

Hyoid bone compression-induced carotid dissecting aneurysm: A case report

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Abstract. Bony structures around the carotid artery, such as the styloid process and the hyoid bone, can cause dissection, compression, plaque formation and plaque rupture of the carotid artery. The present study aimed to present a novel case of hyoid bone elongation causing dissecting aneurysm of the carotid artery. However, the patient had no permanent neurologic symptoms. An 80-year-old man presented with right hemiparesis for >5 h despite preventive therapy with antiplatelets and statins. Magnetic resonance imaging revealed acute infarction in the left parietal lobe. Contrast-enhanced computed tomography revealed two cysts with some calcification located at the bifurcation of the right internal carotid artery (ICA) and the right greater horn of the hyoid bone adjacent to the right ICA. A color duplex scan of the carotid vessels confirmed the relationship between dissecting aneurysm and the hyoid bone. In conclusion, greater attention should be paid to the bony structures around the carotid artery.

Introduction

The carotid artery, a major blood vessel in the neck, has a crucial role in supplying blood to the brain, neck and face. Its anatomical positioning and function make it susceptible to various pathologies, some of which can be life-threatening. One such rare pathology is dissecting aneurysm of the carotid artery associated with the hyoid bone. The hyoid bone, a U-shaped bone situated in the anterior midline of the neck, is unique, as it is the only bone in the human body that does not articulate with any other bone. Instead, it is suspended by muscles and ligaments, giving it mobility that may, in rare instances, lead to mechanical interference with nearby structures. Dissecting

aneurysm of the carotid artery associated with the hyoid bone is rare. Mechanical interference between the hyoid bone and the carotid artery has previously been reported as a cause for stenosis (1,2), pseudoaneurysm (3) or dissection (4,5) of the carotid artery. The case presented in the present study was perhaps the first case of dissecting aneurysm of the carotid artery caused by an elongated hyoid bone.

Case presentation

An 80-year-old man with hypertension, who was taking amlodipine and irbesartan, presented with right hemiparesis for >5 h despite preventive therapy with antiplatelets and statins. The neurologic examination disclosed only right-sided hemiparesis. The patient was admitted to the Third People's Hospital of Chengdu (Chengdu, China) in November 2018.

Diffusion-weighted magnetic resonance imaging revealed acute infarction in the left parietal lobe and magnetic resonance angiography detected stenosis of both posterior cerebral arteries (Fig. 1). No responsive stroke and associated symptoms were found.

Contrast-enhanced computed tomography revealed two cysts with some calcification located at the bifurcation of the right internal carotid artery (ICA) and the right greater horn of the hyoid bone adjacent to the right ICA (Fig. 2).

A color duplex scan of the carotid vessels showed several inhomogeneous plaques in the carotid bifurcation, with irregular thickening of the vessel walls. Two cysts, in which red and blue flow signals were detected, communicated with the right common carotid artery (CCA) and the origin of the right ICA separately, suggesting dissecting aneurysm. The right CCA and the origin of the right ICA were compressed by the hyoid bone repeatedly when swallowing and speaking (Fig. 3). An ultrasound video also showed ICA being collapsed by the hyoid bone during swallowing or speech (Video S1).

The dissecting aneurysm of the carotid artery was associated with an elongated hyoid bone. The patient was treated with aspirin (100 mg qd) and clopidogrel (clopidogrel 75 mg) and symptoms gradually improved. The patient continued to take clopidogrel and atorvastatin orally after discharge. Follow-ups were conducted at least every six months post-discharge. The patient did not experience any further cerebrovascular incidents. Surgical resection of the hyoid bone was recommended for continuous mechanical stimulation, but the patient refused.

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Key words: carotid dissecting aneurysm, case report, contrast-enhanced computed tomography, hyoid bone

