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Bilateral Duplicated Internal Carotid Arteries Presenting as Middle Ear Masses: A Case Report and Review of the Literature

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Abstract

Objectives—To describe an individual with bilateral duplicated internal carotid arteries (ICA) presenting as middle ear masses, to discuss the anatomy and characteristic imaging findings associated with this condition, and to familiarize clinicians with effective methods to prevent and manage complications related to this entity.

Methods—The clinical presentation of an individual with this unusual vascular anomaly was reviewed. A literature search was then performed to identify previously reported studies describing aberrant ICA in order to characterize the presentation, anatomy, imaging findings, and management of this condition.

Results—An aberrant ICA presenting as a middle ear mass is uncommon, as there are only approximately 45 reported cases to date. The majority of these cases presented as a unilateral anomaly without a duplication, were associated with pulsatile tinnitus and hearing loss, and were diagnosed during middle ear procedures. Bilateral aberrant ICAs are exceedingly rare, with only 14 existing reports. Only one of these cases presented with duplicated ICAs. Our report demonstrates an unusual presentation of aberrant ICA as pulsatile tinnitus was absent and previous middle ear surgery had been performed without establishing this diagnosis and without any resulting complications. Furthermore, this case represents the first known instance of a bilateral duplicated ICA system without persistent stapodial arteries. Although vascular middle ear anomalies are unusual, complications of surgical manipulation have been documented.

Conclusions—The clinician must have a high index of suspicion for vascular lesions in patients presenting with a retrotympenic mass. Appropriate diagnostic imaging studies should be performed to exclude this diagnosis prior to middle ear exploration. Should injury to the carotid artery occur, surgeons should follow specific guidelines to avoid potentially serious complications.

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Keywords

Carotid Artery; Vascular Anomaly; Duplication; Congenital Anomaly; Middle Ear

CASE REPORT

A 15-year old male was evaluated in our tertiary care pediatric otology clinic for evaluation of longstanding mixed left-sided hearing loss. His childhood was marked by recurrent acute otitis media for which he underwent bilateral tympanostomy at age nine months. The tubes had extruded and his tympanic membranes had healed uneventfully, however an underlying hearing loss had become evident after extrusion. He had been fitted with hearing aids since his preschool years which he had used consistently until presenting to our clinic.

At the time of evaluation, the patient denied tinnitus, vertigo, aural pressure, and had been performing well in school. His otologic and medical histories were unremarkable. Physical exam revealed bilateral reddish pale masses in both middle ear spaces that could be visualized through the anteroinferior quadrant of the tympanic membrane (Figure 1). A pure tone audiogram (Figure 2) showed a left-sided moderate to severe mixed hearing loss from 250–3000 Hz with borderline normal hearing from 4000 to 8000 Hz. There was an air-bone gap of 30 dB at the low and mid frequencies with fair to poor word recognition (56%). On the right, hearing thresholds and word recognition were normal. There was no interval change from previous audiograms.

The patient underwent high-resolution computed tomography (CT) of the temporal bones (Figure 3) and magnetic resonance angiography (MRA) (Figure 4). These studies revealed bilateral enlargement of the caroticotympanic and inferior tympanic arteries with anastomoses to the horizontal petrous portions of their respective internal carotid arteries. The vertical segments of both internal carotid arteries were present. The foramen spinosum was patent bilaterally and there was no evidence of a persistent stapedia artery. The diagnosis of bilateral duplicated internal carotid artery systems was made. The duplicated internal carotid artery systems were exposed in the middle ear and apposed to the undersurface of the tympanic membranes.

The patient and his parents were informed of the significance of these findings and were counseled on the risks of any procedures involving the middle ear space. He was instructed to continue wearing his left-sided hearing aid. The conductive nature of the patient's left sided hearing loss was attributed to abutment of the vessel against the ossicular chain. The etiology of the sensorineural component remains unclear, although hooding of the vessel over the round window may be a contributing factor.

DISCUSSION

Although an aberrant course of the internal carotid artery (ICA) is a rare phenomenon, it remains an important diagnosis to consider when investigating a middle ear mass. Establishing a correct diagnosis is paramount to avoid possible catastrophic hemorrhagic complications such as bleeding, hearing loss, or neurologic deficits (1). Anatomically, the ICA normally ascends vertically to enter the skull base anterior to the jugular bulb and tympanic canaliculus. The thin bony carotid plate of the carotid canal separates the ICA from the middle ear cavity and is usually < 0.5 mm thick. The ICA then turns anteromedially to enter the cranial cavity superior to the foramen lacerum (2). The blood supply of the middle ear is derived from anastomoses between the external carotid artery (ECA) and ICA. The inferior tympanic artery emerges as a branch off of the ascending

