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Isolated agenesis of the mastoid antrum

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Abstract
The first isolated case of agenesis of the mastoid antrum, previously only described in association with the congenital syndromes trisomy 13 and mandibulofacial dysostosis, is reported. The loss of this important surgical landmark may result in disorientation and iatrogenic trauma. The surgeon must be aware of its existence, and where it is suspected the middle fossa dura should be exposed and followed posteriorly until the lateral sinus is encountered.

Key words: Mastoid, antrum, agenesis

Introduction

The antrum is an important landmark in the surgical approach to the mastoid. Absence of the antrum has previously been reported in association with other congenital otological abnormalities in the syndromes of trisomy 13 and mandibulofacial dysostosis. We report the first case of an isolated absent antrum in a healthy adult.

Case report

A healthy 66-year-old male presented to the Department of Otorhinolaryngology with a 12-year history of an intermittently discharging left ear. Examination revealed a large central perforation, deep posterior attic retraction pocket and inflammation of the middle ear mucosa. Pure tone audiometry showed a 60 dB mixed hearing loss across the frequency range.

Conservative treatment failed and the mastoid was explored post-aurally. The dura was encountered early and sank low into the mastoid below the level of the superior rim of the bony external auditory canal. Only a thin plate of bone separated the superior bony meatus from the middle fossa dura (Figure 1), which was followed posteriorly until the lateral sinus was identified. No cholesteatoma was found. The retraction pocket was excised and the tympanic membrane perforation repaired with temporalis fascia.

A subsequent CT scan has shown the right mastoid antrum to be present. Three months later the left tympanic membrane remains intact and healthy.

Discussion

The tympanomastoid compartment appears in the third week of embryonic life as the tubotympanic recess; an outgrowth of the terminal end of the first pharyngeal pouch (Hammer, 1902). From the 12th week four sacs bud from the recess; anticus, posticus, superior and medius, and expand to pneumatize the middle ear and epitympanum (Proctor, 1964). By the 21st week pneumatization has reached the antrum which is a posterolateral extension of the epitympanum (Allam, 1969). The antrum and the mastoid cells are not completely formed until after birth and expand throughout infancy and childhood (Bast and Anson, 1949).

In the adult the antrum is a 1 ml air-filled cavity. Its anterior wall has the aditus in its upper part and is related to the facial nerve below. Posteriorly the antrum is related to the sigmoid sinus. The roof is separated from the temporal lobe of the brain by the floor of the middle cranial fossa, and the floor is related to the digastric muscle laterally and the sigmoid sinus medially. The medial wall is related to the posterior semicircular canal above, and the endolymphatic sac and posterior fossa dura below. The lateral wall corresponds to Macewen's triangle (Macewen, 1893) which is a standard surgical landmark for identifying the antrum. Its borders are the external canal itself, inferior temporal line, and spine of Henle. The antrum lies just above and behind the postero-superior osseous meatal wall a few millimeters lateral to the annulus and sulcus tympanicum. A well developed Korner's septum (Korner, 1926), an embryological remnant of the petro-squamous suture, can produce the misleading 'false antrum' between those cells that grow outwards into the squamous bone and those that grow downwards into the petrous bone to form the cells of the mastoid process (Glasscock and Shambaugh, 1990).

Agenesis of the mastoid antrum can occur as part of a more extensive congenital otological abnormality in two syndromes i.e. trisomy 13 and mandibulofacial dysostosis.

Trisomy 13 is a condition caused by nondisjunction, translocation or mosaicism and has a three-year survival rate of less than five per cent. It is characterized by microcephaly with a sloping forehead and arrhinencephaly, capillary haemangioma in the glabellar region, cleft lip and/or palate and micrognathia, microphthalmia, iris coloboma and hypertelorism, low set ears, polydactyly, congenital heart defects and mental retardation. In a study of 14 temporal bones of infants with the condition Sando *et al.* (1975) reported the absence of the mastoid antrum amongst other abnormalities of the middle and inner ear; bulky or thick stapes resembling the foetal form, malleus and incus with large bone marrow cavity, absence of pyramidal eminence, flattened horizontal canal cristae, absence or opening of the utricular endolymphatic valve, small facial nerve and obtuse angle of the geniculate ganglion, straight and shortened course of the endolymphatic duct, blindly ending endolymphatic sac, shortening of the cochlear length and widening of the aqueduct.

