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Dermoid cyst in the middle ear

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Abstract
A case of a moderate essentially low tone unilateral conductive hearing loss without a previous history of secretory otitis media in a six-year-old female patient is described. The condition was found to be due to a dermoid cyst in the tympanic cavity, which was confirmed by pathological examination of the surgically removed specimen. This is the sixth documented case of dermoid cyst in the tympanic cavity.

Key words: Dermoid cyst; Ear, middle; Hearing loss, conductive

Introduction

A dermoid cyst is a pathological type of congenital or acquired cyst, and is composed of tissue derived from one or more germinal layers. New and Erich (1937) in their analysis of 1495 dermoid cysts diagnosed between 1910 and 1935 at the Mayo Clinic, identified 24 (1.6 per cent) within the oral cavity but none in the middle ear cavity. The AFIP (Armed Forces Institute of Pathology)—OTR (Otolaryngologic Tumour Registry) records of 1940–1975 contain 10 teratomas (dermoid), three hamartomas, and two christomas involving the middle and inner ear within the temporal bone (Hyams *et al.*, 1988). Unlike teratomas, dermoid cysts are not neoplasms, and dermoid cysts of the head and neck region are predominantly found in the orbital, oral and nasal regions (over 80 per cent), with the remainder found in the neck, occipital or frontal midline, lip or palate (Batsakis, 1979); however, no dermoid cysts of the middle ear were mentioned. Steel's paper in 1976 describes a 67-year-old male patient who presented with secretory otitis media due to a hair-bearing dermoid of the mastoid cavity (Steel, 1976). With reports on this lesion being scarce, he exhaustively reviewed the world literature and thanks to his energetic efforts, two rare descriptions were discovered in the medical archives: one was that of Toynbee (1866), (Steel, 1976) which was recorded in the Transactions of the Pathological Society of London and the other was that of Hinton (1866), (Steel, 1976) was recorded in the same volume. Only a few cases of dermoids in the tympanic cavity have been documented (Scheibe, 1894; Grünwald, 1910; Howie, 1962; Fried and Vernick, 1984). Recently, we treated a patient with a dermoid cyst of the middle ear cavity simulating serous otitis media. The unique location of this tumour and its involvement of the ossicular chain prompted the following case report.

Case report

The patient was a six-year-old female who presented with a conductive hearing loss in the right ear. Neither she nor her parents had been aware of this hearing loss. However, she was seen at her community hospital in August 1991 with a complaint of otalgia in her left ear due to furuncle, at which time her parents were told by an audiologist that her right hearing acuity was considerably diminished. She was treated for three months by her consultant practitioner who performed eustachian tube inflation and administered ear drops, but with no improvement in hearing. There was no previous history of otorrhea, and her parents had thought that the hearing in her right ear had always been normal. She was referred to our clinic for further evaluation. Audiometric testing showed a right low tone conductive deafness, specifically in the frequency range of 125 to 1000 Hz (Fig. 1), and an SRT of 55 dB. Sinus X-ray films showed no evidence of infection, and the lateral view of her head revealed a mild degree of adenoid vegetation. On physical examination, the right ear drum was found to be mobile on pneumatic otoscopy, but its two anterior quadrants were thickened and opaque and had a strange, slightly bulging appearance. The handle of the malleus appeared to be shortened and in a horizontal position, with its apex tipped backwards. No air-fluid level or bubbles could be observed behind the drum. The left ear drum was normal. Impedance audiometry produced a Type B tympanogram in the right ear and a Type A tympanogram in the left. Neither the hearing loss nor the tympanogram of the right ear improved by Politzer inflation,

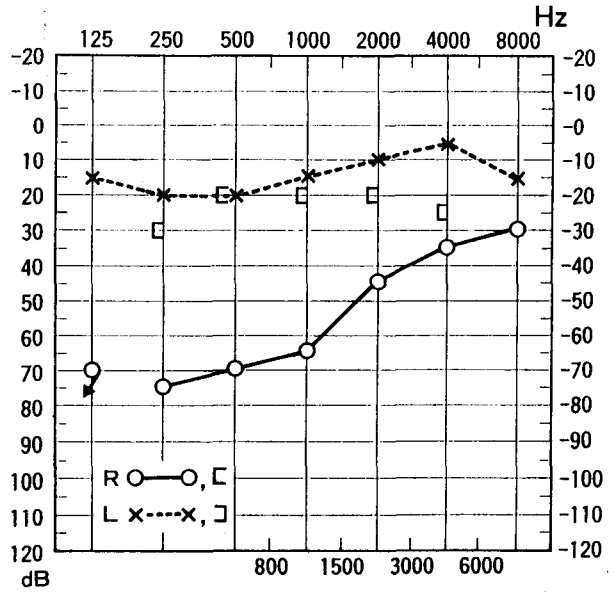

Fig. 1
Audiogram prior to surgery.

