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Hypoglossal nerve palsy due to internal carotid artery dissection with pseudoaneurysm formation: An unusual presentation

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Abstract

Internal carotid artery (ICA) dissection is a dangerous condition that results from disruption of the intimal part of the wall of the internal carotid artery. It is a rare disease that may occur spontaneously or as a result of a trauma. Spontaneous dissections of the carotid artery are rare but important causes of ischemic stroke because they usually affect young and middle-aged patients. Up to date, only a few cases were described in the literature about ICA dissection causing isolated cranial nerve palsies, with the Hypoglossal nerve being the most affected. Here, we report a case of a 56-year-old man presenting with progressive dysarthria, dysphagia to liquid diet, and difficult mastication. He was diagnosed as a case of cervical internal carotid dissection with pseudoaneurysm formation causing mass effect resulting in a compressive ipsilateral Hypoglossal nerve palsy based on magnetic resonance imaging (MRI) findings. Angiography confirmed the presence of dissecting pseudoaneurysm which was eventually managed by stenting. This case was reported to highlight and emphasize the importance of radiology, whether diagnostic or interventional, in managing rare and challenging cases such as ICA dissection.

Keywords

Internal carotid artery dissection, hypoglossal nerve palsy, radiology

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Introduction

Cranio-cervical artery dissection (CAD) is a potentially disabling condition caused by a tear on the arterial wall intima. This can lead to intrusion of blood into the wall of the artery forming an intramural hematoma. CAD is divided into spontaneous or traumatic dissection. Internal carotid artery (ICA) dissection is an uncommon disease with an estimated annual incidence of 2.5 per 100,000 to 3 per 100,000.[1] Spontaneous dissections of the Carotid and Vertebral arteries account for only about 2% of all ischemic strokes. Despite this rare prevalence, they are still considered important causes of ischemic stroke in young and middle-aged patients accounting for 10–25% of such cases.[2]

Some dissections can cause isolated cranial nerve palsies, with the Hypoglossal nerve being involved in most conditions.[3] In this article, a case of cervical ICA dissection

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with pseudoaneurysm formation causing a mass effect in the form of ipsilateral Hypoglossal nerve palsy was reported. This study aims to recall the importance of the role of the diagnostic aspects, as well as the interventional part, of radiology in managing such infrequent and challenging conditions.

Case presentation

A 56-year-old man, with a free past medical and surgical history, known to be a heavy smoker (about 20 pack-year) was transferred to our radiology department with a history that dates back to about one and a half months prior to presentation complaining of progressive difficulty of talking, difficulty of mastication and dysphagia to liquids. The patient did not have any history of trauma. On physical examination, he was found to have an atrophic left hemi-tongue, with deviation to the left side upon protrusion. His blood pressure was 127/75 and the body mass index (BMI) was 24. Routine laboratory investigations were unremarkable.

Magnetic resonance imaging (MRI) of the brain and neck, before and after the administration of intravenous (IV) Gadolinium, demonstrated a crescentic hyperintense area on unenhanced T1-weighted imaging overlying the cervical portion of the ICA, representing a false lumen of a dissection of the artery (Figure 1A). The finding was associated with narrowing of the true lumen of the vessel. In addition to that, there was an area of focal outpouching at the skull base, consistent with a pseudoaneurysm (Figure 1B and Figure 2), abutting the expected course of the Hypoglossal nerve at the exit of the hypoglossal canal. There was tongue denervation manifested by the presence of short tau inversion recovery (STIR) hyperintensity of the involved left hemi-tongue, enlargement of the ipsilateral base of the tongue, causing a protrusion into the oropharynx (Figure 3). No cerebral ischemia or intracranial hemorrhage was found at the encephalic level. Based on the clinical and radiographic findings, the patient was diagnosed as a case of dissecting cervical portion of the ICA, with pseudoaneurysm formation and compressive neuropathy of the Hypoglossal nerve.

Subsequent angiography was done confirming the presence of dissecting pseudoaneurysm (Figure 4A) which was eventually managed by stenting.