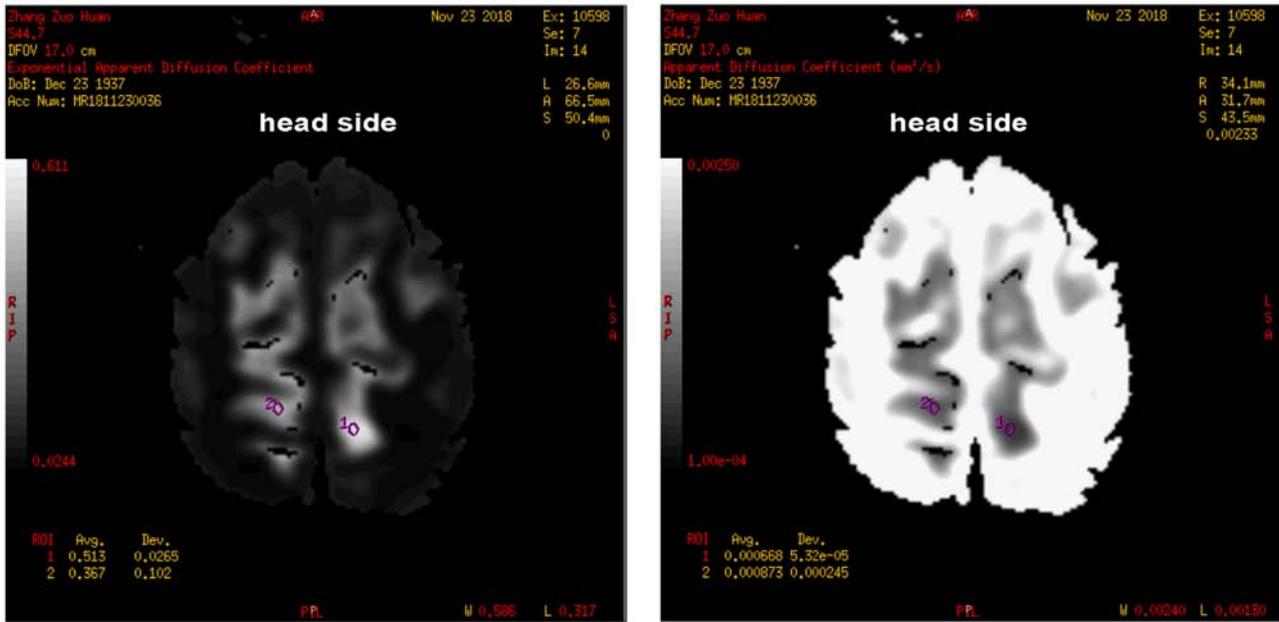


Figure 1. Diffusion-weighted imaging and apparent diffusion coefficient on admission showed acute infarction in the right parietal lobe.

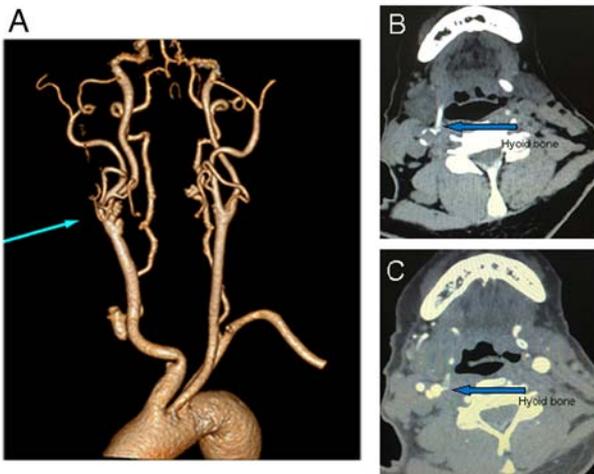


Figure 2. Contrast-enhanced computed tomography revealed two cysts with some calcification. (A) CTA with 3D reconstruction revealed two cysts with some calcification located at the bifurcation of the right ICA and stenosis of the origin of the right ICA. Computed tomography (B) without or (C) with contrast material revealed the greater horn of the right side of the hyoid bone adjacent to the right ICA. CTA, computed tomography angiography; ICA, internal carotid artery.

It is possible that infarction may have been caused by embolization from a thrombus from a dissecting aneurysm. Time-of-flight magnetic resonance angiography image (Fig. S1) demonstrated that a right-sided internal carotid artery lesion does have the potential to cause a left-sided cerebral infarct lesion.

Discussion

Bony structures around the carotid artery, such as the styloid process, thyroid cartilage (6) and hyoid bone, can cause dissection, compression, plaque formation and plaque rupture of

the carotid artery. In 1948, Eagle reported compression and irritation of the sympathetic plexus close to the ICA, resulting in dysphagia with facial and neck pain by elongated styloid process (7). This was further reported in other studies and alternatively called stylo-carotid artery syndrome or Eagle syndrome (8). Kumagai *et al* (6) described a case of symptomatic carotid stenosis caused by mechanical stimulation of the thyroid cartilage. Similarly, compression and trauma from the hyoid bone have also been described as rare causes of carotid stenosis, pseudoaneurysm and dissection. A possible explanation for carotid vasculopathy is the direct compression of the hyoid bone, leading to increasing turbulent blood flow and higher shear force, resulting in endothelial injury (9).

No clear history of head or cervical trauma has been noted in a majority of patients with carotid dissection. The exact cause of these so-called spontaneous dissections is unclear. Recently, certain anatomic characteristics of the hyoid bone (i.e., the length and the proximity to the carotid artery) were reported in only two studies as potential risk factors for carotid dissection (4,5). However, the case described in this study was perhaps the first case in which the elongation of the hyoid bone was diagnosed to be the cause of the dissecting aneurysm.

In the case described in the present study, dissecting aneurysm was detected accidentally because of no responsible ischemic strokes and no associated symptoms or signs such as neck pain, Horner syndrome, cranial nerve palsy and murmur of the carotid artery. No other causes of carotid dissection were found in this case. Computed tomography angiography (CTA) of the neck may characterize the anatomical association of the carotid arteries and the hyoid bone. It was hypothesized that a carotid dissecting aneurysm was related to direct mechanical compression of the carotid artery by the hyoid bone induced by neck rotation or swallowing. This diagnosis was confirmed by a dynamic study of carotid duplex ultrasonography. Swallowing, talking and head rotation were demonstrated to contribute to the hyoid-related vessel injury.

