Holmes-Like Tremor of the Lower Extremity Following Brainstem Hemorrhage

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Abstract: Holmes tremor is an arrhythmic, 2- to 5-Hz resting, postural, and kinetic upper extremity movement disorder that occurs weeks to months after acute mesencephalic pathology. We present a patient who developed tremor in three body parts postbrainstem hemorrhage with subsequent hypertrophic olivary degeneration and discuss the relevant clinical evolution. Our case is unique because in addition to expected upper extremity and cervical dystonic head tremors, the patient also developed a severe lower extremity movement disorder, which we believe to be a form of Holmes tremor. Tremor involving the lower extremity in this setting has not been previously reported. © 2006 Movement Disorder Society

Key words: hypertrophic olivary degeneration; leg tremor; Holmes tremor; hemorrhage; brainstem

In 1904, Gordon Holmes described a unique motor abnormality characterized by low frequency tremor at rest, which was exacerbated by posture and accentuated with intention.¹ Brainstem pathology, such as intracranial hemorrhage, can result in upper extremity Holmes tremor, which occurs late in the course of recovery, and may correlate anatomically with hypertrophic olivary degeneration (HOD).²,³ Hemidystonia and torticollis may rarely result from brainstem lesions that extend rostrally into the thalamic or subthalamic regions.⁴ Cervical dystonia with superimposed dystonic head tremor, upper limb tremor, and severe lower limb tremor have not previously been described in the same patient following a brainstem hemorrhage with unilateral HOD.

CASE REPORT

A healthy 43-year-old man experienced sudden onset left hemiparesis, left hemesthesia, right lower motor neuron facial paralysis, severe dysarthria, and right upper and lower limb dysmetria. The first brain magnetic resonance imaging (MRI) performed 10 days after symptom onset (Fig. 1A) corroborated initial head computerized tomography (CT), which showed hemorrhage extending from the brachium pontis through the dorsal midbrain on the right. Routine serum and urine analysis were normal. Coagulation studies were within the standard range. The patient had no history of hypertension. After an extended Intensive Care Unit stay, the patient was eventually transferred to a Rehabilitation Center at 1 month posthemorrhage where he continued to have left hemiparesis, right peripheral facial weakness, dysphagia requiring a feeding tube, and profound disequilibrium. MRI/MR angiography 2 months posthemorrhage did not reveal an underlying arteriovenous malformation, but T2-weighted sequences showed a central area of hyperintensity surrounded by a hypointense ring suggestive of a cavernous malformation as the cause of the hemorrhage.

Five months after his initial hemorrhage, the patient developed a coarse, large amplitude tremor of the left arm present at rest and posture, but much more severe with action. The patient also developed a torticollis to the left and a resulting irregular dystonic head tremor. Initial pharmacotherapies to treat the tremors were unsuccessful.

This article includes Supplementary Video, available online at http://www.interscience.wiley.com/pages/0885-3185/suppmat

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and included: propanolol (no response), carbidopa/levodopa (no response), primidone (no response), ropirinole (profound nausea/vomiting). The patient was continued on gabapentin 900 mg per day for his sensory complaints in addition to phenobarbital 120 mg at night for sleep.

Nine months after his hemorrhage, he was evaluated in the Movement Disorders Clinic at the University of Washington Medical Center. He was severely dysarthric, but cognitively intact and able to cooperate with examination. He was blind in the left eye secondary to trauma at age 13, but his right eye had a reactive pupil and nearly intact range of motion with the exception of limited upgaze and far right lateral gaze. There was ptosis of the right lid as a result of a gold implant to minimize corneal exposure. No palatal tremor, myoclonus, or clicks were appreciated. He had a dense right facial lower motor neuron weakness and left facial numbness. Violent intermittent dystonic head tremor was noted, with cocontracting posterior and anterior cervical muscles and a hypertrophied right sternocleidomastoid muscle. Cervical dystonia was characterized by left chin rotation and right lateral head tilt. The tremor in the left arm manifested a slow (2 Hz), regular, low-amplitude tremor at rest, and irregular, high-amplitude, low-frequency (2–3 Hz) tremor with posture and movement. Muscle tone on the left side was increased. Strength and tone were normal on the right side, but right upper and right lower limbs were dysmetric on finger-to-nose and heel-to-shin testing, respectively. Tendon reflexes were difficult to elicit in the upper limbs and ankles, brisk at the knees, but without clonus. He had a left Achilles contracture and a positive left Babinski sign. The right toe response to plantar stimulation was equivocal. Sensory examination revealed left hypesthesia. He was unable to ambulate independently.

