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Case Report

Ceruminous adenoma: an unusual mass in external auditory canal: a case report

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ABSTRACT

Most masses in the external auditory canal (EAC) are either polyps or granulations secondary to an underlying inflammatory process. Less common swellings are exostosis and osteomas, a tumour being quite rare. A middle-aged female presented with a mass in the ear which mimicked a meningoencephalocele. However, further study revealed it to be a benign neoplasm of the ceruminous gland of the EAC-a ceruminous adenoma. The tumour was successfully excised with no recurrence on follow-up. A clinician must keep in mind the differential diagnosis of a mass in the EAC. A thorough examination and radiological investigations help aid in the diagnosis.

Keywords: Ceruminous adenoma, EAC tumour, EAC mass

INTRODUCTION

The external auditory canal (EAC) is a skin-lined cul-de-sac, with the tympanic membrane at the blind end of this pouch. The medial one-third of the canal is bony, lined by thin skin of 0.1 mm thickness. The lateral two-thirds of the ear canal is cartilagino-membranous. In the cartilaginous canal, the thickness of the skin ranges from 1.0 to 1.5 mm. The skin in this cartilaginous part has appendages: ceruminous glands, sebaceous glands, and hair.[1]

The anatomy of the EAC is such that whilst protecting the fragile tympanic membrane from external insults, the canal itself is susceptible to blockage, which reduces its primary function of sound conduction. The most common causes of EAC obstruction are wax, otitis externa, and foreign bodies. Here, we describe an unusual mass in the EAC causing obstruction of the EAC-a ceruminous adenoma. Ceruminous adenomas are rare benign tumours arising from ceruminous glands in the EAC. They may be asymptomatic when small, but the patient presents when the tumour increases in size.[2]

CASE REPORT

A 39-year-old female presented with the sensation of left ear block since the past six months. She also complained that she felt a swelling in her ear but had no complaints of pain or discharge from the ear. On oto-endoscopic examination, a solitary mass was seen descending from the roof of the EAC, obscuring the tympanic membrane. The mass appeared pale, bluish, with a leash of blood vessels on it (Figure 1). It was soft in consistency, with attachment to the postero-superior wall of EAC.

The appearance and location of the mass led us to suspect a meningocele or a meningoencephalocele. However, it was not pulsatile. There was no history of previous trauma. Hence, high-resolution computed tomography (HRCT) of the temporal bone was sought, which revealed that the mass was limited to the EAC. Also, the mass was

notably distant from the tegmen, as seen in Figure 2. Pure tone audiometry showed conductive loss of 35 dB.

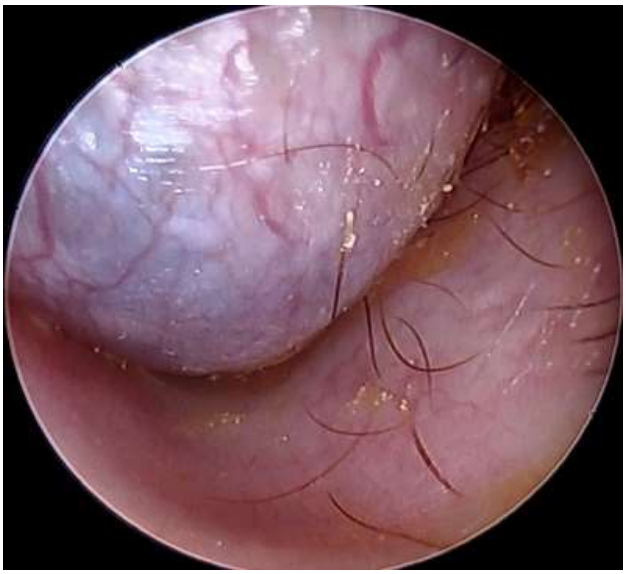

Figure 1: Solitary mass in the left EAC, obscuring the tympanic membrane.

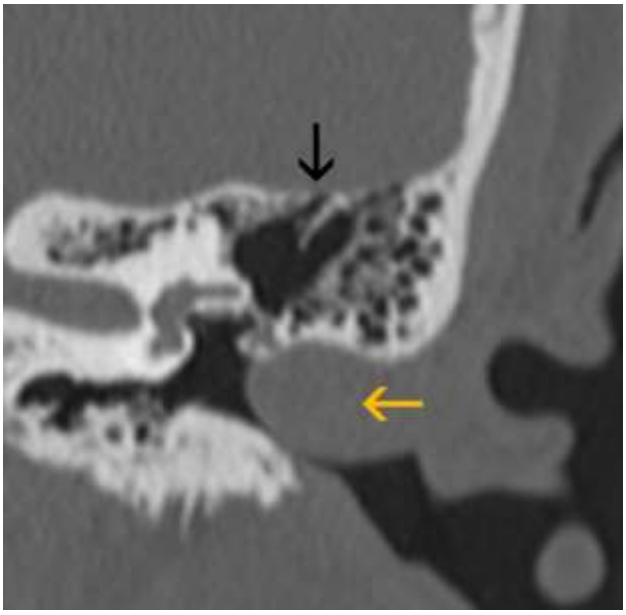

Figure 2: HRCT Temporal bone-mass in EAC (yellow arrow), distant from tegmen (black arrow).

