

[CASE REPORT]

Takayasu's Arteritis with a Thrombosed Aneurysm on the Common Carotid Artery Causing Ischemic Stroke

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Abstract:

Takayasu's arteritis is an inflammatory disease of unknown etiology that causes stenosis, occlusion, or dilatation of the aorta and its major branches, the pulmonary arteries, and the coronary arteries. The incidence of extracranial carotid artery aneurysm in patients with Takayasu's arteritis is reportedly 1.8-3.9%. We herein report a patient with Takayasu's arteritis who presented with transient left hemiplegia immediately after neck massage. Carotid ultrasonography revealed a thrombus within the fusiform aneurysm on the right common carotid artery. We speculated that fragmentation from the intra-aneurysmal thrombus was caused by neck massage.

Key words: extracranial carotid artery aneurysm, Takayasu's arteritis, stroke

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Introduction

Takayasu's arteritis (TA) is an inflammatory disease of unknown etiology that causes stenosis, occlusion, or dilatation of the aorta and its major branches, the pulmonary arteries, and the coronary arteries (1). Although TA is most commonly diagnosed in young women of Asian descent, it occurs worldwide, with an incidence ranging from 0.3 to 40 per million (2-4).

Approximately 10-20% of patients with TA are complicated with ischemic stroke or transient ischemic attack (TIA) (5), while the incidence of extracranial carotid artery aneurysm (ECAA) ranges from 1.8-3.9% in patients with TA (6-8). We herein report a case of TA with transient left hemiplegia immediately after neck massage, accompanied by a thrombosed right common carotid artery (CCA) aneurysm detected by carotid ultrasonography.

Case Report

A 42-year-old woman was admitted to our department because of transient left hemiplegia that lasted for 15 minutes immediately after neck massage. She had a 22-year history of TA and had been taking aspirin (100 mg/day) and predni-

solone (5 mg/day). The most recent contrast-enhanced computed tomography (CT) results five years before the onset revealed an aneurysmal dilatation on bilateral CCAs without aneurysmal thrombus. The patient also had a history of bypass surgery for stenosis in the left subclavian artery, ascending aortic arch replacement, and thoracic aortic aneurysm stenting.

Upon admission, the patient's blood pressure was 100/62 mmHg, pulse rate was 82 beats/min, and body temperature was 36.7°C. The patient's neurological findings were normal. Blood chemistry test results showed a white blood cell count of 12,660/mm³ and a C-reactive protein concentration of <0.1 mg/dL. D-dimer was elevated at 1.8 µg/mL, but no other abnormalities were observed.

Diffusion-weighted imaging showed acute infarcts in the right insular cortex (Fig. 1a) and the right parietal lobe (Fig. 1b). Magnetic resonance angiography showed no occlusive disease in the intracranial arteries (Fig. 1c). CT angiography showed a thrombosed right CCA aneurysm (Fig. 1d). The size of the aneurysm remained unchanged from five years before the onset. Carotid ultrasonography revealed a thrombus within the fusiform aneurysm of the right CCA (Fig. 2a). The macaroni sign, as a characteristic sign of TA (9), was not observed on carotid ultrasonography. Although we were unable to perform advanced vessel wall im-

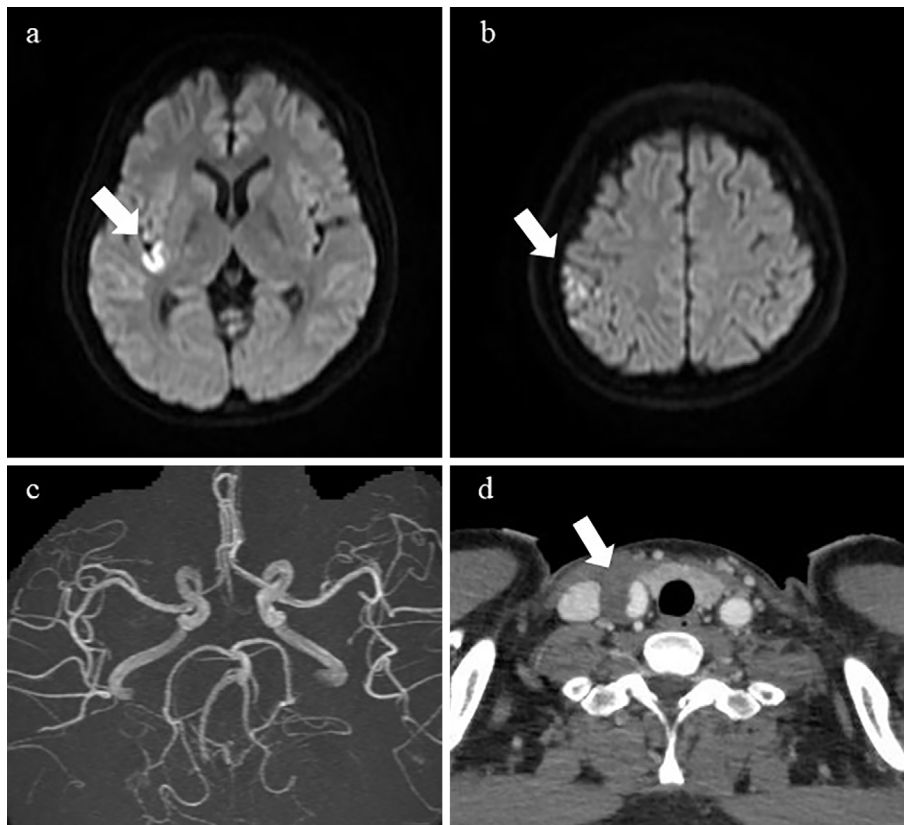


Figure 1. Brain MRI (a, b), MRA (c), and contrast-enhanced CT (d) of the neck. Diffusion-weighted brain MRI revealed acute cerebral infarctions scattered in the right insular cortex (a, arrow) and the right parietal lobe (b, arrow). MRA (c) did not show any occlusion or stenosis of major cerebral arteries. Contrast-enhanced CT showed a fusiform aneurysm of the right CCA and an intra-aneurysmal thrombus (d, arrow). CCA: common carotid artery, CT: computed tomography, MRA: magnetic resonance angiography, MRI: magnetic resonance imaging

