



Recurrent Acute Ischemic Stroke Associated with Dolichoectatic Aneurysm of the Internal Carotid Artery

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Key words

- Acute ischemic stroke
- Best medical treatment
- DOAC
- Dolichoectatic aneurysm
- ICA
- Vertebrobasilar dolichoectasia

Abbreviations and Acronyms

ICA: Internal carotid artery

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INTRODUCTION

Cranial artery dolichoectasia is characterized by dilated, elongated, and tortuous cranial arteries such as the distal internal carotid arteries (ICAs).¹ Dolichoectasia and dolichoectatic aneurysm—particularly of the ICA—are rare (prevalence 0.05%–0.06%)^{2,3} but can lead to stroke. We report a case of acute cerebral infarction associated with a dolichoectatic aneurysm of the left ICA combined with right dolichoectatic aneurysm, vertebrobasilar dolichoectasia, and atrial fibrillation. Written informed consent was obtained from the patient for publication of this case report and the accompanying images, and the study design was approved by the appropriate ethics review board.

CASE DESCRIPTION

An 80-year-old man had episodic aphasia for a duration of 3 days. He had a history of hypertension and atrial fibrillation. He was taking rivaroxaban (10 mg/day) and

■ **BACKGROUND:** Cranial artery dolichoectasia is characterized by dilated, elongated, and tortuous cranial arteries. Dolichoectasia and dolichoectatic aneurysm—particularly of the internal carotid arteries (ICAs)—are rare but can lead to stroke. We report a case of suspected recurrent acute cerebral infarction and dolichoectatic aneurysm of the left ICA combined with right dolichoectatic aneurysm, vertebrobasilar dolichoectasia, and atrial fibrillation.

■ **CASE DESCRIPTION:** An 80-year-old man experienced 3 days of recurrent aphasia episodes. He had a history of atrial fibrillation and hypertension. Neurologic findings indicated Wernicke aphasia. Magnetic resonance imaging showed acute cerebral infarction in the left temporal and parietal lobes. Magnetic resonance angiography and cerebral angiography showed dolichoectatic aneurysms in the C1-2 portion of the bilateral ICAs and vertebrobasilar dolichoectasia. Possible diagnoses included convulsion, cardioembolic infarction, atherothrombotic cerebral infarction, and acute ischemic stroke associated with the dolichoectatic aneurysm of the left ICA. Oral antiplatelet drugs, direct oral anticoagulants, and anticonvulsants were administered. Nine months after initial hospitalization, the patient had a recurrence of acute ischemic stroke and was rehospitalized. He was treated with a new direct oral anticoagulant, clopidogrel, statins, and ethyl icosapentate. At 2 years after initial hospitalization, he has had no recurrence of stroke, has a modified Rankin Scale score of 2, and is currently being followed as an outpatient.

■ **CONCLUSIONS:** A direct oral anticoagulant (edoxaban) and clopidogrel were effective for suspected recurrent acute ischemic stroke associated with dolichoectatic aneurysm of the ICA combined with contralateral dolichoectatic aneurysm, vertebrobasilar dolichoectasia, and atrial fibrillation.

antihypertensive medication and had good compliance. Neurologic assessment revealed Wernicke aphasia but no apparent quadriplegia or other neurologic deficits. The patient scored 6/42 on the National Institutes of Health Stroke Scale. Blood analysis (including blood cell counts, biochemistry, and coagulation parameters) revealed no abnormalities. Electrocardiography revealed no atrial fibrillation at hospitalization. Cardiac ultrasound examination showed no intra-atrial thrombus.

Imaging Findings

Head computed tomography showed marked elongation, dilation (>4.5 mm),

and calcification from the left vertebral artery to the basilar artery. Both ICAs were dilated, elongated, and partially calcified; the vascular lumens were uniform with slightly increased density (Figure 1A). Diffusion-weighted magnetic resonance imaging showed areas of high intensity from the left temporal lobe to the parietal lobe consistent with the vessel-dominant areas of the M2 inferior trunk (Figure 1B). Magnetic resonance angiography revealed aneurysms in the C1-2 portion of both ICAs and poor visualization of both middle cerebral arteries (Figure 1C). Time-of-flight magnetic resonance angiography showed heterogeneous high-intensity changes from the left

