The Journal of Laryngology & Otology
March 2001, Vol. **115,** pp. 216–219

Adenoma in the middle ear: a report of two cases

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Abstract
Middle-ear adenoma has been reported only in small numbers by surgeons. The few large series reported have been presented by histopathologists. We add two cases of middle-ear adenoma to the published literature, together with pre-, per- and post-operative imaging of one case, as a demonstration of this rare clinical entity. We discuss the pathology of middle-ear adenoma, its diagnosis and treatment, and suggest ways of improving its management.

Key words: Adenoma; Ear, Middle; Histology

Introduction

Adenoma of the middle-ear mucosa is a rare entity. The pathology of the tumour remains the subject of some discussion, but most recent authors concur with its description by Hyams and Michaels,[1] and more recent papers describing the two distinct histological patterns.[2] Due to its relative rarity the pre-operative diagnosis can be difficult, and it may be confused per-operatively with other middle-ear diseases. Relatively few of these tumours have been recorded in the medical literature since their first description and most large case series are from pathologists rather than surgeons. We present two further cases with photographs, computed tomogram (CT) and magnetic resonance (MR) scans demonstrating the appearances. We also suggest improvements in the operative technique for their management to ensure the best possible outcome.

Case 1

A 33-year-old female Caucasian was referred to The Ipswich Hospital in 1994, having noticed a gradual loss of hearing, earache and bubbling noises in her left ear since 1993. She did not notice any ear discharge, dizziness or facial sweakness. Pure tone audiometry then showed normal hearing on the right and a conductive hearing loss of 20 dB in the left ear (Figure 1). A lesion was seen behind the tympanic membrane that could not be identified. A left tympanotomy and biopsy of the lesion was carried out in October 1994 by the same surgeon. Histology showed a lesion consisting of bland relatively uniform glandular cells with eosinophilic cytoplasm arranged as sheets with glandular spaces. The appearances were consistent with a middle-ear adenoma. CT of the temporal bone showed a soft tissue mass within the middle-ear cavity surrounding the ossicles and extending into the attic and the mastoid cells. There was no bony erosion or intracranial extension. An MR scan of the temporal bone confirmed the growth to be confined to the middle ear and mastoid cells, and with no significant enhancement after intravenous gadolinium. The surgeon then attempted to remove the tumour in December 1994 by a tympano-mastoidectomy procedure. The tumour was found to be arising from the promontory. The main bulk of the tumuor was removed, together with the malleus, incus and part of the eardrum. An attico-antrostomy cavity was created in order to remove the tumour in the attic, aditus and mastoid antrum. Part of the tumour was found to be adherent to the stapes and extended into the sinus tympanum and was therefore left behind. After the operation, the patient had no more ear-ache although there was intermittent discharge from the left ear. However, the residual tumour showed signs of continuous

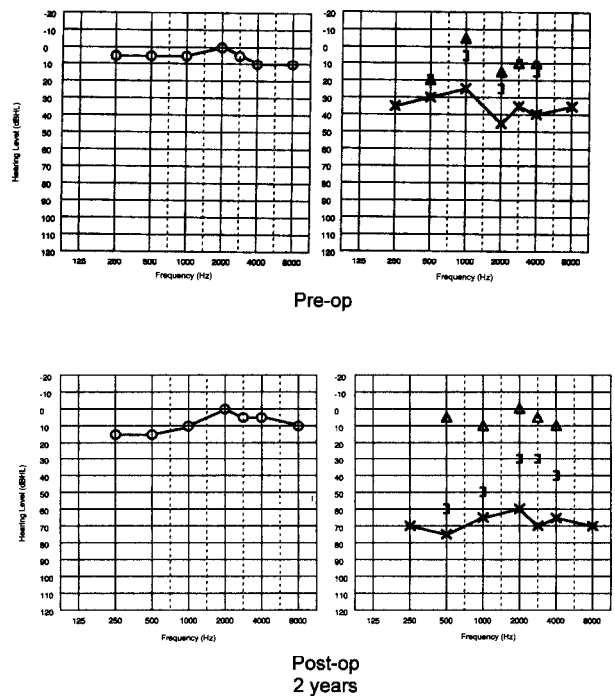

Fig. 1
Pre- and post-operative audiograms of *Case 1*.

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Accepted for publication: 26 September 2000.

enlargement and the patient was then referred to MWY in July 1998 for further treatment.

