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CASE REPORT

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Isolated hypoglossal nerve palsy due to spontaneous internal carotid artery dissection: Two case reports and review of the literature

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ABSTRACT

We report on two patients who presented to the ear, nose, and throat clinic. Clinical examination demonstrated tongue deviation toward the left side, without other neurological symptoms. Imaging studies, neck computed tomography, and magnetic resonance angiograms revealed intramural hematoma, thickening, and stenosis of the right distal extracranial internal carotid artery consistent with spontaneous dissection.

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Spontaneous; internal carotid artery dissection; hypoglossal nerve palsy; MRI; CT; angiogram

Case 1 Report

A 58-year-old male presented to the ear, nose, and throat clinic complaining of tongue deviation to the left for 21 days. No history of head or neck trauma or other neurological deficits was detected. A neck computed tomography angiogram (CTA) was performed. Imaging revealed an asymmetry of the oropharyngeal airway secondary to right hemi-tongue atrophy and segmental wall thickening of the right distal extracranial internal carotid artery (ICA) with luminal narrowing and expansion of its diameter below the carotid foramen at the skull base, consistent with right internal carotid artery dissection (ICAD) (Fig. 1).

For a better characterization, contrast enhanced-magnetic resonance (MR) angiogram was performed, showing a reduction of the right ICA lumen, intramural high T1 signal, and expansion confirming dissection (Fig. 2).

Case 2 Report

A 76-year-old woman with slurring of speech with prior orofacial dystonia under Botox treatment. She noticed deviation of her tongue 2 years before but the slurring of speech began 6 months earlier. Her neurologist noted a right hypoglossal palsy.

Past medical history included dystonia, depression, diabetes, hearing loss, osteoarthritis, seasonal allergies, hyperlipidemia, bursitis, colonic adenoma, gastroesophageal reflux disease, obstructive sleep apnea, restless legs, obesity (BMI 30), neuropathy, and right hip replacement. She also developed daily blurred vision and dizziness. On exam, there was subtle slurring of speech. The tongue deviated to the right with a loss of bulk on the right hemi-tongue.

She underwent MR imaging of the head and neck (Figs. 3 and 4) and whole body fludeoxyglucose (FDG) positron emission tomography (PET)-CT. Imaging confirmed fatty atrophy of the right hemi-tongue and chronic dissection of the right ICA. No tumor was seen on FDGPET-CT.

Discussion

Hypoglossal nerve palsy (HNP) usually is caused by intra-extracranial expansive mass, infection, surgical or anesthesiology procedures, vascular

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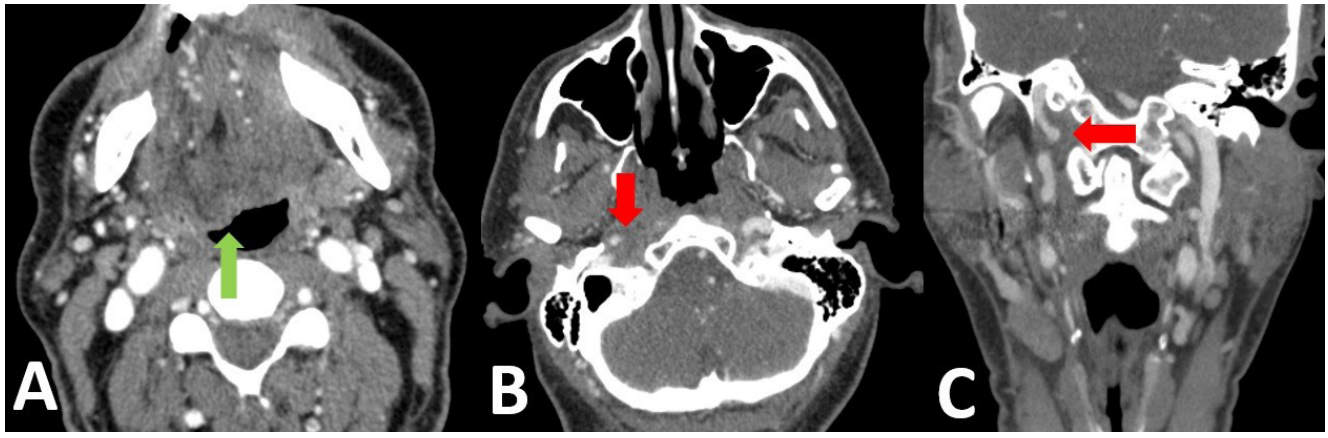


Figure 1. Neck CTA: (a) axial view shows bulbous protrusion of the right hemi-tongue into the oropharynx (green arrow); (b) axial view reveals wall thickening in the right ICA with luminal narrowing (red arrow); and (c) coronal view demonstrates wall non-enhancing intramural hematoma (red arrow) corresponding to the dissection of the right ICA.

