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LETTER TO EDITOR

A Patient Presenting with Repeating Transient Hemiballismus Due to Critical Stenosis of the Internal Carotid Artery

Dear Editor,

A 62-year-old male patient was admitted to our emergency service due to transient involuntary movements of his left upper extremity which had abruptly started 6 h before admission and lasted for a few hours. The patient described these movements as violent, coarse, and wide-amplitude movements of his left hand including proximal part of the upper limb. It was learned that he had a 40 pack-year smoking history. The patient had no another medical history of a disease constituting risk factor for vascular disease such as hypertension, hyperlipidemia, or diabetes mellitus. Besides, he had no history of a seizure disorder, parkinsonism, or prior use of a neuroleptic. The movements had recovered approximately 2 h before arrival. At admission to our center, the patient was evaluated as fully orientated and cooperative, and other neurological examination including motor, sensory, and cerebellar tests were evaluated as normal. Examination of vital signs revealed high blood pressure (180/90 mm H_2O). The results of the blood tests including hemogram, full biochemistry, lipid profile, fasting glucose, HbA1c, thyroid-stimulating hormone, B12, and folic aside were within normal ranges. Diffusion-weighted imaging (DWI) revealed millimetric diffusion-restricted lesions in the right superior parietal lobule and within the right periventricular white matter [Figure 1]. To exclude a possible mechanism of ictogenesis, we have performed routine electroencephalography which was evaluated within normal limits. On the other hand, brain/neck computed tomography angiography (CTA) revealed critical stenosis of the cavernous portion of the right internal carotid [Figure 2]. Conventional angiography also confirmed the stenosis [Figure 3]. However, interventional treatment option was discussed with an interventional neurosurgeon (ED), and it was evaluated as a strictly high-risk procedure. Further investigations of stroke including cardiac evaluations (echocardiography, electrocardiography, and 24-h rhythm Holter monitoring) were evaluated as normal Ergo, the patient was disharged with dual antiplatelet therapy (clopidogrel and aspirin) and atorvastatin (20 mg). Nevertheless, he applied to our polyclinic three weeks later again due to repeating attacks of involuntary movements of his left upper limb over the last two days which occurred abruptly and lasted for 10 min. Cranial DWI, at this time, revealed newly developed multiple millimetric diffusion-restricted lesions corresponding to the territories of the right internal carotid artery (ICA) [Figure 4]. Brain/neck CTA was also repeated which revealed critical stenosis of the right ICA similar to the rate at the previous imaging. Dual antiplatelet therapy was switched to warfarin therapy. During the four days period of hospitalization on anticoagulant therapy, no attacks have occurred. In conclusion, the patient was discharged after providing therapeutic INR value. He was symptom free on his last follow-up visit four months later under warfarin therapy.

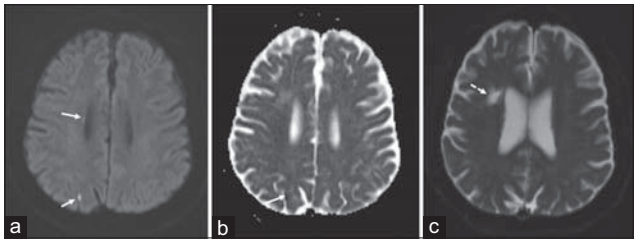

Figure 1: Cranial diffusion-weighted imaging and apparent coefficient sequences show diffusion restricted regions in the right superior parietal lobule and within the right periventricular white matter (a-b, arrows). A chronic ischemic lesion in the right corona radiata corresponding to the region of internal watersheds (c, jagged arrow)

DISCUSSION

Poststroke movement disorders represent a significant rate of secondary movement disorders (up to 22%).[1] However,