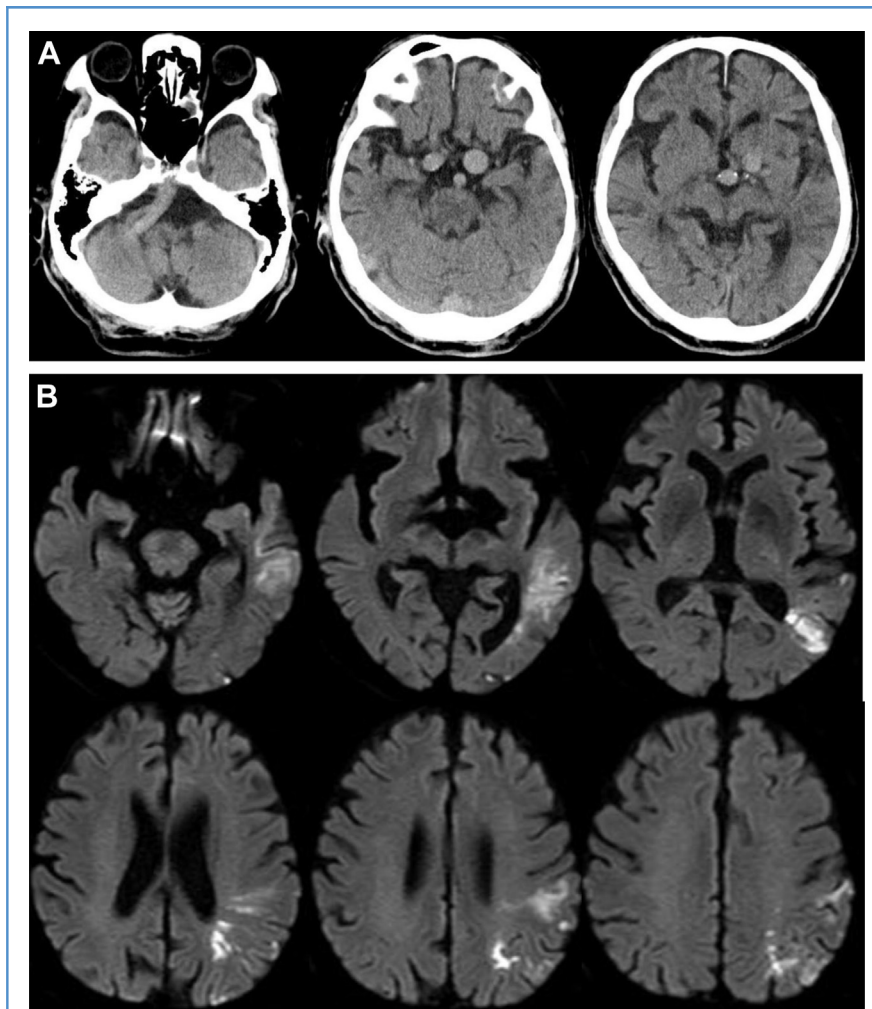


Figure 1. (A) Head computed tomography images showing remarkable elongation, dilation, and calcification from the left vertebral artery to the basilar artery. The vascular lumen was uniform with slightly increased density. Both internal carotid arteries (ICAs) also showed expansion with calcification. The vascular lumens were uniform with slightly increased density. (B) Diffusion-weighted magnetic resonance imaging showing high-intensity areas in the left temporal lobe

and parietal lobe, consistent with the vessel-dominant areas of the M2 inferior trunk of the left middle cerebral artery. (C) Magnetic resonance angiography showing aneurysms in the C1-2 portion of the bilateral ICAs, with poor visualization of the bilateral middle cerebral arteries. (D) Time-of-flight magnetic resonance angiography showing heterogeneous high-intensity changes from the left vertebral artery to the basilar artery and in the bilateral ICAs.

vertebral artery to the basilar artery and in both ICAs (Figure 1D).

Acute cerebral infarction and thrombus progression was assessed by cerebral angiography. Giant cerebral ICA aneurysms at the C1-2 level (right, approximately 20 mm; left, approximately 25 mm; with both anterior choroidal arteries diverging from the aneurysms) were

found. Dolichoectatic aneurysms of the bilateral ICAs were diagnosed (Figure 2A and B). Aneurysmal blood flow was turbulent and delayed bilaterally. Left ICA angiography poorly visualized the M2 inferior trunk from the origin, but aneurysmal thrombosis was absent. Right ICA angiography showed cross-flow to the left ICA and left middle

cerebral artery in the late arterial phase during the Matas test and right M2 superior trunk stenosis. Bilateral cervical ICAs were elongated but without severe stenosis. Bilateral external carotid artery angiography revealed an absence of intracranial anastomosis. Vertebral angiography revealed significant elongation and dilation from the left vertebral artery