A follow-up carotid computed tomography (CT) angiogram study after 3 months showed normal caliber of the stented left cervical ICA with disappearance of the pseudoaneurysm (Figure 4B). The patient reported subjective partial improvement of difficulty of mastication and liquid dysphagia, with a total resolution of talking difficulty.

Discussion

Cranio-cervical artery dissection (CAD) is an incapacitating rare condition that mostly affects young and middle-aged adults.[4] The disease has become increasingly detectable due to the improved availability of MRI and the continuous upgrading in sequences, making a positive diagnosis more likely.[5] CAD may be traumatic or spontaneous. Spontaneous CADs, as the case reported in our study, are more common and they usually occur in healthy arteries with no identifiable etiological factors.[2] Spontaneous dissections of the Carotid and Vertebral arteries can affect all age groups including children, but the maximum incidence is in the fifth decade.[1] The peak is at 46 years, with no specific gender predisposition.[5]

A certain number of events have been associated with the onset of CAD like constitutional anomalies of the wall.

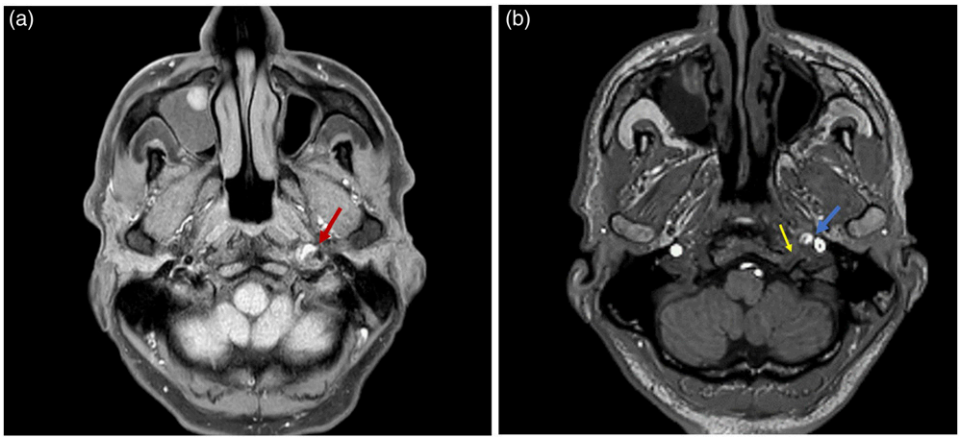

Figure 1. Axial T1-weighted fat-saturated image through the upper neck (image A) demonstrates a crescentic hyperintense rim (red arrow) surrounding the narrowed flow-related signal void of the left cervical ICA, likely representing subacute hemorrhage within the false lumen of a dissection of the internal carotid artery. There is focal dilation at this level (blue arrow) and the hypoglossal nerve canal is shown in yellow arrow (image B).

Examples of these anomalies include fibromuscular dysplasia or connective tissue diseases like Ehlers-Danlos and Marfan syndrome, polycystic kidney disease, osteogenesis imperfecta type I, infections, and transient vasculitis.[5,6] An elongated styloid process, which is called Eagle Syndrome, can also cause a spontaneous internal carotid artery dissection.[7]

The wide variation in signs and symptoms makes the diagnosis of carotid artery dissection challenging. Typically, a patient with carotid artery dissection presents with pain on one side of the head, face, or neck accompanied by a partial Horner's syndrome (oculosympathetic palsy), and followed hours or days later by cerebral or retinal ischemia. However, this classic triad is only found in less than one-third of the patients. Cranial nerve palsies can be detected in about 12% of patients with carotid artery dissections. The lower cranial nerves are the commonest to be affected, particularly the cranial nerve XII (Hypoglossal nerve) as described in this case. The Hypoglossal nerve provides motor innervation to the tongue. It arises from the ventral surface of the medulla before traversing the hypoglossal canal. On exiting the canal, the nerve runs beneath the ICA and the internal jugular vein. The relationship between the nerve and ICA explains how mural expansion may cause nerve compression. Vascular expansion does not exclusively occur as a result of the dissection itself. Sometimes it is the nutrient arteries which become occluded by vessel wall damage, or distal embolization.[3]