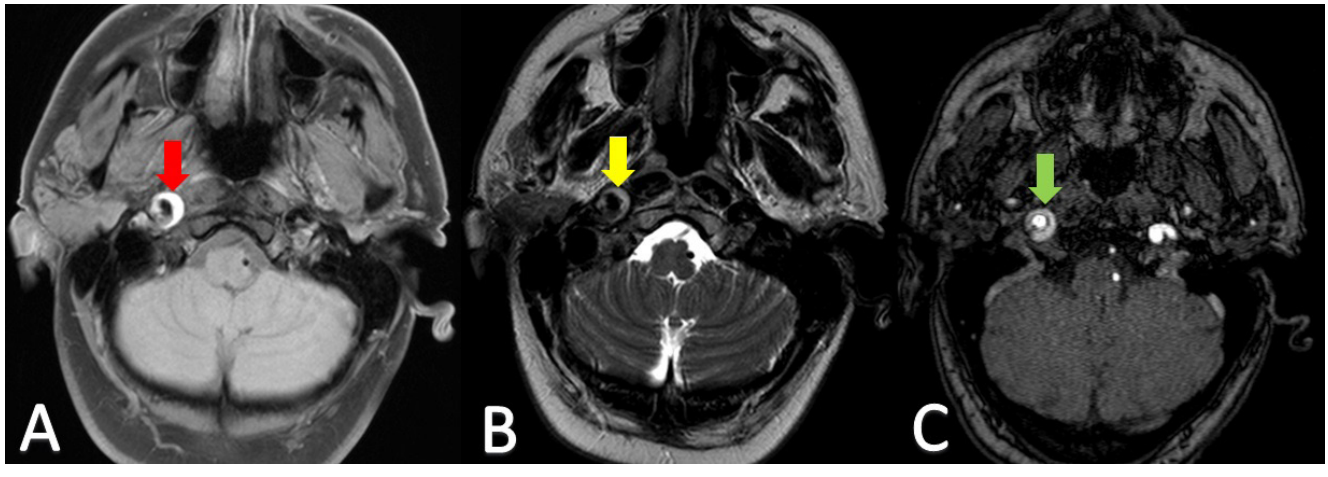


Figure 2. Magnetic resonance imaging. (a) T1-weighted fat-suppressed imaging demonstrates intramural high signal hematoma (red arrow) of the right ICA; (b) T2-weighted imaging reveals thickening (yellow arrow) of the right ICA; and (c) MR-angiogram shows right ICA stenosis (green arrow).

anomalies, trauma, motor neuron diseases, head and neck injuries, neuropathy, or autoimmune diseases. The idiopathic cause is very rare [1]. Clinically, it affects action extrinsic tongue muscles (genioglossus) resulting in tongue deviation toward the enervated side [2].

Spontaneous ICAD [3] has a relatively wide age range, and it is considered an important cause of acute cerebrovascular disease in young and middle-aged patients. Its classic manifestations include ipsilateral head and neck pain, Horner syndrome, and ischemic contralateral focal cerebral lesions [4]. Recognizing this clinico-radiological pathology is very important for specific treatment and reducing the risk of a stroke [5].

Arterial hypertension, hypercholesterolemia, diabetes mellitus, smoking, high body mass index, and migraine are mentioned as the risk factors predisposing to ICAD [4]. Other etiologies include, but are not limited to, autosomal dominant polycystic kidney disease, fibromuscular dysplasia, imperfect osteogenesis, estrogen–progesterone hormone therapy, ICA redundancy, infectious diseases, and connective tissue disorders [6]. In addition, heritable connective tissue disorders such as Marfan's syndrome and Ehlers–Danlos syndrome are associated with a high risk of spontaneous ICAD [7].

Stino et al. [8] analyzed a total of 245 cases of HNP. The most frequent etiologies included: postoperative (29.3%), idiopathic (15.1%), primary neoplastic (14.2%), metastatic (13.0%), inflammatory (7.3%), radiation-induced (6.1%), traumatic (4.1%), vascular (3.3%), congenital (2.8%), cystic (2.4%), and motor neuron disease-related (1.6%) [8].