The patient received partial improvement of his cervical dystonia and dystonic head tremor with botulinum toxin injections. However, his left upper extremity tremor remained debilitating and a follow-up MRI 10 months after his event showed residual hemosiderin in the area of hemorrhage, no evidence of new hemorrhage, and T2-weighted hyperintensity of the right inferior olivary nucleus suggestive of HOD (Fig. 1B).

Almost 1 year after the acute hemorrhage, he developed a severe irregular, low-frequency (2–3 Hz), high-amplitude left lower extremity tremor at the hip and knee, present at rest which was exacerbated with movement, suggestive of a Holmes-like tremor (see Video). The tremor was severe enough to be transmitted to the trunk. His left upper limb tremor had become less severe than his left lower limb tremor. Rehabilitation has been helpful in providing assistance helping him adapt and perform at the highest level of independence possible. He remains wheelchair-dependent and relies on his wife for most activities of daily living.

DISCUSSION

The Consensus Statement of the Movement Disorder Society on tremor published in 1998 describes Holmes tremor as a rest, intention, and sometimes postural tremor that is not as rhythmic as other tremors. Literature review reveals several monikers for this condition, including: rubral tremor, midbrain tremor, thalamic tremor, cerebellar outflow tremor, and Benedikt’s syndrome. The tremor frequency is usually less than 4.5 Hz, and the delay between the time of the lesion and the occurrence of the tremor is usually between 4 weeks to 2 years. Published reports have largely focused on the upper limb when it comes to tremor, and Holmes tremor is no exception. The case of the patient reported here illustrates that Holmes tremor is not limited to the upper limb and may be present and perhaps more severe in the lower limb.
Upon reviewing the literature, we found no specific references to lower limb Holmes tremor, but we found one case report on a large amplitude tremor in the left hemibody after a right midbrain hemorrhage.\(^7\) Review of the videotape segment of that case report shows a severe left upper limb Holmes tremor and a more subtle left lower limb tremor. In our patient, the lower limb tremor was dramatically worse than the upper limb tremor at 1 year posthemorrhage (see Video). Furthermore, unlike our patient, the previously reported case did not have HOD and responded well to levodopa. While the tremor of the lower extremity is unique, other features of this case have been very predictable based on multiple similar cases. The absence of palatal myoclonus and the temporal evolution of HOD in our patient are noteworthy and reported elsewhere.\(^6\)–\(^12\)

The features of the left lower limb tremor in our patient fit the characteristics of a Holmes tremor: low and irregular frequency, high amplitude, present at rest and with action, and occurring approximately 1 year after a brainstem hemorrhage. The pathophysiology of Holmes tremor is not fully understood, and there is no anatomic reason to discount the involvement of a lower extremity. This is clinically relevant because of the additional morbidity a lower extremity tremor may impart. Pharmacotherapy alone is less likely to be effective because both the dopaminergic nigrostriatal system and the cerebello-thalamic system (or their connections) may be involved.\(^{13}\) Consequently, a multimodal approach in combination with lesioning or high-frequency stimulation of more than one deep brain target may be necessary,\(^{14,15}\) especially when Holmes tremor extends to the lower extremities.

**LEGEND TO THE VIDEO**

Patient with Holmes tremor of the left upper limb and Holmes-like tremor of the left lower limb. The tremor is irregular in frequency, of high amplitude, and is transmitted to the trunk. The left lower limb tremor is briefly suppressible with complete knee extension.

**REFERENCES**

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