■ CASE STUDIES

Abnormal Internal Carotid Artery in the Middle Ear and Severe Otorrhagia After Myringotomy

Jorge L. Merán Gil, Elisabeth Masgoret Palau, Francisco J. Avilés Jurado, Esther Domènech Vadillo, Enric Figuerola Massana, María D. Martínez Novoa, and Juan C. Flores Martín
Servicio de Otorrinolaringología y Patología Cérvico-Facial, Hospital Universitari Joan XXIII, Tarragona, Spain

We present a clinical-surgical case of a 72-year-old woman referred from another hospital due to presentation of otorrhagia following myringotomy. We later determined by means of imaging studies that it was a case of an aberrant internal carotid artery. All masses in the middle ear, especially pulsing masses, must be studied by imaging methods such as computerized tomography and, preferably, magnetic resonance angiography.

Key words: Otorrhagia. Aberrant carotid artery. Myringotomy.

Arteria carótida aberrante. A propósito de un caso
Los autores presentan un caso clínico-quirúrgico de una mujer de 72 años de edad, remitida desde otro centro hospitalario por una otorragia grave tras una miringotomía. Mediante estudios de imagen se determinó que se trataba de un caso de arteria carótida aberrante de oído derecho. Toda masa en oído medio, especialmente las que son pulsátiles, deben ser estudiadas mediante pruebas de imagen como tomografía computarizada y, preferentemente, angiografía por resonancia magnética.

Palabras clave: Otorragia. Arteria carótida aberrante. Miringotomía.

INTRODUCTION

Aberrant courses of the petrosal vessels are extremely rare; as a result, to date only a mere 50 cases of aberrant internal carotid artery (AICA) in the middle ear have been reported. In 1899, Marx was the first to describe an aberrant carotid artery in the middle ear.[1] The internal carotid artery (ICA) may exhibit 4 variants: *a)* normal location, albeit exposed, lacking bony cover; *b)* congenital vascular abnormality secondary to regression of the cervical ICA during embryogenesis, exposed, and following an abnormal course; *c)* aneurysm with protrusion toward the middle ear; and *d)* aberrant course.[2]

The most common symptoms presented by patients with AICA tend to be pulsating tinnitus, conductive hearing loss, and earache, although it must be pointed out that many of these patients may remain asymptomatic. Otoscopy may reveal a normal tympanic membrane with a mass in the middle ear that, in some cases, is pulsating. Imaging studies are fundamental for diagnosis; high resolution computerized tomography (CT) is the technique of choice and, in some cases, can be complemented with a magnetic resonance (MRI) angiography.[3]

CASE STUDY

We report the case of a 72-year-old female referred from another hospital due to severe otorrhagia following myringotomy.

Of interest in her personal history we would highlight untreated hypergammaglobulinaemia, monitored by the haematology department, cardiac arrhythmia due to atrial fibrillation treated with Sintrom®, mild aortic insufficiency, and allergic rhinitis.

According to the medical report sent by the referring hospital, in the context of recurrent otitis media, the patient was scheduled to undergo surgical myringotomy under sedation. The surgical report specifies that, when the tympanic membrane of the right ear was punctured, severe otorrhagia resulted that required anteroposterior packing of the ear and nose with gauze and intubation. The patient was transferred to our centre, where she was admitted to the ICU and kept there for 4 days until discharge once her general status had improved significantly.

The CT performed revealed a difference in diameter between the 2 ICA along their course through the carotid canal (Figure 1A), as well as a round mass of soft tissue that continued along the carotid canal and projected into the

Correspondence: Dr. J.L. Merán Gil.
Ernest Vilches, 1C, 5.º, 1.ª. 43006. Tarragona. España.
E-mail: orltarragona@hotmail.com

Received March 12, 2007.
Accepted for publication July 13, 2007.

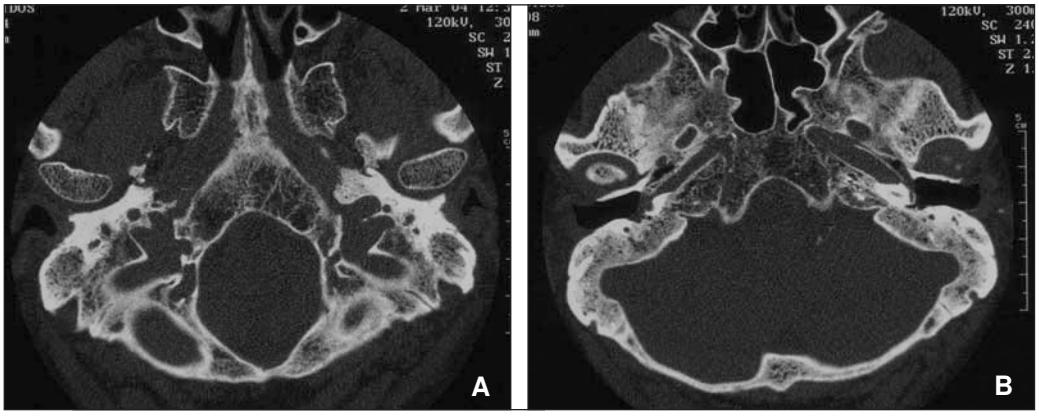

Figure 1. Computerized axial tomography. A: asymmetry in the course of both carotid arteries. B: absence of part of the bony wall of the carotid canal that projects toward the right middle ear.

anterior part of the lower region of the tympanic cavity (Figures 1B and 2).