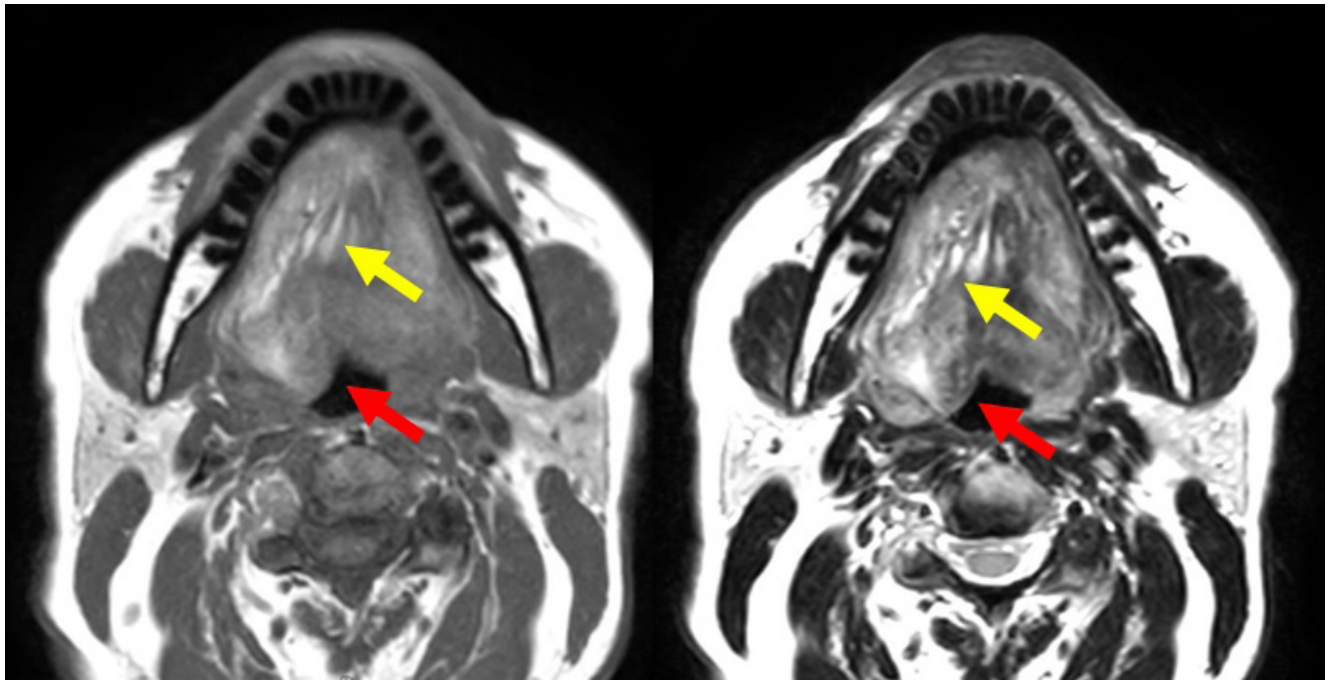


Figure 3. Axial T1 and T2 weighted images of case 2 displaying right hemi-tongue atrophy with fatty replacement (yellow arrows) and bulbous protrusion of the right hemi-tongue into the oropharynx (red arrows).

