Multiple Strokes Associated With Elongation of the Hyoid Bone

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Abstract

Hyoid bone elongation is an uncommon cause of stroke. Here, we report a case of hyoid bone elongation causing localized trauma to the internal carotid artery, resulting in multiple strokes. A 32-year-old woman presented with unilateral weakness and history of a recent stroke. Imaging revealed the greater horn of the hyoid bone extending between the external and internal carotid with associated thrombus at the carotid bifurcation and acute stroke. Carotid ultrasound demonstrated movement of the hyoid bone in and out of the space between the external carotid artery and internal carotid artery with neck rotation. Treatment involved anticoagulation and partial hyoid bone resection. After resection, the stroke symptoms had not recurred in the patient. Hyoid bone-related carotid injury is an infrequent etiology of stroke, with no established treatment guidelines. Partial hyoid bone resection and antithrombotic therapy are likely a reasonably safe and effective treatment.

Keywords

stroke, hyoid bone, carotid artery diseases

Introduction

Hyoid bone elongation, causing compression or localized trauma to the carotid artery, is an uncommon etiology of stroke. The hyoid bone is a midline structure in the anterior neck below the mandible, above the thyroid cartilage and at the level of the third cervical vertebrae. It lies medial to the carotid artery and typically ends anteriorly and superiorly to the carotid artery bifurcation. The position of the hyoid bone changes with swallowing, talking, and neck rotation. Anatomic variations occur and elongation of the hyoid bone has been implicated in internal carotid artery compression,^{1,2} stenosis,³ dissection,⁴ and pseudoaneurysm.⁵ Here, we present a case of stroke related to thrombus in the internal carotid artery associated with an elongated hyoid bone.

Case Report

A 32-year-old woman presented initially to a local hospital with episodic left-sided numbness and weakness. She was diagnosed with right middle cerebral artery embolic strokes. She was 12 weeks pregnant, and hypercoagulability of pregnancy was felt to be her major risk factor for stroke. Aspirin 81 mg was started for stroke prevention and the pregnancy was terminated. Three months later, she presented to our institution with recurrent transient left-sided symptoms. Neurologic

examination demonstrated equal pupils without ptosis, decreased sensation, and mild weakness in her left arm. She denied neck pain.

Diffusion-weighted magnetic resonance imaging revealed small areas of acute infarction of the right frontal and parietal lobes. Computed tomography (CT) angiogram of the neck demonstrated a small thrombus in the right carotid bifurcation with extension into the proximal internal carotid artery. The greater cornu or horn of the hyoid bone was noted to be elongated bilaterally, with the right horn extending between the right external and internal carotid arteries (Figures 1 and 2).

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Figure 1. Computed tomography (CT) angiogram of the neck axial cuts. A, Left posterior hyoid bone elongation medial to carotid arteries. B, Right posterior hyoid bone elongation between the internal carotid artery and external carotid artery.

Figure 2. Coronal view of the right hyoid bone between internal carotid artery and external carotid artery. Demonstrates right superior skew of hyoid bone.

A workup for multiple causes of young adult stroke was unrevealing. This included testing hemoglobin A1c, thyroid-stimulating hormone, sedimentation rate, C-reactive protein, human immunodeficiency virus, rapid plasma reagin, anticardiolipin antibodies, beta-2-glycoprotein antibody, hexagonal phospholipid neutral, Russell viper venom test ratio, antithrombin III activity, prothrombin G20210A mutation, factor V Leiden mutation, homocysteine level, and urine toxicology. Urine pregnancy test was negative. Low-density lipoprotein cholesterol was 95. A transthoracic echocardiogram did not demonstrate thrombus, patent foramen ovale, or valvular disease. Telemetry demonstrated episodic bradycardia, but no atrial fibrillation. Her case was discussed with otolaryngology and vascular surgery for guidance in treatment options.

Based on the size and position of the hyoid, the elongated greater cornu was suspected of causing trauma to the internal carotid artery, leading to thrombus and subsequent embolic

Figure 3. Intraoperative duplex. A, Neck is neutral demonstrating hyoid in between the internal carotid artery and external carotid artery. B, Neck rotated to the left, the bone has exited and is no longer in between the vessels.

strokes. Given ongoing events, despite antiplatelet treatment, heparin was initiated and episodic left hemiparetic episodes did not recur on anticoagulation. She was discharged on anticoagulation with anticipated right hyoid bone partial resection. Prior to surgical intervention, a carotid ultrasound in neutral head position demonstrated the right hyoid bone extending between the external carotid artery and internal carotid artery. Neck rotation repositioned the hyoid bone out of this space (Figure 3). The patient was taken to the operating room for hyoid bone resection. During surgery, it was confirmed that the right greater horn was positioned between the internal and external carotid arteries and could be rotated in and out of this position with repositioning of the head. The right greater cornu was resected to the body of the hyoid without complications. Several weeks after resection, anticoagulation was stopped, and she was continued on monotherapy with aspirin 81 mg daily. Stroke symptoms have not recurred since resection. Consent was provided by the patient for the publication of this report.

Discussion

Rare cases of hyoid bone–related carotid artery disease have been described. Internal carotid artery pathology related to hyoid bone injury includes direct compression,^{1,2} stenosis,³ dissection,^{4,6} and pseudoaneurysm.⁵ Direct compression can result in increasing turbulent flow and higher shear forces, resulting in endothelial injury. Injury can lead to atheromatous plaque and clot formation, which can in turn cause arterial stenosis and artery-to-artery embolic ischemic stroke.^{1,3,7} A case-control study by Renard et al⁸ compared the distance between the internal carotid artery and styloid and hyoid bones in patients with or without carotid artery dissections. They found that the hyoid bone was anatomically closer to the internal carotid artery in the dissection group. It was concluded that anatomical factors likely play a role in carotid artery dissection. Repetitive or extreme head rotation has also been demonstrated to contribute to the hyoid bone–related

vessel injury.^{9,4} Elongation of the styloid process has also been implicated in carotid artery injury. A case-control study by Raser et al¹⁰ demonstrated an association between carotid artery dissection and longer styloid process length.

Our case demonstrates hyoid bone-related carotid artery clot and ischemic stroke. Our patient did not have evidence of focal carotid stenosis, but there is high suspicion that localized trauma caused endothelial injury leading to continued clot formation. One possibility is that a hypercoagulable state in the setting of pregnancy may have triggered her clot formation at the area of injury. One limitation in this case is that a magnetic resonance arterial imaging with fat suppression was not obtained and vessel dissection could not be confirmed. Carotid ultrasound demonstrated movement of the hyoid bone in and out of the space between the internal carotid artery and external carotid artery with neck rotation. There was no history of repetitive or extreme neck rotation. Routine neck rotation with hyoid bone movement between the carotid bifurcation was likely the cause of vessel injury. Several months after surgery, she could fully rotate her head laterally without eliciting any symptoms.

Hyoid bone-related carotid artery injury continues to be a rare etiology of stroke. There are no established treatment guidelines. Anticoagulation and/or antiplatelet treatment is usually initiated. In this case, partial hyoid bone resection proved to be a safe and effective treatment option. Hyoid bone elongation may be an important and underrecognized cause of young adult cervical internal carotid artery dissection and ischemic stroke.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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