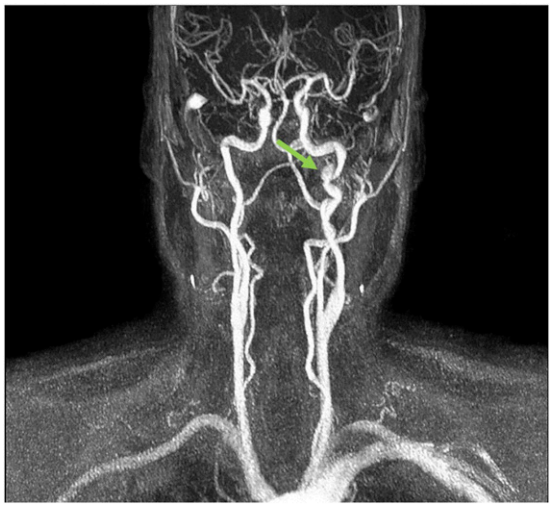

Figure 2. Coronal MIP reconstruction of MRA of the carotid arteries demonstrates a segmental area of narrowing of the left cervical ICA) with a focal area of aneurysmal dilation of the left internal carotid artery (green arrow).

In imaging, non-specific signs of dissection, such as stenosis or occlusion with hemodynamic repercussions, are frequently observed during Doppler examination and can help to reach the diagnosis. The main limitation of ultrasound resides in the inability to examine the entire vascular axis along its whole course because some regions are inaccessible to the ultrasound beam (sub- and intra-petrous ICA), leading to numerous false-negative results.[5] Evaluation via CT scan does not reveal any specific signs of CAD. Non-contrast brain CT scan is insensitive for dissection but may demonstrate cerebral ischemia or infarct. Arterial wall hematoma can be seen in the upper portion of the ICA as a crescent-shaped hyperattenuating focus. However, after injection of iodinated contrast medium, an enlargement of the dissected artery can be noted, in addition to an abnormal vessel with a narrowed eccentric lumen surrounded by a crescent-shaped mural thrombus and thin annular enhancement.[8] It can also show an intimal flap or dissecting aneurysm. A sub-intimal dissection tends to result in

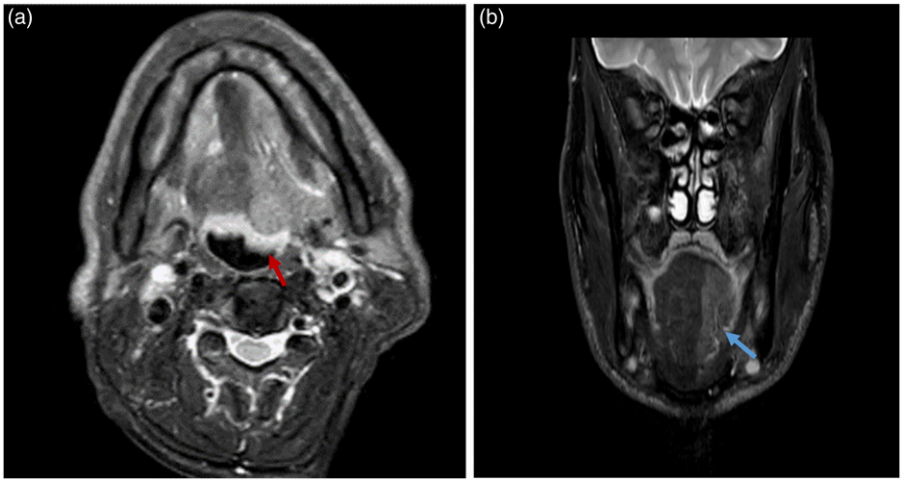

Figure 3. A well demarcated atrophy and STIR hyperintensity on the left hemi-tongue (blue arrow) and a dorsal bulge (red arrow) due to loss of muscle tone.