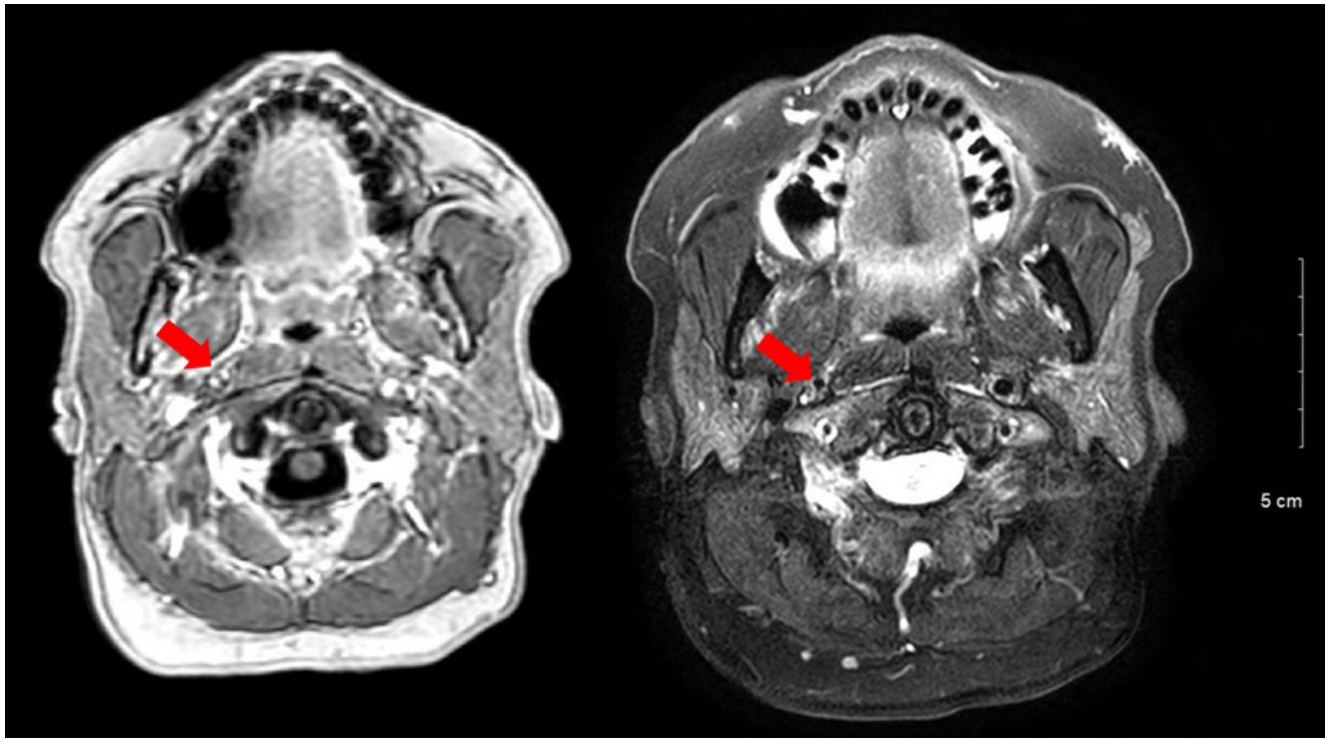


Figure 4. Axial T1 postcontrast and T2 fat sat displaying reduced caliber of the right ICA with linear low-intensity flap (red arrows).

ICAD rarely presents with HNP, but in some cases, it should be considered as the etiology of the damage to the lower cranial nerves including the glossopharyngeal (IX), vagus (X), spinal (XI), and hypoglossal (XII) nerves [9]. The mechanisms of nerve palsy could be a mechanical compression or stretching of the nerve by mural hematoma and nerve ischemia due to an impairment of the blood supply to the cranial nerve [6,7,10]. The hypoglossal nerve exits the skull through its own canal and

descends into the retro styloid space along with cranial nerves IX, X, and XI, the sympathetic chain, internal jugular vein, and ICA. The anatomical relationship between the XII nerve and ICA explains how mural expansion, due to ICAD in its higher extracranial portion, may cause nerve compression [11]. However, not all arterial dissections result in vascular expansion [12]. Case 1 had ICA expansion while case 2 did not, while both had ipsilateral hypoglossal nerve paralysis.

Therefore, nerve ischemia is a potential contributor to hypoglossal palsy at least in case 2. Hypoglossal nerve tumors are rare and are diagnostically challenging, especially in cases involving minor tumors [13].

Due to the subsequent risk of embolic ischemic stroke, ICAD requires treatment, especially within the first 2 weeks [7]. Drug treatment primarily consists of antithrombotic therapy (i.e., anticoagulant and antiplatelet therapies) [3]. However, after ICAD, there is no superiority of anticoagulation or antiplatelet therapy in the prevention of stroke [14]. In general, in more than 90% of cases, ICAD has a favorable evolution, with a good prognosis [9].

Finally, the differential diagnosis of an isolated hypoglossal cranial nerve lesion includes ICA aneurysm or thrombosis, tumors or trauma affecting the skull base, internal jugular vein thrombosis, and retropharyngeal masses [5].

Conclusion

Isolated HNP due to spontaneous ICAD is rare but has been reported. The relationship between the XII nerve and the ICA immediately below to carotid canal in the skull base explains how mural expansion may cause nerve compression, but nerve ischemia is another possible mechanism. CT and MR neck angiographies provide accurate mapping of vessels with Magnetic Resonance Angiography (MRA) being superior to CTA for demonstrating the vessel wall hematoma, important to confirm the clinical diagnosis of ICAD. It is important to recognize the association of ICA dissection with hypoglossal paralysis to avoid unnecessary further imaging in such cases.

Conflict of interest

None.

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