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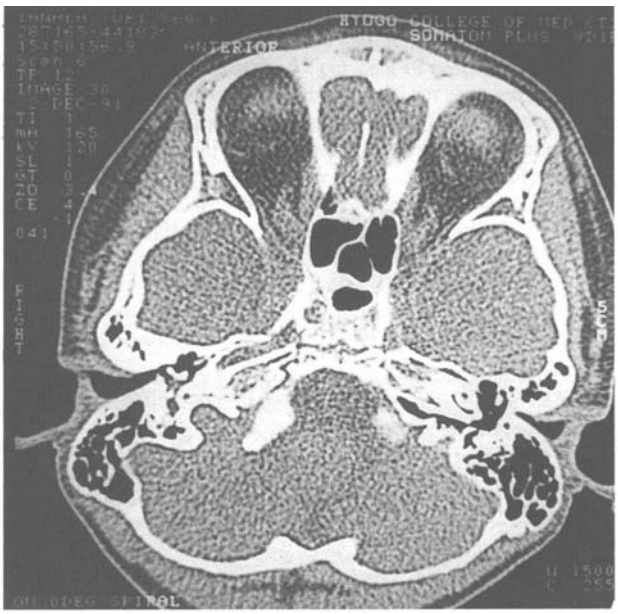

FIG. 2

CT image. Soft tissue mass and conglomerated shadow of the ossicula can been in the right epitympanum.

and the preliminary diagnosis was secretory otitis media. It was felt, however, that this case differed from ordinary otitis media with effusion (OME) because of the presence of tubal dysfunction, and the severity of the hearing loss was incompatible with OME. Accordingly, it was thought advisable to perform a computerized tomographic (CT) scan in anticipation of exploratory middle ear surgery. The CT image (Fig. 2) demonstrated an opacification of the right middle ear cavity with the questionable presence of a soft tissue mass and a conglomerated shadow representing the malleus and incus.

On 3 February 1992, middle ear surgery was performed with the patient under general anesthesia. When the tympanomeatal flap was elevated through a postauricular approach, a yellowish encapsulated lobulated mass was found to be occupying the tympanic cavity. The mass, which was fibrotic, filled the mesotympanum and extended superiorly into the antrum. Examination of the ossicular chain revealed a conglomerated and deformed malleus, incus and stapes (Fig. 3). Both the horizontal semicircular canal and the Fallopian canal were in the normal position; unfortunately, however, the fibrotic wall of the mass adhered strongly to the stapedial footplate and extended between the anterior and posterior crura. During the manoeuvering of the tumour and the conglomerated ossicular chain, the stapes was dislocated, so the oval window was subsequently sealed with temporal muscle fascia. After the tumour was completely removed, secondary reconstruction of the ossicular chain was considered secondarily.

The post-operative course was uneventful, and at six months post-peratively, there was no evidence of any recurrence of the tumour and the SRT had improved to 30 dB. The histopathological diagnosis was dermoid cyst of the middle ear (Fig. 4).

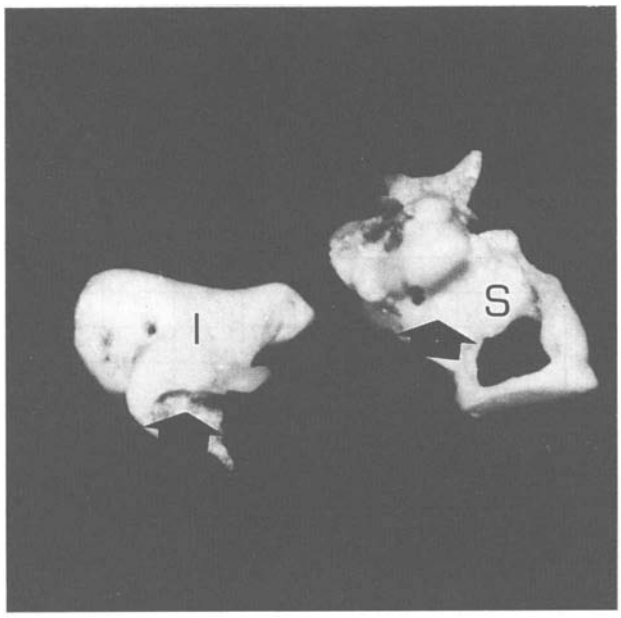

FIG. 3

Extracted ossicula. The long process of the incus (I) and the stapes (S) conglomerated with the deformed incus are enclosed within the mass (arrows).