pharyngeal artery and travels vertically to enter the tympanic canaliculus with Jacobson's nerve. It then anastomoses on the promontory with the caroticotympanic artery which passes through a small dehiscence in the carotid plate to join with the horizontal segment of the ICA (3). An aberrant course of the ICA is often associated with an absence of the vertical segment of the ICA in the carotid canal. Lasjaunias hypothesized that decreased or absent blood flow during embryological development through the vertical segment of the developing carotid canal would result in failure of that vessel segment to develop. An alternate circulation pathway from the external carotid system would then develop to bypass this segment, with the anastomosis of the inferior tympanic artery to the caroticotympanic artery in the middle ear serving as the alternate route (4). An altered course of a normally established carotid system has also been reported in several instances in the literature, with the carotid artery being displaced posteriorly and laterally into the middle ear space through a dehiscence in the carotid plate. Both embryologic (persistent vasculature providing posterior traction on the ICA, congenital dehiscence of the carotid plate) and acquired (trauma, cholesteatoma) etiologies have been proposed as mechanisms to account for these occurrences (5).

Conservative management of an aberrant ICA is generally advocated, and middle ear surgical procedures are avoided. Incidentally our patient had tympanostomy tubes placed early in life, fortunately without complication. Excellent communication must be established with all physicians involved in the patient's care to convey the significance of this finding. In situations where debilitating tinnitus or chronic middle ear infections are present, Ruggles and Reed describe covering the artery in the middle ear with a fascia graft followed by a bone graft and then an additional superficial fascia graft and then positioning the artery to a more normal anatomic position through an existing bony dehiscence (6). If a middle ear mass is encountered during an operative procedure, attempts at diagnosis such as angiogram should be made prior to performing a biopsy of the lesion. The lesion can also be aspirated with a 27-gauge needle and blood gas analysis can be performed on the aspirate to confirm an arterial source (7). If bleeding is encountered during surgery, the middle ear should be packed with Surgicel or Gelfoam, a neurosurgery consult should be obtained, and an angiogram should be performed to assess the adequacy of cerebral blood flow provided by the contralateral side. The patient should be followed closely and if bleeding occurs despite conservative management, embolization using balloons or coils may be attempted and is preferred over ligation of the ICA in the neck when possible (1). Once the patient is discharged, regular follow up should be performed.

In conclusion, there must be a high index of suspicion for vascular lesions in patients with retrotympanic masses. The presence of a vascular lesion should be ruled out using high resolution CT, MRA, and, if needed, diagnostic angiogram prior to middle ear exploration. Our case represents an unusual presentation of aberrant ICA as prior middle ear surgery had been performed without establishing a diagnosis and without any resulting complications. To the authors' knowledge, only one other case describing bilateral duplicated ICAs has been reported in the literature, and this case was associated with persistent stapedia arteries (8). Our report therefore represents the second reported instance of duplicated ICAs and is the only known example of duplicated ICAs not associated with persistent stapedia arteries.

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Figure 1. Photograph of the patient's left ear revealing a pale red retrotympanic mass best visualized in the anteroinferior quadrant (arrow). This finding was also observed on otoscopic exam of the patient's right ear.

Figure 2.

Pure tone audiogram showing a left-sided moderate to severe mixed hearing loss from 250 to 3000 Hz with borderline normal hearing from 4000 to 8000 Hz. A 30 dB air-bone gap is present at the low and mid frequencies. This patient displayed fair to poor word recognition (56%) on the left. On the right, hearing thresholds and word recognition were normal. Immittance testing was normal bilaterally. X = Left ear air conduction; O = Right ear air conduction; J = Bone conduction.

Figure 3.

Axial (A, B) and coronal reconstruction (C) CT images of the left temporal bone show duplication of the left internal carotid artery. There is a normal vertical petrous carotid canal (A,*). The aberrant internal carotid artery represents enlargement of the inferior tympanic and caroticotympanic arteries. At the level of the skull base, the inferior tympanic artery passes through the inferior tympanic canaliculus within the caroticojugular spine (A, arrow), which is normally imperceptible. This vessel then anastomosis with the caroticotympanic artery (B, arrow) within the middle ear cavity over the cochlear promontory. The caroticotympanic artery in turn passes through a dehiscence carotid plate to anastomose with the petrous portion of the internal carotid artery. Notice the close proximity of the aberrant vessel to the ossicular chain and tympanic membrane (C, arrow).

Figure 4.

Axial source (A) and 3 D volume rendered MRA (B) images show duplication of the internal carotid arteries bilaterally with the medial vessels representing the normal internal carotid arteries (long arrows) and the lateral vessels representing the aberrant vessels (short arrows). These vessels anastomose just proximal to the horizontal petrous segment. Notice the normal position of the middle meningeal arteries with no evidence of a persistent stapedial variant (arrowheads).