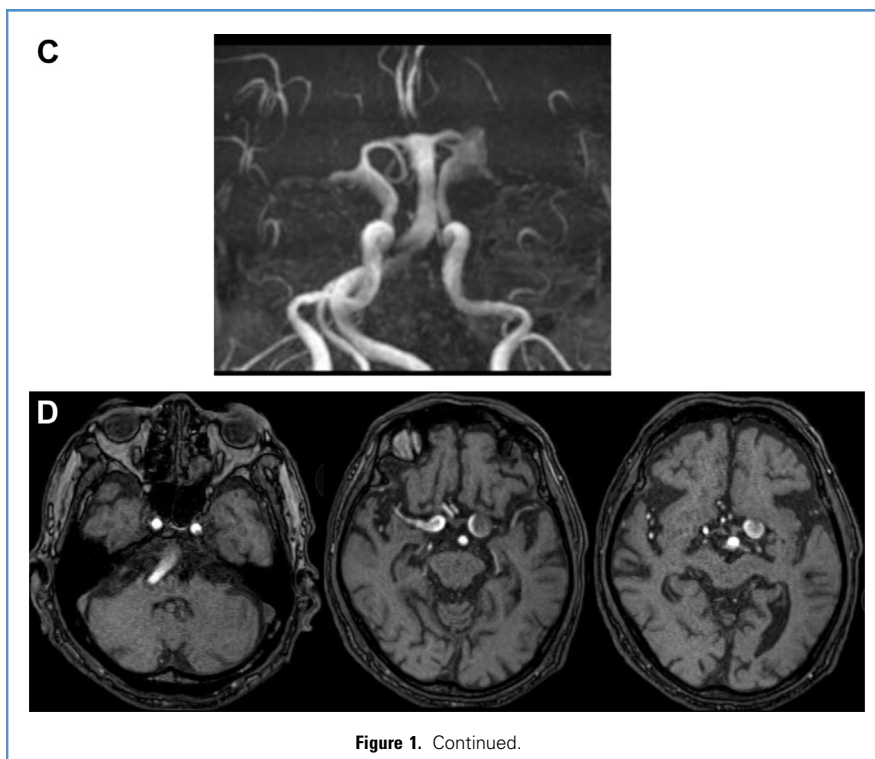


Figure 1. Continued.

to the basilar artery and dolichoectasia (Figure 2C and D). Allcock test revealed no cross-flow to the anterior circulatory system.

Posthospitalization Course

The patient was treated with argatroban by intravenous injection, an oral antiplatelet drug (aspirin, 100 mg/day), an anticoagulant (rivaroxaban, 10 mg/day), and an anticonvulsant (levetiracetam, 1000 mg/day). Electroencephalograms conducted during oral administration of the antiepileptic drugs did not show epileptic waves. During hospitalization, the patient did not have progressive cerebral infarction or a seizure attack. One month after hospitalization, his aphasia had subsided, and he was transferred to a rehabilitation hospital. At this time, he had a modified Rankin Scale score of 2.

Nine months after his initial hospitalization, the patient had mild right upper limb paralysis and was transported to our hospital again. His oral compliance was good. Magnetic resonance imaging

showed acute lacunar infarction in the left centrum semiovale (Figure 3A and B). He was treated with edaravone by intravenous injection, an oral antiplatelet drug (clopidogrel, 75 mg/day), an anticoagulant (edoxaban tosylate hydrate, 30 mg/day), rosuvastatin calcium (2.5 mg/day), and ethyl icosapentate (1800 mg/day). The antithrombotic drugs administered at the time of the patient's initial discharge were changed. The patient's right upper limb paralysis improved, and he was discharged 1 week after his second hospitalization. Two years have passed since the patient's initial hospitalization. He has not been readmitted for the recurrence of stroke; however, he has aftereffects of aphasia with a modified Rankin Scale score of 2, and is currently being followed as an outpatient.