Pre-operative assessment revealed a small attico-antrostomy cavity in the left ear with a partially epithelialized lining. The ear-drum was intact but its posterior part was pushed laterally by the tumour. The ear was operated on in September 1998 using a post-auricular incision. The middle-ear adenoma was found to be located in the mesotympanum, sinus tympanum, attic region and engulfing the whole stapes suprastructure. It also lay over the round window and the intact Fallopian canal. The tumour was removed using a combination of sharp dissection and the Argon laser. The removal of the tumour in the sinus tympani was controlled using a 30 degree endoscope. The stapes superstructure was removed in order to facilitate total tumour removal. The footplate was found to be mildly subluxed after the tumour was completely removed, hence a type IV tympanoplasty was carried out instead of a total ossicular reconstruction. The size of the mastoid cavity was reduced with a tragal cartilage-perichondrium composite graft. Histology of the recurrent tumour was similar to the previous biopsy.

The patient has been followed-up regularly in the out-patient clinic. So far, there is no evidence of recurrence of the tumour. The mastoid cavity is small and epithelialized and the most recent audiogram in January 2000 showed a mixed hearing loss of 65 dB in the left ear.

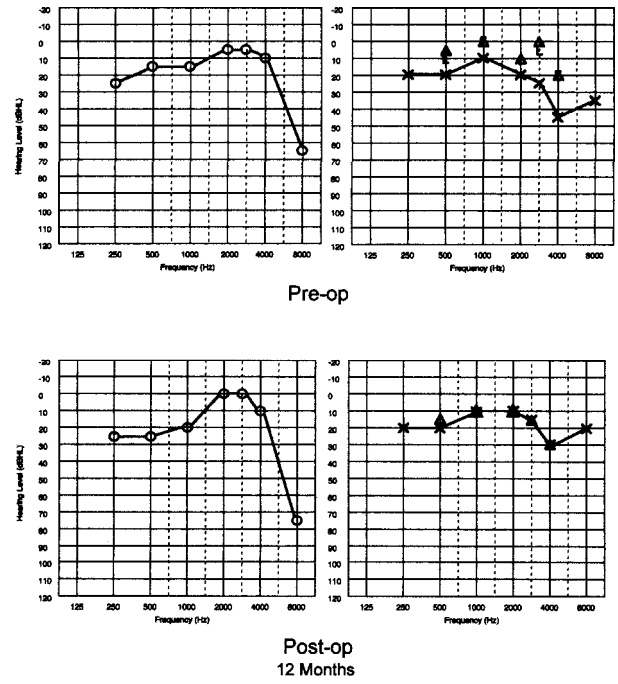

FIG. 3

Pre- and post-operative audiograms of *Case 2*.

Case 2

A 40-year-old female Caucasian was referred to the ENT clinic at The Ipswich Hospital in March 1999. She had had a pressure sensation in her left ear for two years. There was no history of hearing loss, ear-ache, tinnitus, ear discharge or facial weakness. Otoscopy revealed a pink shadow behind the left ear-drum (Figure 2). The pure tone audiogram showed mild high tone sensorineural hearing loss in both ears but no evidence of conductive deafness in the left ear (Figure 3). CT of the ear revealed a soft tissue mass arising from the medial wall of the middle ear, lying mainly above the promontory and surrounding the stapes and incus. It was confined to the epitympanum and mesotympanum with no evidence of bony erosion (Figure 4a). MR scans of the ear showed the mass to be of high signal on T1 and intermediate signal on T2 images (Figure 4b). Fat suppressed MR sequence did not show any change in signal. There was also only minimal increase in signal after intravenous gadolinium was given.

The left ear was explored by MWY in June 1999 using a post-auricular incision. The tumour was found to be arising from the promontory, extending upwards to engulf the head of malleus, incus and stapes, but it did not extend into the mastoid antrum. It obliterated the oval window and lay over the facial nerve and the round window area. The facial nerve was not exposed. Adenoma of the middle ear was diagnosed on a per-operative frozen section and confirmed by later paraffin sections (Figure 5). An atticotomy was carried out to expose the whole tumour

