothalamic pathway²⁻⁴; to our knowledge, anterior thalamic involvement has not been described. We describe a patient who developed an acute, predominantly dystonic tremor after a contralateral anterior thalamic infarct.

REPORT OF A CASE

A 65-year-old right-handed Hispanic woman with a medical history of hypertension, type 2 diabetes mellitus, hypercholesterolemia, and coronary artery disease developed a tremor in her left upper extremity immediately after undergoing a 2-vessel coronary artery bypass at an-

other institution in June 1998. Although she was right-handed, she used her left hand for most activities of daily living. She was seen in our clinic 8 months later because her tremor continued to impair her daily activities. For example, she had difficulty holding a cup or sewing. She denied associated weakness, numbness, pain, gait difficulty, or visual changes. She denied any neurologic symptoms before cardiac surgery.

On neurologic examination, the patient's mental status and cranial nerves were normal. Her muscle power was 5/5 in both the upper and the lower extremities. Her muscle tone was normal; cogwheel rigidity was absent. An irregular, coarse, postural flexion-extension tremor of the left upper extremity, mostly involving the hand, was observed. Mild finger extensor dystonia was also present when the patient's left hand was outstretched. The tremor was not limited to specific tasks and was only minimally present at rest; no action or intention component was present. The dystonia and tremor did not respond to a geste antagonistique. Coordination and gait were intact. Deep tendon reflexes were symmetrically normal, and the plantar responses were flexor bilaterally. Sensation to all modalities was preserved.

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Figure 1. Axial section of T2-weighted cranial magnetic resonance imaging scan showing an infarct in the anterior thalamus.

The results of laboratory investigations and determinations, including complete blood cell count, serum chemistry profile, VDRL, serum ceruloplasmin levels, and thyroid function tests, were normal. Cranial magnetic resonance imaging (MRI) scans (**Figure 1** and **Figure 2**) revealed an infarct in the ventral portion of the right thalamus. Scattered small, subcortical ischemic white matter hyperintensities were also noted. There were no lesions involving the brainstem, cerebellum, or basal ganglia.

COMMENT

A rapid-onset dystonic-postural tremor developed in our patient shortly after she underwent coronary bypass surgery, suggesting that the tremor resulted from a perioperative stroke. We believe that the tremor was caused by a contralateral anterior thalamic infarct, which was detected on delayed cranial MRI scans. Although no perioperative neuroimaging was performed, the MRI scans that we obtained revealed no other lesions that have been previously associated with stroke-related movement disorders.²⁻⁷ It is unlikely that the anterior thalamic infarct seen on MRI scans was incidental, since this is a rare location for ischemic stroke.⁸

The clinical features of tremor associated with thalamic abnormalities have been well described. Miwa et al² reviewed a series of 12 cases involving patients with delayed-onset tremors that developed after posterior thalamic infarction or hemorrhage. The tremors were contralateral to the thalamic lesion and predominantly involved the upper extremity; they appeared from 1 month to 4 years after thalamic stroke; and accompanying sensory disturbances and transient hemiparesis were usu-



Figure 2. Sagittal section of T2-weighted cranial magnetic resonance imaging scan showing an infarct in the anterior thalamus (arrow).

ally present as well. Four patients had dystonia or athetosis in the affected limb. In all cases, the tremors were postural (2 to 5 Hz); a resting component was found in some patients. Holmes³ described a series of patients with coarse, irregular tremors characterized by resting and intention components, usually with postural exacerbation. Autopsy in 2 patients demonstrated lesions in the contralateral midbrain area. Further studies have shown that the Holmes tremor is due to a cerebellar or midbrain lesion disrupting the dentatothalamic or dentatorubrothalamic tract.^{1,9}

The tremor in our case does not clearly follow the patterns previously described.^{2-4,7,8} The unique characteristics of the tremor, such as the absence of associated sensory disturbances and the lack of action or intentional component, distinguish it from tremor of posterior thalamic or dentatorubrothalamic origin. We suspect that these differences are related to the specific localization of the tremor to the anterior thalamus.

It is possible that the tremor in our patient may represent a forme fruste of a focal dystonia.¹⁰ Focal dystonia with and without tremor has been described in association with thalamic stroke.^{2,4,6,7} The lesions were confined to the posterior thalamus in each case.

The pathogenesis of thalamic tremor and dystonia is not precisely known. Structures that may be involved in lesions causing tremor include the ventrolateral nucleus, ventral posterolateral nucleus, ventral posteromedial nucleus, pulvinar nucleus, lateral geniculate body, and thalamic reticular nucleus.^{2,11} Interruption of afferents to the posterolateral thalamus from the brainstem and cerebellum, destruction of thalamic neurons, abnormal volleying of the circuitry between the thalamus and the cerebral cortex, and damage to the thalamic reticular nucleus are some of the proposed pathophysiologic tremorigenic mechanisms.^{2,11,12}

Thalamic dystonia may result from dysfunction of the cerebellopontothalamic pathway.⁶ Alternatively, a disruption of the corticostriatopallidothalamocortical loop after a lesion develops that involves the centromedian nucleus may disinhibit the ventrolateral nucleus, producing increased thalamocortical drive.⁷

The anterior thalamus has not been previously linked to the origin of acquired movement disorders.^{2,4,6} The anterior thalamus consists of the lateropolaris, anterior ventrolateral, and intralamellaris nuclei.^{11,12} The anterior ventrolateral nucleus is further subdivided into the anterior and posterior ventro-oralis subnuclei.^{11,12} The infarct in our case probably involves the lateropolaris, intralamellaris, and anterior ventro-oralis nuclei.

The lateropolaris nucleus participates in basal ganglia circuits, which include pallidothalamic GABAergic neurons that produce an inhibitory tone on thalamic neurons.¹² The efferents project to the prefrontal cortex, insula, and supplementary motor area.¹² The lateropolaris nucleus possibly partakes in the initiation and progression of movement.¹² The anterior ventro-oralis nucleus is a relay nucleus for the pallidal afferents via the ansa and fasciculus lenticularis, and its efferents project to area 6 of the motor cortex.¹² The intralamellaris nucleus also has motor control function.¹¹ It is conceivable that injury to the lateropolaris, intralamellaris, or anterior ventro-oralis nucleus could produce tremor by disrupting planned motor activity.^{11,12}

Alternatively, an anterior thalamic infarct may disturb interneuronal modulatory pathways to posteriorly located nuclei. Studies have demonstrated the presence of GABAergic inhibitory interneurons throughout the thalamus.¹³ The posteriorly located ventrointermedius nucleus is targeted for stereotactic ablation or deep brain stimulation to ameliorate parkinsonian or essential tremor.^{13,14} An anterior thalamic lesion might produce uninhibited oscillations of neurons of the ventrointermedius nucleus, clinically manifesting as a dystonic tremor.

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