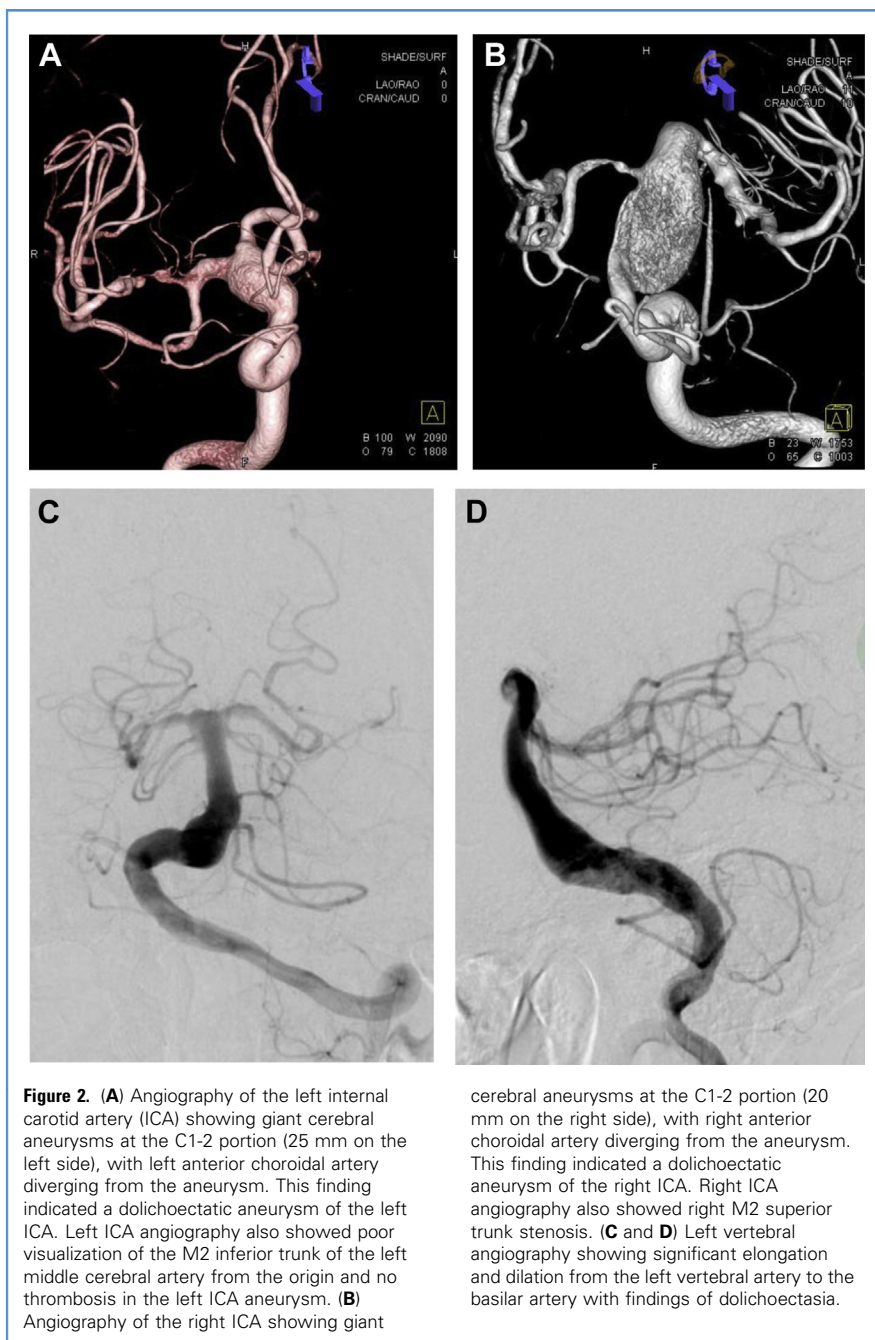
DISCUSSION

The main causes of dolichoectasia include atherosclerosis and hypertension,³ although

other conditions have been reported.⁴⁻⁶ From a pathogenic perspective, dolichoectasia is a dilatative arteriopathy that occurs as a result of deficiencies in the muscularis and internal elastic lamina, Marfan syndrome, Ehlers-Danlos syndrome, pseudoxanthoma elasticum, and other less well-defined connective tissue disorders.⁷ The patient in our case had a history of chronic hypertension but no family history of Marfan syndrome, Ehlers-Danlos syndrome, or other relevant pathologies. Therefore, atherosclerotic vertebrobasilar dolichoectasia and dolichoectatic aneurysms of both ICAs were possible diagnoses. Hyperimmunoglobulinemia syndrome⁸ and infection⁹⁻¹¹ may also cause arterial dilation, but the patient's IgE level was not elevated, and there was no evidence of infection. Dolichoectasia involving both the vertebrobasilar and the carotid systems is rare.¹ Our case represents a particularly rare manifestation, as dolichoectasia existed in both the anterior and the posterior circulation, with dolichoectatic aneurysms of the bilateral intracranial ICAs. Coupled with the absence of cross-flow from the contralateral ICA and posterior circulation, the flow velocity of the left middle cerebral artery was likely further decreased, possibly contributing to cerebral infarction.