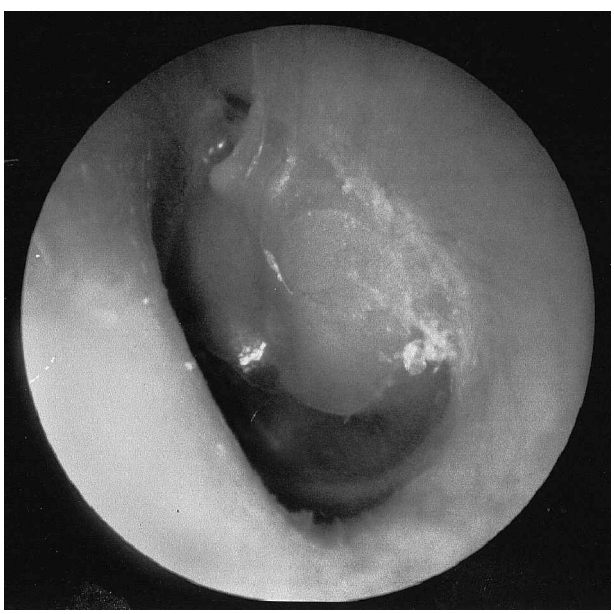

FIG. 2

Pre-operative otoscopy of *Case 2*, showing polypoid mass through tympanic membrane.

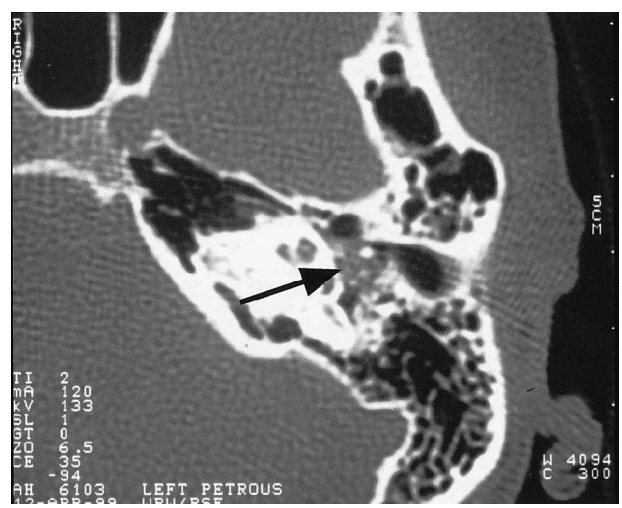

FIG. 4a

CT scan of *Case 2* showing soft tissue density (arrowed) in middle ear surrounding ossicles.

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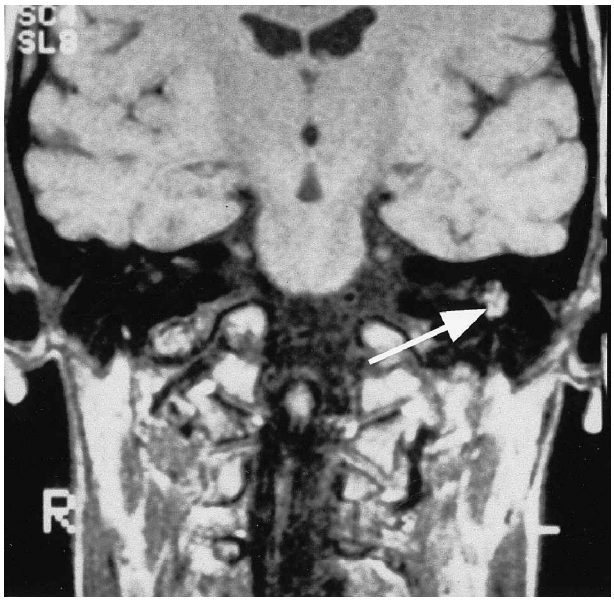

FIG. 4b
MRI scan of *Case 2* showing enhancing mass in middle ear (arrowed).

before its removal. The tumour removal was performed using a combination of sharp dissection and the Argon laser. The incudostapedial joint and the crura of the stapes were divided early during the operation to avoid subluxation of the footplate due to tumour removal at the oval window area. The head of the malleus, the incus, the stapes suprastructure and the posterior half of the eardrum were removed together with the tumour. The tumour was eventually completely removed whilst preserving the footplate and the facial nerve. The ear-drum was grafted using temporalis fascia and a hydroxyapatite total ossicular prosthesis was interposed between the grafted drum and the footplate. The scutum was then reconstructed using a tragal cartilage-perichondrium composite graft.

The patient made an uneventful recovery. Post-operative follow-up revealed a near normal looking ear canal and eardrum. The most recent audiogram in June 2000 showed the threshold in the left ear to be better than 20 dB (Figure 3).

