

case report

Unusual presentation of internal carotid artery aneurysm in the middle ear

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Ann Saudi Med 2007; 27(4): 293-295

An aberrant course of the internal carotid artery (ICA) through the middle ear is uncommon. Approximately one percent of the population presents with a dehiscant ICA canal. Embryological malformation of the first and second branchial arches would prevent the formation of the ICA bony covering over the tympanic part of the middle ear.¹ These abnormalities are usually asymptomatic and often diagnosed during ear surgeries.² We report a case of unilateral ICA aneurysm in the middle ear. The literature is reviewed and the diagnosis and risks in clinically treating this anomaly are discussed.

CASE

A healthy 25-year-old Saudi female presented to the Otorhinolaryngology, Head and Neck Section of King Abdulaziz Medical City, Jeddah, Saudi Arabia, with left-side hearing loss and no record of otorrhea, tinnitus, vertigo, hemorrhage or otalgia. There was a history of suppurative bilateral chronic otitis media. The patient had undergone a left canal wall-up tympanomastoidectomy three years previously. This same procedure was done two years later to the right side without intra- or post-operative complications.

Otосcopy revealed a dry central residual tympanic membrane perforation in the left ear and an intact graft in the right ear. There was no facial weakness, nystagmus or other ear, nose, and throat abnormalities. Rinne's test was negative bilaterally and Weber's test was lateralized to the left ear. An audiogram showed bilateral mild-to-moderate conductive hearing loss more on the left side with an air bone gap of 20 decibels (dB).

The patient was booked for left revision tympanoplasty. During the operation, after elevation of the tympanomeatal flap, adhesions between the middle ear mucosa and a previously inserted silastic sheet were found. After releasing the adhesions and removing the sheet, severe bleeding was encountered, raising the suspicion of a dehiscant jugular bulb. The middle ear was tightly

packed with a *Surgicel* absorbable hemostat, the external auditory canal packed with non-absorbable gauze, and the procedure was aborted. The patient was hospitalized for one week. During this period there were no further episodes of bleeding. The patient was discharged with a new external ear pack, which was to be changed again at a 7-day follow-up visit. Five days after discharge the patient presented to the emergency room with bleeding from the left ear, tachycardia, a normal blood pressure and her hemoglobin had dropped from 13 to 9 g/dL. The external ear pack was extruding and another pack was applied to control the bleeding. The patient went into hypovolemic shock, was resuscitated, and admitted to the intensive care unit (ICU).

The patient developed right hemiparesis and underwent CT imaging the next day with enhancement for the brain and the temporal bone which showed a bilateral intact jugular bulb, a bony rarefaction around left petrous carotid, and a small brain infarction in the left temporal (Figure 1). On the third day an angiography was performed, which confirmed the presence of an aneurysm in the left ICA through the hypotympan-



Figure 1a. Axial CT of the temporal bone shows the aneurysm of the internal carotid artery in the left middle ear.

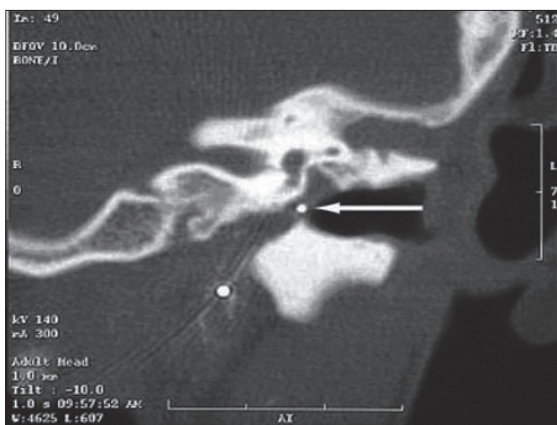


Figure 1b. Coronal CT of the temporal bone shows anomalies around the left petrous carotid artery.

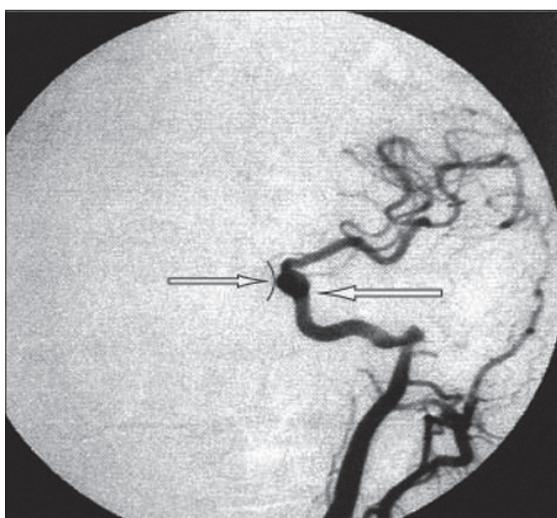


Figure 2. Angiography of the carotid artery shows an aneurysm of the internal left middle ear.

num (Figure 2). Two days later in the ICU, the bleeding stopped, the weakness improved and the patient was transferred to the ward. After six days, the aneurysm was successfully embolized with detachable platinum coil fillings and the patient recovered neurologically.