Discussion

Unlike another epidermal cyst which occurs frequently in the middle ear, cholesteatoma (congenital or postinflammatory), the dermoid cyst is an epithelial-lined cavity containing varying skin appendages, including hair, hair follicles, and sebaceous glands. A review of treatomas, which are true tumours composed of multiple tissues foreign to the part of the body in which they arise, in the middle ear and mastoid, excluding dermoid cysts, was reported by Silverstein *et al.* in 1967. Dermoid cysts can occur anywhere in the body; however, little is known about dermoid cysts in the middle ear and mastoid. McAvoy and Zuckerbaum (1976) investigated the occurrence of several dermoid cysts of the head and neck in a series of 594 epidermal cysts in children, in which 33 such lesions were found. They emphasized that dermoid cysts in children (by their embryologic origin) have a propensity for forming deep tracts which may adhere to the subjacent periosteum.

As described in the Introduction, Steel (1976) reviewed the reports on dermoid cysts in the mastoid, from the first description by Toynbee in 1866 to 1976. According to Steel's report, the first detailed clinical record was made by Wagenhäuser (1888), who described a case of dermoid cyst presenting in a young man with a history of mastoid abscesses. He also found four cases in the literature which referred to non-hair-bearing cysts in the mastoid.

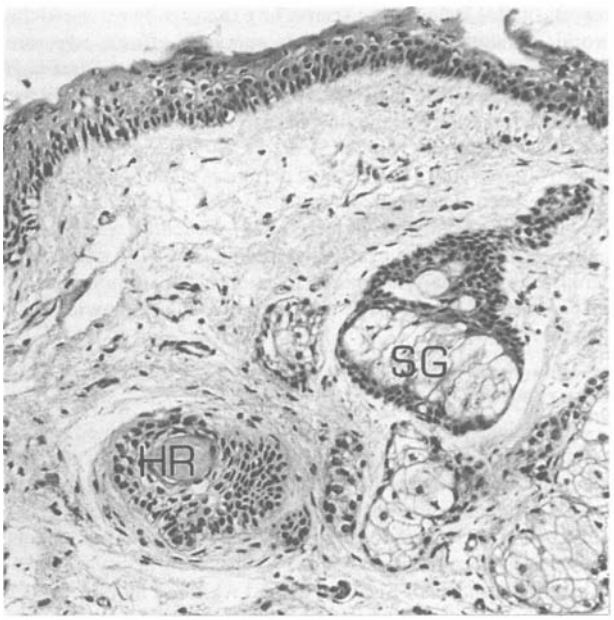

FIG. 4

Histopathological appearance of the mass (H & E stain ×200). The interstitial tissue contains a hair root (HR) and sebaceous glands (SG).

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TABLE I
DERMOID CYSTS CONFINED TO THE TYMPANIC CAVITY REPORTED IN THE LITERATURE

Authors	Age	Sex	Presenting symptoms	Treatment
Scheibe (1894)	41	Male	Recurrent otorrhea of the left ear and polyp	Polypectomy and conservative
	24	Male	Recurrent otorrhea of the right ear	Polypectomy with snare
Grünwald (1910)	27	Female	Recurrent otitis media and polyp	Conservative, later surgery
Howie (1962)	29	Female	Vertigo, vomiting followed by hearing loss (simulating glomus tympanicum)	Surgery
Fried and Vernick (1984)	22 months	Male	Recurrent episodes of otitis with purulent otorrhea	Surgery
Present study	6	Female	Conductive hearing loss without previous history of otitis media	Surgery