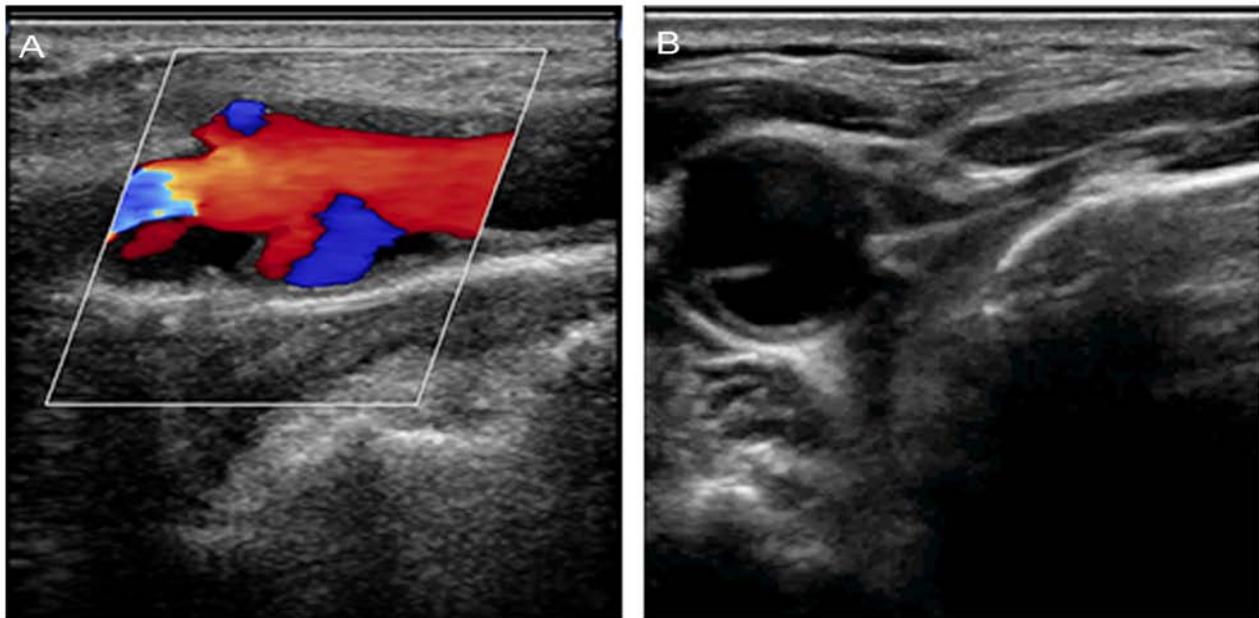


Figure 3. Color duplex scan of the carotid vessels. (A) Two cysts, in which red and blue flow signals were detected by carotid ultrasonography, communicated with the right CCA and the origin of the right ICA separately. (B) Right CCA and the origin of the right ICA were compressed by the hyoid bone repeatedly when swallowing and speaking. Video is available. CCA, common carotid artery; ICA, internal carotid artery.

No established treatment guidelines exist for hyoid bone-related carotid artery injury. Anticoagulation and/or antiplatelet treatment is usually initiated. At present, there is no established consensus regarding the treatment of this disease. As will be detailed subsequently, both anticoagulants and antiplatelet agents are viable options. Following a thorough discussion with the patient, we opted for antiplatelet therapy. If the hyoid bone is left untouched, it likely continues to cause mechanical stimulation. Although no responsible ischemic stroke was observed in the case reported in the present study, surgical resection of the hyoid bone was an appropriate treatment option. Partial hyoid resection is a safe and effective treatment in most case reports. Therefore, arterial reconstruction combined with partial hyoid resection was suggested. However, the patient refused to undergo this surgery and opted for oral antiplatelets.

In summary, despite only a few case reports of hyoid bone-related dissecting aneurysms, attention should be paid to the elongated hyoid bone as an underlying factor and treatable cause of dissecting aneurysm of the carotid artery. CTA of the neck and carotid duplex ultrasonography are useful treatment modalities. Partial resection of the hyoid bone is a safe and effective approach to prevent further ischemia.

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Availability of data and materials

All data generated or analyzed in this study are included in this published article.

Authors' contributions

QY, YL, HT, QZ and HZ collected patient data. QY and HL contributed to the study design and manuscript writing. QY and HZ confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

The ethics committee of The Affiliated Hospital of Southwest Jiaotong University and The Third People's Hospital of Chengdu (Chengdu, China) approved the present study (approval no. 20181123).

Patient consent for publication

The patient provided written consent for the publication of the case report and images.

Competing interests

The authors declare that they have no competing interests.

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