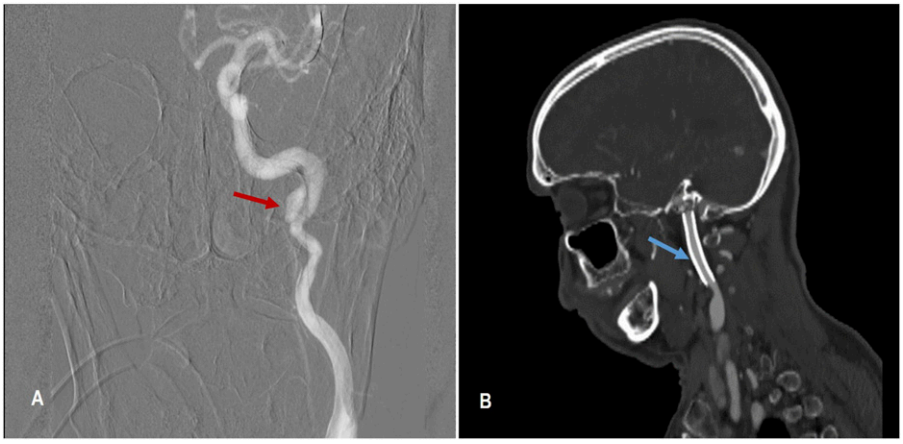

Figure 4. Digital subtraction angiogram AP (image A), confirming the presence of dissecting pseudoaneurysm of the left ICA (red arrow) and therapeutic stenting were applied. Follow-up carotid CT angiogram after therapeutic stenting (image B) showed normal caliper of the stented left cervical ICA with disappearance of the pseudoaneurysm of the left ICA (blue arrow).

stenosis of the arterial lumen, whereas a sub-adventitial dissection can cause an aneurysmal dilatation of the artery. Although such aneurysms are often referred to as "pseudoaneurysms," this is a misnomer because their walls are composed of blood vessel elements (media and adventitia).

When cervical ICA dissection is clinically suspected, the appropriate MRI protocol includes magnetic resonance angiography (MRA) of the neck to evaluate the lumen, and axial fat-suppressed T1-weighted images (T1 FS) to determine the presence of subacute intramural hematoma. Pathognomonic features of ICA dissection are intramural hematoma, intimal flap, and double lumen. All of these findings can be identified using vessel-wall (VW)-MRI.[9] The main advantages of vessel wall imaging over lumen-based methods are detection of non-stenotic lesions, as well as further characterization of stenotic lesions that have already been detected with common angiographic methods. In a recent meta-analysis, the reported frequency of intramural hematoma, vessel wall enhancement, aneurysmal dilatation, and intimal flap/double lumen sign differs, being, respectively, 86%, 75%, 71%, and 47%.[10] However, when dissection is not clinically suspected and therefore, MRI protocol is not tailored accordingly, subpetrous ICA dissection with intramural hematoma can still be detected on routine brain MRI sequences.[11] Other entities can mimic the radiographic findings of ICA dissection such as fibromuscular dysplasia, dysgenesis of the ICA, atherosclerosis, neck irradiation treatment, Takayasu's arteritis, Behcet's disease, and Giant Cell arteritis.[4]

Conventional angiography is the reference imaging technique for studying the arterial lumen but does not provide any information about the vascular wall. This technique does not enable the detection of the mural hematoma; therefore, it may be considered normal in the event of Carotid artery disease, especially if the latter does not cause any alteration on the arterial lumen.[5] This invasive and irradiating technique is not performed in first intention for the positive diagnosis of Carotid artery disease. Nowadays, it is only practiced for therapeutic purposes. It is indicated when the patient is symptomatic with ongoing ischemic events that are not well-controlled by medical management, in the form of narrow stenosis with hemodynamic repercussions or recurrent infarctions despite antithrombotic treatment.[12] In these cases, angioplasty, with or without stenting, may be proposed. In our case, the pseudoaneurysm formation and hypoglossal nerve palsy were the two main arguments that led to angioplasty with stent placement.

In conclusion, despite being rare, ICA dissection should be considered in the differential diagnosis of Hypoglossal nerve palsy; with nerve compression seems to be the most likely cause of palsy. When cervical ICA dissection is clinically suspected, MRI is the reference examination, enabling identification of the mural hematoma. Endovascular treatment including gentle angioplasty and stent placement may be of benefit in this aspect.

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