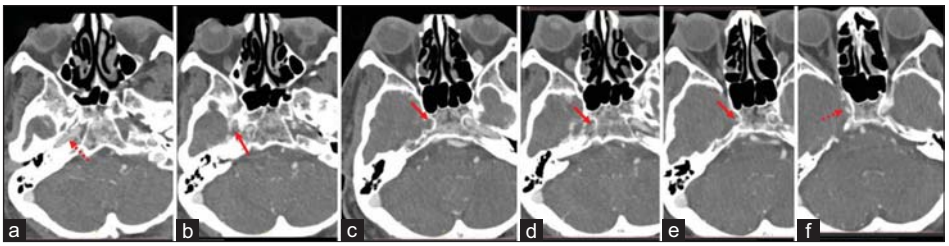

Figure 2: Axial cerebral computed tomography angiography images. (a) Patent internal carotid artery in the petrous portion (jagged arrow). (b-e) Critical stenosis of the right internal carotid artery in the cavernous segment (obvious in the images of c and d, arrows). (f) Image showing the patent flow at the supraclinoid segment of the right internal carotid artery (jagged arrow)

their occurrence in all strokes is rare which is estimated to be 1%–4% of all strokes.[1] Among these, poststroke ballismus is rather a common subtype, and its causal association with carotid artery occlusive disease has been multiple times illustrated previously.[2-8] Arrestingly, the transient occurrence of ballismus as the only sign of transient ischemic attack is considerably rarely reported.[8,9] In our patient, the only clinical presentation was the history of a transient limb shaking, and DWI showed minor ischemic lesions which could easily be underestimated. However, computed tomography (CT) angiography showed critical stenosis in the cavernous portion of the right ICA. In a previous report by Joseph *et al*., a remarkable patient who had presented with left-sided hemiballismus, facial paralysis, and dysarthria was reported.[8] In this patient, the cranial magnetic resonance imaging (MRI) result was unremarkable, but the CT angiography had revealed 90% stenosis of the right ICA. Via the presentation of this patient, the authors suggested involving neurovascular imaging in the initial workup of hemiballismus to avoid underdiagnosis and administer timely treatment.[8] Similarly, the clinical presentation of our patient may give substantial considerations to be kept in mind in this regard. Of note, there was no additional clinical manifestation other than monoballismus in our patient which surely makes the diagnosis more sophisticated.

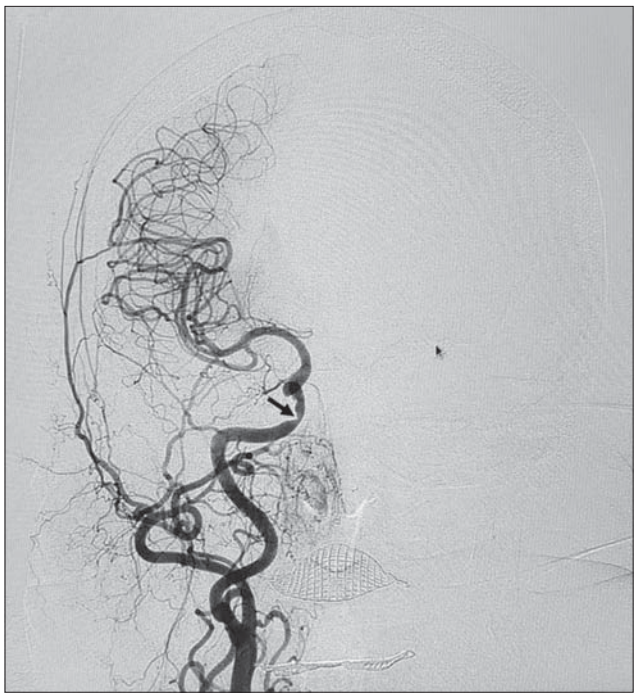

Figure 3: Digital angiography confirms the critical stenosis of the right internal carotid artery in the cavernous segment (arrow)

While there are many debates regarding the pathophysiology of ballismus (and/or hemichorea) following a stroke, the exact mechanisms and responsible lesion sites remain to be elucidated. In the majority of the cases of ballismus due to ischemic etiology, the reported ischemic lesion sites include basal ganglia or subthalamic nucleus.[2,3] Disruption of the direct and indirect pathways through the