DISCUSSION

Anomalies of the ICA are very rare and the incidence is difficult to determine. Glasscock et al suggested an incidence of one percent of the population.³ Cohen and Briant considered the incidence of bone dehiscence over the ICA to be considerably less than reported.^{4,5} A review of the literature showed some case reports involving children and adults. Only 42 clinical cases of an aberrant ICA in the middle ear had been reported

before 1991. Most cases of intratympanic ICA occurred in women (female:male ratio, 5:1), often occurring unilaterally on the right side (female:female ratio, 2:1). However, it has been reported bilaterally.⁶ The present estimate is about 50 published cases.

Normally, the ICA enters the petrous bone medial to the styloid process via the carotid canal, separated from the internal jugular vein by the carotid ridge. The initial vertical segment is anterior to the cochlea and separated from the tympanic cavity by a plate of bone approximately 0.5 mm thick that was possibly cribriform in childhood and may reabsorb in old age. The ICA then turns anteriorly to lay inferior and posteromedial to the eustachian tube, traverses the foramen lacerum, and enters the middle cranial fossa.^{2,6}

The precise causes of an aberrant ICA in the tympanic cavity remain unclear.² Anomalies of the ICA within the temporal bone include aneurysms and arterial displacement, and both can be either congenital or acquired.⁷ Congenital aneurysms are thought to originate from a persistent stapedia artery or the abnormal persistence of embryonic vasculature that may supply enough traction to pull the ICA into the tympanic cavity. This traction may pull or fix the ICA laterally into the middle ear. Proposed acquired causes include head trauma, previous surgery, atherosclerosis, chronic infections, cholesteatoma and malignancies.^{6,8}

ICA anomalies in the middle ear represent a diagnostic problem since both symptoms and signs are non-specific. Patients may be asymptomatic or complain of pulsatile tinnitus, hearing loss, otalgia or aural fullness. Otoscopy may show a vascular bluish-red mass in the anteroinferior mesotympanum—the “rising sun sign”. An audiogram usually shows conductive hearing loss while impedance testing may reveal a pulsating tympanic membrane.^{1,2} These symptoms and signs are not distinct to ICA aneurysm and could signify vascular anomalies in the middle ear space (Table 1).^{6,9} Anomalies of the ICA have a typical appearance on a high resolution CT scan: an enhancing intratympanic mass, with the vessel lying lateral to its normal position, the absence of the bony plate between the hypotympanum and the ICA, and the jugular fossa is separated from the carotid canal by a spur of bone.¹⁰

Even though it has been suggested that a CT scan alone is sufficient to confirm the diagnosis, it does not always differentiate between a vascular anomaly and a high vascular glomus tumour.¹¹ Conventional carotid angiography remains a standard reference for diagnosis as it shows the lateral displacement of the vertical segment of the ICA in the anterior/posterior view; it is important in the assessment of vascular supply and

Table 1. Differential diagnosis of a vascular mass in the middle ear.

- Aberrant ICA
- Petrous ICA with aneurysm with dehiscence bone
- Dehiscent jugular bulb
- Glomus tympanicum
- Glomus Tumours
- Cholesterol granuloma
- Persistent stapedia artery
- Other vascular temporal bone lesions

the confirmation of the presence or absence of an aneurysm.¹

Lapayowker et al described the “vestibular line”, which is a vertical line drawn tangential to the lateral most extent of the vestibule.¹² This line indicates the lateral limit of the normal intratympanic ICA. If the artery lies lateral to this line it is considered aberrant. These features can be demonstrated by the less invasive magnetic resonance angiogram (MRA), suppressing the need for conventional angiography.¹³ The limitation of MRA relates to the reduced flow of such large aneurysms, and the signal intensity is reduced with limited visualization.¹⁴

The best treatment of aberrant or aneurysm of the ICA is to avoid manipulation, informing any physi-

cian involved in the patient's care of this precaution.^{5,6} However, in the case of chronic middle ear infection, surgical techniques to restore the middle ear cavity and to cover the aberrant vessel permanently with a bone graft are advocated by Ruggles and Reed.¹⁵ The surgeon should be aware of the risks of hemiparesis, temporary aphasia, permanent hearing loss or even death. In case of iatrogenic injury during myringotomy or middle ear surgery, immediate treatment should consist of packing of the middle ear with absorbable material such as *Surgicel*, then tight packing of the external canal to control the bleeding. The surgeon should be careful not to damage the middle and inner ear structures as a result of haphazard pack placement or complete occlusion of the ICA by too firm packing, which can lead to neurological sequelae. Additional steps to control hemorrhage include a posterior epistaxis pack or balloon, ICA ligation and balloon occlusion at the middle ear level under EEG monitor as the last resort.² The treatment for a confirmed aneurysm would be embolization by filling the aneurysm with detachable platinum coils.¹⁶ The patient should have regular follow up after hemostasis.

Awareness of the existence of vascular anomalies in the middle ear, although they are uncommon, is essential. Any abnormal clinical or radiological findings should be considered suspect. Failure of accurate diagnosis could have disastrous consequences. The preferred method of treatment is avoidance of middle ear manipulation.

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