The delayed flow velocity in our case may be related to thrombus formation. Transcranial Doppler studies of intracranial dolichoectasia have demonstrated reduced peak flow velocity.¹² Reduced flow can lead to stagnation of the blood column to give spontaneous echo contrast and increase the risk of formation of a thrombus with embolization distally.¹³ Our patient exhibited increasing symptoms similar to a transient ischemic attack before hospitalization, suggesting convulsion, a cardioembolic infarction, a hemodynamic cerebral infarction, or an artery-to-artery embolization mechanism, all of which are associated with dolichoectatic aneurysms of the left ICA and/or an atherothrombotic cerebral infarction in the M2 inferior trunk.

Previously, in dolichoectasia cases, anticoagulant therapy was recommended



for the prevention of thrombi and embolisms when ischemic symptoms occurred¹⁴; antiplatelet therapy was always recommended, regardless of ischemia.¹⁵ In our case, given the standard treatment for atherothrombotic cerebral infarction and the patient's atrial fibrillation, antiplatelet therapy

and anticoagulant therapy were administered during his initial hospitalization. Although these therapies did not resolve his aphasia, they may have prevented progression of cerebral infarction during his initial hospitalization. Although there is no evidence for dolichoectasia causing

ischemic strokes, we treated the patient using the best medical treatment. The patient was given a direct oral anticoagulant (edoxaban), clopidogrel, statins, and ethyl icosapentate following the recurrence of ischemic stroke, which was based on the mechanism of thrombi formation.

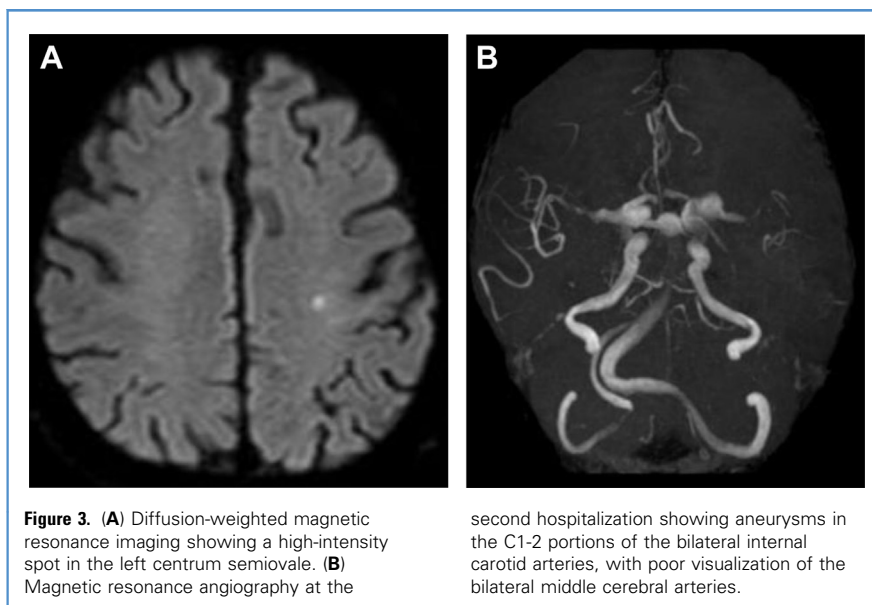


Figure 3. (A) Diffusion-weighted magnetic resonance imaging showing a high-intensity spot in the left centrum semiovale. (B) Magnetic resonance angiography at the

second hospitalization showing aneurysms in the C1-2 portions of the bilateral internal carotid arteries, with poor visualization of the bilateral middle cerebral arteries.

CONCLUSIONS

We report an extremely rare case of suspected recurrent acute cerebral infarction associated with a dolichoectatic aneurysm of the ICA combined with dolichoectatic aneurysm of the contralateral ICA, vertebralbasilar dolichoectasia, and atrial fibrillation. Standard medical care, which included a direct oral anticoagulant (edoxaban), clopidogrel, statins, and ethyl icosapentate, was effective for our patient.

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