An MRI was then performed and revealed the packing material in the external auditory canal and haemorrhagic occupation in the middle ear.

Audiometry performed 10 days after puncture reveals bilateral mixed hearing loss predominantly on the right side with an ADT of 20 dB HL (Figure 3).

On the basis of the radiological findings and the history of haemorrhage resulting from the prior myringocentesis, a definitive diagnosis of aberrant internal carotid artery in the middle ear was made.

DISCUSSION

To date, fewer than 50 cases of aberrant carotid artery have been reported. More than 90% of the cases described in the literature were found in females and most involved the right side. No convincing explanation has been put forth for this predominance in females and on the right.

In 9 of these cases, the ICA lesion was the result of myringotomy and 5 of these 9 patients developed pseudoaneurysm. In fact, only 24 cases have been reported as involving the area of the pars petrosa.[4]

Ordinarily, the vertical portion of the carotid artery runs upward, is covered by a 0.5-mm bony wall, and enters the carotid aperture, which is medial and anterior to the styloid process in the petrous bone. It then curves and the horizontal portion passes obliquely frontward and inward with respect to the axis of the petrous bone, passes underneath the Eustachian tube and the cochlea, and reaches the endocranial orifice (foramen lacerum).[5,6]

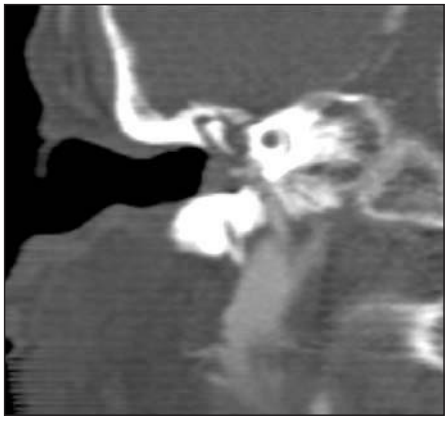

Figure 2. Coronal computerized tomography reconstruction: occupation in the middle ear by the aberrant course of the internal carotid that enters the hypotympanum contiguous to the cochlea.

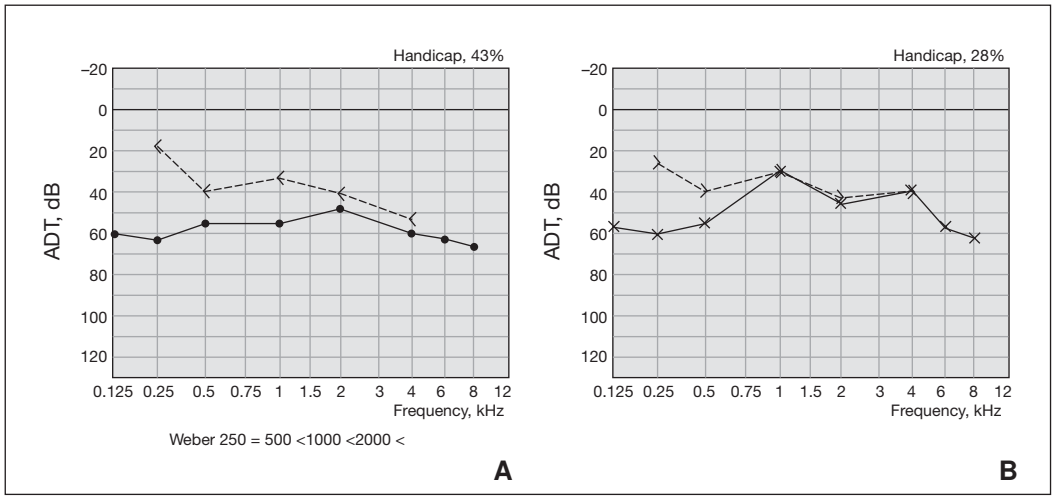

Figure 3. Pure tone audiometry: moderate, bilateral, mixed hearing loss predominantly on the right with a transmissive component with a 20-dB ADT.

There are 3 hypotheses as to the origin of an aberrant ICA:

1. The absence of the bony wall along its vertical portion that allows lateral displacement of the artery to take place.

2. The arterial vessels present during embryonic development fail to disappear, remaining as vestiges of the stapedial artery that would pull on the ICA bringing it into an anomalous position.