Dermoid cysts in the mastoid and antrum have been described by Toynbee (1864); Hinton (1866); Wagenhäuser (1888); Watanabe (1938); Weaver (1943); Portmann and Corgnet (1947); Waltner and Karatay (1947); Richardson (1956); Steel (1976). Concerning the dermoid cysts confined to the middle ear and tympanic cavity, the patients characteristics of reported cases are summarized in Table I. In 1962, Howie reported a dermoid cyst in the left middle ear of a 29-year-old female who presented with the symptoms of hearing loss and vertigo. A small sausage-shaped mass was found to be situated in the lower part of the tympanic cavity, masquerading as a glomus jugulare tumour. Fried and Vernick (1984) published the latest report on a patient 22 months of age, who was the youngest patient with a histologically confirmed dermoid cyst of the middle ear and mastoid. Both Howie (1962) and Fried and Vernick (1984) had overlooked the achievements which were buried in the history: two cases in Scheibe (1894) and one case in Grünwald (1910) which had been documented nearly a century ago. Our patient was thus the sixth case in the literature and unique in some respects. It would be expected from eustachian dysfunction that the initial symptoms could have been due to recurrent episodes of otitis media with effusion; however, this was not applicable to our patient. The CT image and the strange appearance of the ear drum were both consistent with a soft tissue mass in the middle ear cavity. The histopathological diagnosis of the mass was a dermoid cyst. Another unique point was the presence of a curiously deformed malleus and incus, and a part of the stapes assembly, as shown in Fig. 3. The pathogenesis of epidermal or dermal cysts remains as obscure as it was half a century ago (Batsakis, 1979), but dermoid cysts are generally considered to arise from epithelium that has been enclosed in the tissue either on closure of an embryogenic process or from traumatic implantation. In fact, Steel (1976) speculated that the dermoid cyst in the mastoid of his case may have arisen secondary to congenital inclusion of epithelial cells in the region of the squamo-petrous suture.

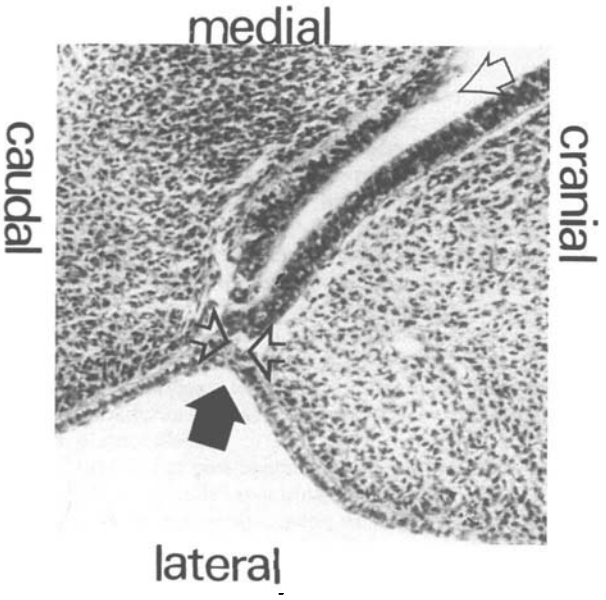

FIG. 5

Transverse section through the branchial groove of human embryo of Carnegie stage 16, showing both ectodermal and endodermal epithelial layers at the most adjacent position. This area would be the primitive tympanic membrane. Arrowheads: primitive tympanic membrane, open arrow, tubotympanic recess, arrow: first branchial groove. (By courtesy of Professor Yoshihiko Nishimura, M.D.)

The aetiology of the dermoid cyst in the tympanic cavity of our case will now be considered. A shallow pit of the primitive external auditory canal appears at Carnegie stage 16 of the embryo (about 6–7 weeks), and the epithelium at the medial end of this pit is temporarily in contact with the thin endoderm of the first pharyngeal pouch (later, the tympanic cavity and eustachian tube) (Fig. 5). The mesoderm later grows in between these two epithelial layers, separating them (Nishimura *et al.*, 1992). During the sixth week of embryonic development, the malleus and incus appear as a single mass, derived from Meckel's and Reichert's cartilage, and are separated. Therefore, some minor developmental anomaly between the ectodermal and endodermal layers, or migration of ectodermal cells into the endodermal layer might occur during an early stage of embryonic life. In the present case, it seems likely that the dermoid cyst in the tympanic cavity may have arisen due to congenital inclusion of these vestigial ectodermal cell components. Regarding the deformed ossicles in this case, the aetiology of this anomaly is far from certain, because both migration of ectodermal epithelium cells in the early stage of embryonic life and a developing dermoid cyst in the tympanic cavity could cause such a developmental anomaly of the ossicular chain.

Since malignant degeneration appears unlikely, treatment for all patients has been complete surgical excision. As with most cysts, and as was described in Howie's case (Howie, 1962), local recurrence at the primary site is common unless the entire wall of the cyst is removed. Therefore, complete excision of the cyst during surgery must be ensured.

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