Limb-Shaking Transient Ischemic Attacks: Diagnosis, Prognosis, and Management

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Abstract
Limb-shaking transient ischemic attacks (LSTIAs) are a rare manifestation of carotid occlusive disease. We report the case of an 81 year old woman who presented with recurrent LSTIAs and received conservative treatment with cessation of her anti-hypertensive medications. Multi-Slice Detector CT Angiography (MSCTA) demonstrated a right internal carotid artery occlusion with evidence of cerebral autoregulatory compensation via leptomeningeal vessels. This case characterizes LSTIAs and demonstrates the use of MSCTA in diagnosis, prognosis, and management.

INTRODUCTION
Limb-shaking transient ischemic attacks (LSTIAs) are a rare manifestation of cerebral ischemia due to severe internal carotid artery stenosis or occlusion. Multi-Slice Detector CT angiography (MSCTA) is an emerging non-invasive technique that can quickly examine the neurovascular circulation from the Great Vessels to the distal leptomeningeal branches of the cerebral vasculature. Its utility in the emergent evaluation of complex neurovascular disease is becoming increasingly recognized. This case explores the phenomena of LSTIA and demonstrates the use of MSCTA to identify occlusive carotid vascular disease and outline downstream collateral circulation/autoregulatory compensatory responses important in carotid disease reoccurrence prediction.

CASE REPORT
An 81 year old left handed woman presented with left arm “shaking.” She reported repeated rhythmic finger extension with anterior forearm protrusion in discrete 15-20 minute episodes over a 2 hour period. At onset, the patient was standing and performing lower extremity exercises. She also experienced a 30 minute episode of word finding difficulty in the emergency department (ED). Her past medical history included a permanent pacemaker, hypertension, hyperlipidemia, coronary artery disease, and atrial fibrillation without anticoagulation. Ten months prior to presentation, the patient had a TIA with word finding difficulties and left arm numbness; a 95% right internal carotid artery (ICA) stenosis was noted on MSCTA at that time.

Examination in the emergency department, approximately 4 hours after symptom onset, was significant for an irregularly irregular pulse, a blood pressure of 158/72 and a mild pronator drift of the left upper extremity. Non-contrast head CT (NCHCT) was negative for acute signs of infarction. MSCTA demonstrated a right ICA occlusion at the bifurcation. The left ICA was normal with diminutive posterior communicating arteries. Insular and cortical branches were prominent throughout the right middle cerebral artery territory suggestive of autoregulatory dilation to draw collaterals from the Circle of Willis and leptomeningeal vessels. (see Figure 1) An electroencephalogram was negative for epileptiform activity.

The patient had an uneventful hospital course with slow reintroduction of blood pressure medications. Anticoagulation was held for a planned surgery of an incidental mass discovered on chest x-ray. On the day following discharge, she experienced a second left arm shaking episode and transient left hemiparesis that resolved with sitting. On readmission, the patient was not orthostatic and a NCHCT was unchanged from the previous admission. She was discharged with compression stockings and instructed to stop her antihypertensives indefinitely.
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Figure 1

Figure 1

MSCTA of the Carotid Bifurcation and Circle of Willis. A. Complete occlusion of the right internal carotid artery at the bifurcation (black arrow). B. Occlusion of the ICA with no flow at the level of the supraclinoid ICA with a very small posterior communicating artery on the right (white arrow). B-D. Note increased leptomeningeal vessel markings (black arrows) in the right hemisphere compared to the left consistent with autoregulatory, vasodilatory compensation for the right internal carotid occlusion. D. Prominent leptomeningeal vessel at the middle and posterior cerebral artery watershed zone indicative of pial-pial collateralization across vascular territories (most posterior arrow).

DISCUSSION

Limb-shaking transient ischemic attacks were first described in the medical literature in 1962 by Fisher. Nonetheless, individuals presenting with LSTIAs remain a diagnostic challenge because the phenomena is rare and may be difficult to differentiate from focal motor seizures. A careful description of the shaking distinguishes the ischemic etiology of LSTIAs from a true Jacksonian March. Furthermore, onset upon standing, with exercise, or hyperventilation suggests LSTIA. Presumably, the underlying mechanism of LSTIA involves cerebral ischemia, but the electrophysiological and/or neural circuits involved have yet to be elucidated. It has been argued that selective impairment of inhibitory networks with subsequent release of motor activity may be involved, or that cerebral ischemia itself in the proper context may result in direct transient neuronal excitability.

Our patient presented with classic manifestations of LSTIAs and experienced an episode of expressive aphasia consistent with left hand/right hemispheric dominance. The close temporal association of her limb shaking, expressive aphasia and left hemiparesis suggests a common mechanism. The prominence of the patient’s right hemispheric leptomeningeal vessels in conjunction with a complete right ICA occlusion indicates lateralized autoregulatory compensation. Activities such as lower extremity exercise and standing which increase local arterial blood flow and impair venous return/preload may exceed the capacity of the leptomeningeal collaterals and precipitate symptoms.

This case highlights the importance of considering transient cerebral ischemia in the differential diagnosis of involuntary limb movements. Individuals with carotid stenosis and TIA are at increased risk for stroke – the majority of recurrences occur in the first few days after TIAs. It is critical to identify patients quickly as they benefit from carotid revascularization. Individuals with carotid occlusion and recurrent symptoms may be managed with changes to their antihypertensive medications in an effort to maintain adequate cerebral perfusion. Revascularization by extracranial-intra cranial anastomosis has been reported to relieve limb shaking attacks and can be considered in recurrently symptomatic patients.

There are several clinical and radiologic variables that modify the risk of stroke in patients with symptomatic carotid disease of > 70% stenosis. Our patient had several important clinical features that are predictors of recurrent stroke. They include: cerebral rather than isolated ocular symptoms, age > 75, and recurrent ischemic events over a period greater than six months. Her angiographic predictors of recurrence on MSCTA were the presence of primarily leptomeningeal collaterals and the absence of significant Circle of Willis collaterals. The anterior communicating and posterior communicating arteries were not contributing significant collateral circulation. The strikingly increased intensity of the leptomeningeal vessels in the right hemisphere suggests maximal dilation. This may explain why exercise and standing – activities that normally do not lower cerebral perfusion induced neurologic dysfunction in this patient. Importantly, the patient had a normal contralateral ICA which decreases the long-term risk of
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recurrence and is associated with improved autoregulatory reserve in the territory of the ipsilateral carotid occlusion, as previously demonstrated by transcranial Doppler CO2 reactivity testing.\textsuperscript{15}

CONCLUSION

In summary, LSTIAs are a rare manifestation of severe or occlusive internal carotid artery disease. Early recognition is critical for prevention of recurrent events, as both medical and surgical/endovascular options are available. Knowledge of the clinical and radiologic factors associated with recurrent events is important to assess the need and timing for treatment. Use of MSCTA can minimize exposure to invasive angiographic procedures and provide diagnostic and prognostic information essential to management.

References

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