3. Sinnreich's hypothesis, the most widely accepted theory, that proposes dehiscence of the intrapetrous portion of the CA and absence of its vertical segment, either by involution or agenesis, resulting in an alternate vascular pathway toward the horizontal portion of the ICA.[7]

In 30% of cases, the AICA is associated with an aberrant stapedial artery.

Ear pain has been reported to be one of the first symptoms. Other signs and symptoms include tinnitus, conductive hearing loss, vertigo, and the sensation of having a plugged ear. Tinnitus may be due to direct mechanical transmission of pulses from the vessel to the tympanum and to the ossicles, or to the audible sound produced by arterial blood flow through the abnormal vessel inside the middle ear. The hearing loss may be due to the vessel's mass effect (softening of the vibrations of the tympanum or pressure, or erosion on the ossicles). Many patients remain asymptomatic.[8]

The main CT findings are, as we have seen in our case, a soft tissue mass in the middle ear (Figure 2), abnormal position of the carotid canal, visualization of the abnormal course along the lateral and posterior portion, and the continuity of this canal with the mass in the middle ear.

These findings alone suffice to establish the diagnosis, rendering angiography unnecessary, thereby avoiding the possible angiography-derived neurological complications that occur in 2.6% of the cases, with 0.3% of permanent damage.[3,9]

Conventional MRI does not reliably identify AICA; better results are obtained by means of angiographic MRI, which does reveal flow-dependent structures, without constituting an invasive testing procedure.[3]

The differential diagnosis should include a high jugular bulb with bony or dehiscent coverage, glomic tumours (tympanic, jugular, or both), arteriovenous malformations, persistent stapedial artery, cavernous angioma, and some cases of cholesterol granuloma that give rise to a "blue eardrum."

A correct diagnosis would avoid serious complications, such as perforating a mass of this type in the middle ear, as occurred in this case.

The best treatment following the diagnosis of AICA is to avoid manipulation and inform the patient of the risk it entails. Immediate treatment following accidental puncture consists of packing the auditory canal to control bleeding, leaving the packing in place for several weeks so as to avoid recurrent bleeding.

Additional steps to control bleeding include posterior nasal packing, ligature of the ICA, and balloon occlusion. Nevertheless, interruption of blood flow through the ICA is not generally necessary.[10]

Vascular abnormalities in the middle ear are rare; however, given the associated clinical relevance and possible complications, they represent a diagnostic and therapeutic challenge.

A correct diagnosis will avoid serious complications caused by attempting to puncture or excise this type of mass in the middle ear as Lapayowker et al[11] and Goodman et al[12] indicated. This is the basis for deciding to adopt a wait-and-see attitude with tomography and the importance of instructing patients and their relatives on the nature of the disease, as well as the potential risk involved in confusing it with ototubaritis or otitis media with effusion and puncturing it, which can be life-threatening for the patient. Special care must also be taken to avoid possible trauma or acute or chronic infection of the ear that may injure this kind of vascular abnormality.

REFERENCES

1. Biller JA, Linthicum FH Jr. Aberrant carotid artery. Otology and Neurotology. 2002;23:407-8.
2. Schuknecht HF. Pathology of the Ear. Philadelphia: Lea & Febiger; 1993. p. 672.
3. Hernández Valencia G, Solís Rábago L, González González L, Cano Bonilla.A. Arteria carótida aberrante en oído medio. Acta de Pediatr Mex. 2004;25: 262-8.
4. Figuerola E, Gómez E, Teruel J, Sánchez JM, Llano A, Rosell R. Carótida interna aberrante en oído medio. Acta Otorrinolaring Esp. 1989;40:47-50.
5. Jürgen G, Fradis M, Schipper J. Aberrant internal carotid artery in the middle ear. The Annals of Otology, Rhinology & Laryngology. 2001;Sep:110-9.
6. Phelps P, Lloyd G. Vascular masses in the middle ear. Clin Radiolog. 1986;37:359-64.
7. Selesnick SH, Lessow AS. Aberrant internal carotid artery. Am J Otology. 1999;20:403-4.
8. Hasso A, Broadwell R. Anomalías congénitas. In: Som P, Bergeron R, editors. Radiología de cabeza y cuello. Madrid: Mosby; 1993. p. 976-8.
9. Teruel J, Gómez E, Figuerola E, Castañer F, Ferrer J, Pont J, et al. Diagnóstico mediante tomografía computarizada de masas vasculares de oído medio. Radiología. 1990;32:407-11.
10. Ridder, GJ. Aberrant internal carotid artery in the middle ear. Annals of Otology, Rhinology & Laryngology. 2001;110:892-4.
11. Lapayowker MS, Liebman EP, Ronis ML, Saber JN. Presentation of the internal carotid artery as a tumor in the middle ear. Radiology. 1971;98:293-7.
12. Goodman RS, Cohen NL. Aberrant internal carotid in the middle ear. Ann Otol. 1981;90:67-9.