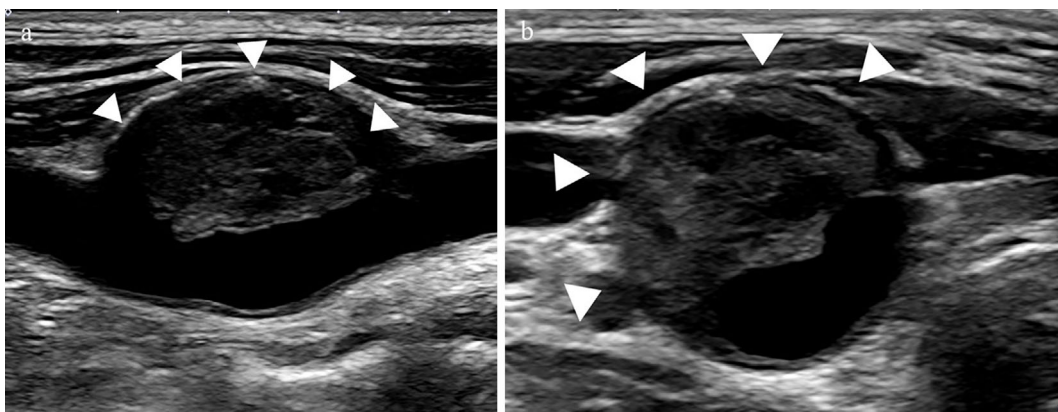


Figure 2. Long-axis (a) and short-axis (b) images of carotid ultrasonography of the right CCA. A thrombus within the fusiform aneurysm (arrowheads) was revealed on the right CCA. CCA: common carotid artery

aging, such as three-dimensional T1-weighted magnetic resonance imaging, carotid ultrasonography revealed the association between the carotid intima and thrombus and the absence of carotid plaques (Fig. 2a, b). There was no other embolic source on electrocardiography (ECG), Holter ECG, or transthoracic echocardiography. In the acute phase, the patient was treated with clopidogrel at an initial dose of 300

mg, followed by 75 mg per day, as well as aspirin, which had been prescribed originally. At discharge, aspirin was changed to cilostazol (100 mg twice daily), and no recurrence was observed thereafter.

Discussion

ECAAs are rare and account for <1% of all arterial aneurysms (10). The most common site of ECAA is the CCA, just proximal to the carotid bulb (10). Although the common causes of ECAA include atherosclerosis, trauma, a history of surgery followed by infection, neck irradiation, and fibromuscular dysplasia (11-14), less frequent causes include Marfan syndrome, Behcet's disease, and TA (7). Previous reports suggested that the percentage of patients presenting with ischemic stroke or TIA ranged 12-51% in patients with ECAA (15-18).

Medical, surgical, and endovascular treatments are used for ECAA (11, 15, 19, 20). Although there is insufficient evidence to suggest the optimal treatment for ECAA (19), conventional surgery is regarded as the standard treatment, especially for symptomatic or growing ECAAs requiring removal with or without arterial replacement. Endovascular repair for ECAA has only been reported in a small number of case series (21). However, the incidence of aneurysm in patients with TA ranges from 4.9% to 31.9% (22-25). The most common site of aneurysm in patients with TA is the aorta, followed by the subclavian artery, the brachiocephalic artery, and the CCA. The incidence of ECAA in patients with TA ranges from 1.8% to 3.9% (6-8, 24); however, thrombosed ECAAs in patients with TA are rarely reported (26, 27). In the present case, we were unable to identify any other factors that may have induced thrombus formation other than oral prednisolone (28). Careful follow-up with carotid imaging, including carotid ultrasonography, which is minimally invasive and provides useful information, should be considered for patients with TA to detect aneurysmal dilatation and intra-aneurysmal thrombus of the carotid artery.

In terms of treatment, although the indications for ECAA surgery in patients complicated with TA have not been established because of its scarcity, there are some reports of successful surgical treatment for ECAA in patients with TA (6, 7). In addition, there is insufficient evidence to support the use of thrombolysis and endovascular treatment for patients with acute ischemic stroke and TA. Of note, the present case suggests that neck massage can induce intra-aneurysmal thrombus fragmentation. Although formation of the intra-aneurysmal thrombus suggested a recurrence risk of distal emboli from the ECAA, we decided to continue medical treatment while considering the risk of triggering stroke with external stimuli after discussing the situation with the patient and consulting a surgeon. We would like to emphasize that external stimuli that compress the carotid artery should be avoided in patients with TA and ECAA. To reduce the risk of recurrence, dual antiplatelet therapy with aspirin and clopidogrel was administered in the acute phase (29), followed by a combination of clopidogrel and cilostazol to minimize the long-term bleeding risk with dual antiplatelet therapy (30).

Conclusion

In conclusion, we reported a case of TA with ischemic stroke due to a thrombosed CCA aneurysm evaluated by carotid ultrasonography. External stimuli that compress the carotid artery should be avoided in such patients. Attention should be paid to the development of aneurysmal dilatation and thrombus formation within an aneurysm; thus, follow-up should be performed with carotid imaging, including carotid ultrasonography, in patients with TA.

The authors state that they have no Conflict of Interest (COI).

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