Mandibulofacial dysostosis is an autosomal dominant syndrome consisting of hypoplasia of the malar bones, antimongoloid obliquity of the palpebral fissures, coloboma of the outer

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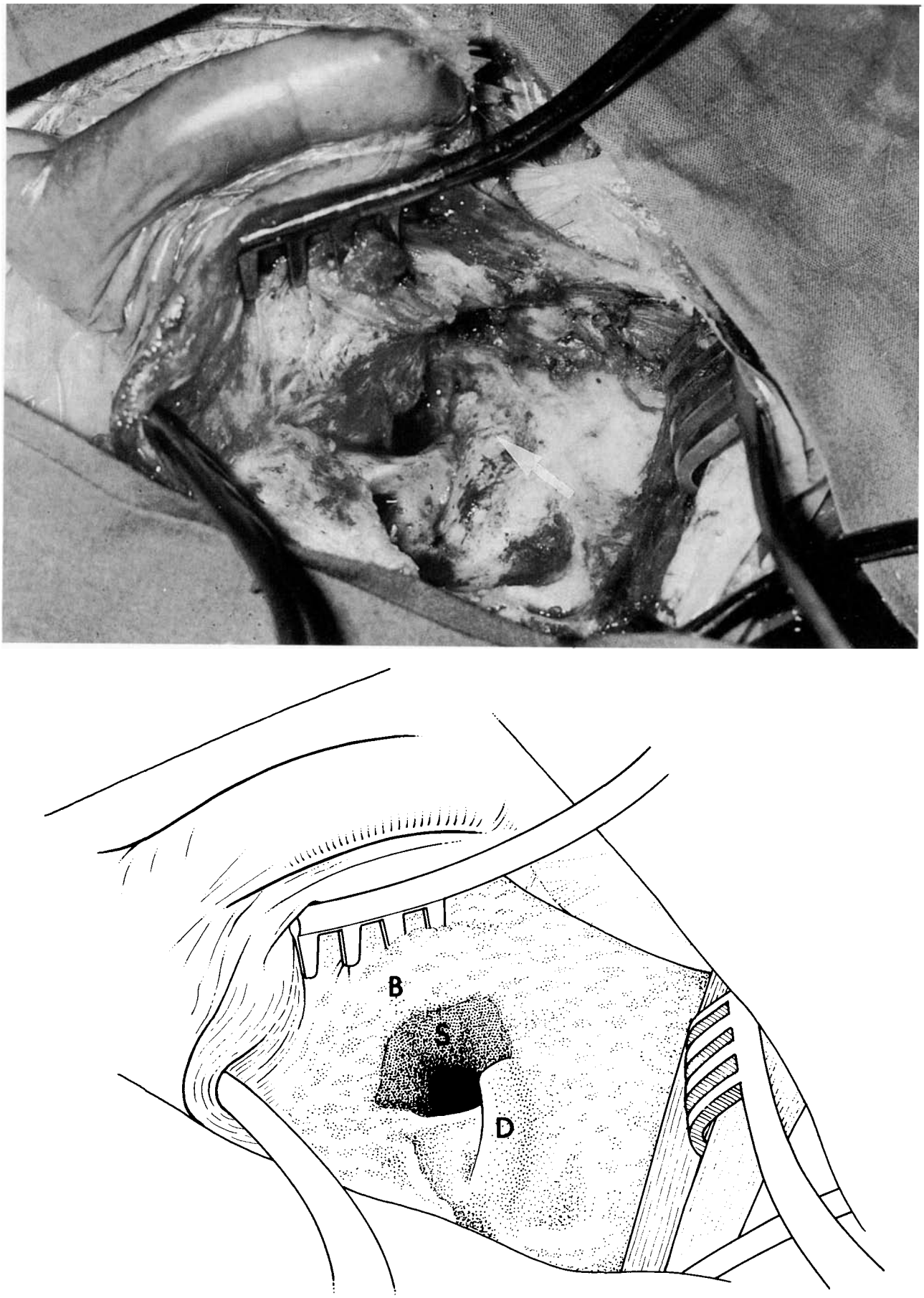

FIG. 1
Uniformly low lying middle fossa dura (arrowed) in agenesis of the antrum. D = dura; S = skin; B = bone.

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third of the lower eyelid, a tongue-shaped process of hair extending toward the cheek, obliteration of the nasofrontal angle, dysplasia of the mandible and microtia. In a study of 16 patients (32 ears) Hutchinson *et al.* (1976) found the mastoid and antrum to be poorly developed or absent in 29 ears. Other otological abnormalities described were mild symmetric deformity of the auricle, absence of the external auditory canal, marked narrowing or agenesis of the middle ear cleft, agenesis or severe malformation of the malleus and incus, deformed stapes superstructure, and anteriorly located facial nerve.

Although isolated agenesis of the antrum has not previously been reported, underdeveloped mastoid pneumatization as a consequence of chronic otitis media is well recognized (Mafee *et al.*, 1986). The inflammatory process is believed to affect the periosteum, mesosteum, endosteum and bone marrow of the spongy bone leading to bone necrosis and reactive osteogenesis (Ferlito, 1974). The osteogenic process causes increased thickening of the trabeculae and finally obliteration of the mastoid air cells but the dura still lies high in its normal position. The association of the uniformly low dura with only a thin plate of bone separating the bony meatus from the middle fossa dura indicates a congenital aetiology.

Conclusion

Agenesis of the antrum, which has previously only been reported in association with the congenital syndromes trisomy 13 and mandibulofacial dysostosis, can occur in isolation. The surgeon must be aware of its existence as disorientation could result in iatrogenic trauma to adjacent structures particularly the lateral semicircular canal and facial nerve. Where it is suspected the middle fossa dura should be exposed and followed posteriorly until the lateral sinus is encountered.

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