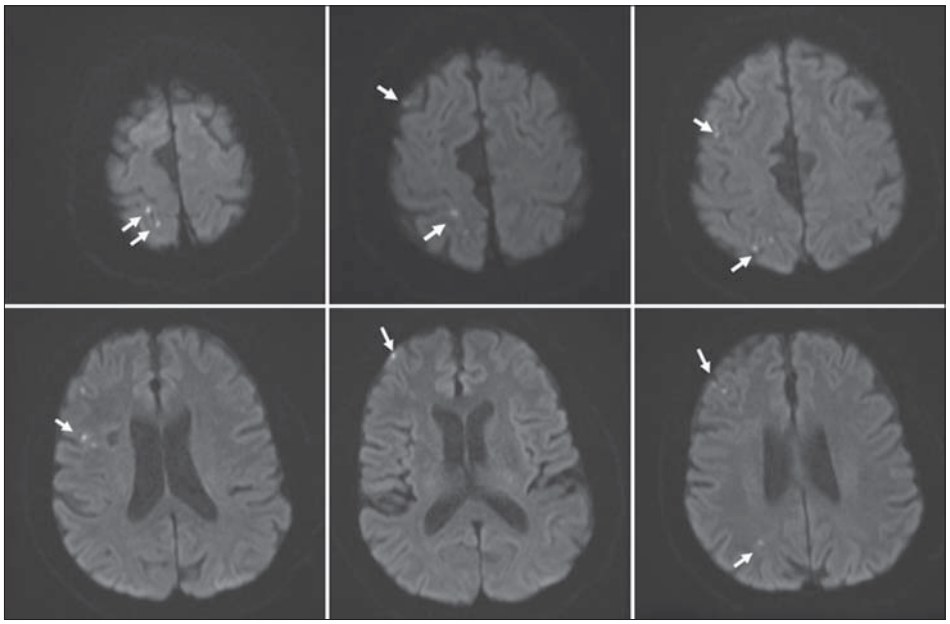

Figure 4: Cranial diffusion-weighted images (from rostral to caudal), performed at second admission, show multiple millimetric diffusion restricted lesions corresponding to the territories of the right internal carotid artery (arrows)

basal ganglia is the main pathophysiological mechanism suggested by these authors. However, there are also authors reporting cases without lesions within the basal ganglia.[4,5] Besides, lesions of the subcortical white matter have also been associated with chorea/ballismus in a few reports.[6] In light of these evidence, the underlying pathophysiology of repeating monoballistic movements in our patient may be a critical point of discussion. Arrestingly, there is a growing amount of evidence that hemodynamic failure is the likely mechanism for limb-shaking transient ischemic attacks from severe carotid artery disease.[5,7-9] On the other hand, cases of ballismus due to lesions of the caudate nucleus and patchy multifocal hemispheric white matter regions have also been reported.[6,10,11] In these reports, it has been hypothesized that an interruption in the corticostriate pathway by vascular lesion diminishes the rate of firing of striatal neurons, liberating the pallidum from striatal inhibition and causing the abnormal movements of hemichorea-hemiballismus. In our opinion, the main limitation of our report was that no perfusion imaging was performed to understand the possible role of hypoperfusion. On the other hand, another discussion may be associated with the differential diagnostic process of monoballismus in our patient. The most common cause of ballistic movements is vascular insult including basal ganglia. However, it can occur secondary to various causes including tumors, encephalitis, neurodegenerative disorders, drugs, auto-immune diseases, and metabolic disorders such as nonketotic hyperglycemia.[12] In our patient, laboratory investigations revealed normal blood glucose level and HbA1A level excluding nonketotic hyperglycemia. There was no previous history of autoimmune disease or use of a neuroleptic. Finally, cranial MRI revealed the diagnosis of vascular movement disorder clearly.

In conclusion, we think that this report is important by the reason of that isolated monoballismus due to ICA stenosis is a strictly rare presentation. We think that our case represents a smart illustration in this regard drawing attention to ICA stenosis as a critical cause of acute onset monoballismus. The clinicians should be aware of this critical etiology in clinics with acute-onset ballismus.

Declaration of patient consent

The consent form has been obtained from the patient. In the form, the patient has given his consent for his clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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