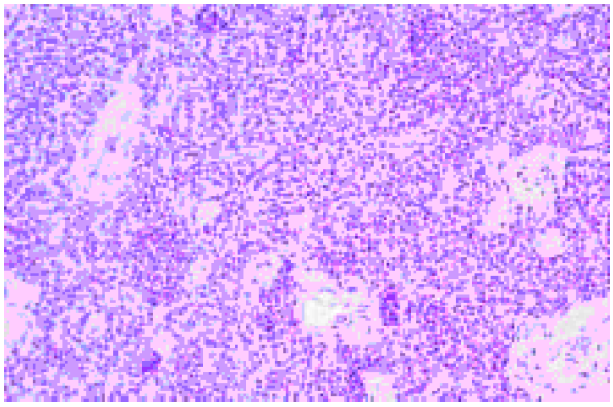

FIG. 5
Sheets and trabeculae of relatively bland epithelial cells. The morphology of the cells is similar to those in *Case 1* although glandular structures are less well defined (H & E; ×125)

Discussion

Many different terms have been used to describe benign glandular tumours of the middle ear, including ceruminoma, adenomatous neoplasms, glandular tumours, cystadenomas, carcinoid tumours of the ear, cylindromas, pleomorphic adenomas and mixed tumours. This partly reflects the relatively poor understanding of the pathology.[3] Even though they were first described over 20 years ago there is still some debate regarding their cellular origin.

The term adenoma of the middle ear was first used in 1976 by Hyams and Michaels.[1] The tumours have been subdivided into different histological types of mixed and papillary patterns.[2] Patients with mixed-type tumours may present with conductive hearing loss, aural fullness or tinnitus. The tumours are always confined to the middle ear and mastoid and rarely erode bone. They are the more common and less aggressive type of middle-ear adenoma. Papillary type adenomas may also present with hearing loss, which may be conductive or sensorineural, but may also have facial nerve paralysis. This type commonly erode bone and may extend intracranially. Both types are seen in a wide age range from the second to the ninth decades.[1,4] These two new cases showed the mixed histological pattern. In *Case 2* there was focal evidence of neuroendocrine differentiation.

It has been suggested that the neuroendocrine differentiation found in the tumours makes them carcinoid tumours rather than adenomas.[5,6] The anatomical site of origin is also debated. Some authors have suggested that these tumours may arise from the external ear canal and extend medially into the middle ear.[1] Both of the tumours in our series were arising from the promontory, and both extended through the obturator foramen.

The appearances of the mixed type of tumour on CT scans may be that of a soft tissue mass confined to the middle ear and mastoid, with cloudy mastoid air cells due to sarcomas or inflammation. The ossicles are often embedded in the mass, but intact. Papillary tumours may show the typical bony destruction on CT, with involvement of the petrous pyramid and middle and/or posterior cranial fossa extension.[7]

The tumuor is rare, therefore its diagnosis is easy to mistake for other causes of conductive hearing loss such as otitis media, mastoiditis, congenital cholesteatoma, glomus tymanicum or choristoma. It was fortunate that the pathologist examining the frozen section specimen was able to give the diagnosis in *Case 2*, as it can be difficult in such tissue types, and paraffin specimens may be required. This allowed treatment by one single operation.

It is important to dislocate the incudostapedial joint and transect the crura at the earliest opportunity, to prevent subluxation of the stapes footplate. It is possible that if this had been done for *Case 1* that the post-operative hearing loss may not have been as severe. As the ear with middle-ear adenoma often has normal eustachian tube function and middle-ear mucosa, ossiculoplasty should have a successful outcome as shown in *Case 2*.

It is suggested that management of the mixed type of adenoma should be complete excision with ossicular reconstruction. For the papillary type wider surgical excision is suggested. No recent authors have suggested that radiotherapy is of benefit when total excision has been achieved.

Conclusion

These cases demonstrate the need to make a diagnosis pre- or per-operatively, and the usefulness of frozen section biopsies. The patient in *Case 2* avoided further procedures

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by having an early diagnosis and full removal of the tumour at the first attempt. Early transection of the crura of the stapes and use of the argon laser for removal of the tumour has reduced trauma to the inner ear for one of the patients.

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Mr S Jones takes responsibility for the integrity of the content of the paper.
Competing interests: None declared

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