Thereafter, the patient underwent complete excision of the EAC mass through a post-aural approach under local anaesthesia. A mass of approximately 1×0.7 cm was meticulously elevated off the posterosuperior bony canal wall, excised, and sent for histopathological examination. No invasion to the underlying bone or middle ear was seen. The tympanic membrane was intact. The canal skin defect was covered using a post-aural split skin graft.

The cut surface of the mass revealed a cystic lesion. Histopathologic examination revealed a cyst lined by an apocrine type of epithelium. The cyst had multiple invaginations into the stroma, lined by a similar apocrine epithelium, with areas of papillary projections and complex cystic/ glandular spaces. Also, the cells showed apocrine decapitation type of cytoplastic projections, with brownish pigment within the cytoplasm (Figure 3). No cellular pleomorphism or abnormal mitosis was seen, which ruled out malignancy. Based on the histopathological examination, it was diagnosed as a benign tumour of the EAC-ceruminous papillary cystadenoma, a type of ceruminous adenoma.

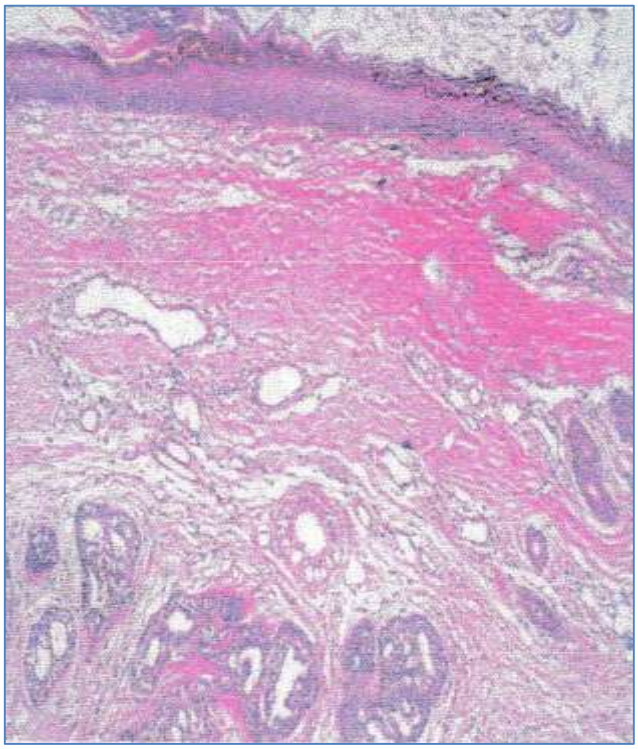

Figure 3: HPE showing apocrine epithelium, with areas of papillary projections and complex cystic/ glandular spaces, apocrine decapitation type of cytoplastic projections, with brownish pigment within cytoplasm.

Postoperatively, the patient had normal hearing and was seen to have a complete recovery with healthy skin lining the EAC. She was followed up for five years after excision, with no recurrence.

DISCUSSION

Most tumours in the EAC are aural polyps or granulations secondary to inflammation in the external and/or middle ear due to conditions such as cholesteatoma, keratosis obturans, or malignant otitis externa. However, several other benign tumours can also present in the EAC.[3] Malignant tumours of the EAC are rare, representing 0.2% of all head and neck cancers.[4]

Table 1 lists the differential diagnosis for a mass in the EAC.

Table 1: Differential diagnosis of masses in the EAC.[3,4]

Inflammatory	Tumours		Others
	Benign	Malignant	
Aural polyp	Hemangioma	Squamous cell carcinoma	Glomus tumours
Granulation	Ceruminous adenoma	Lymphoma	Osteoma, exostosis
	Ceruminous, pleomorphic, adenoma	Basal cell carcinoma	Meningocele, encephalocele, meningoencephalocele
	Ceruminous, syringocystadenoma, papilliferum	Adenoid cystic carcinoma	Sebaceous cyst, branchial cleft cyst
		Adenocarcinoma	TMJ ganglion cyst
		Ceruminous carcinoma	Heterotopic glial tissue
		Malignant fibrous histiocytoma	
		Rhabdomyosarcoma	

Ceruminous glands are seen in the outer one-third to one-half of the EAC overlying the cartilage. They are modified apocrine sweat glands. The luminal secretory cells have golden yellow-brown, lipo-fuschin-like ceroid pigment granules in their cytoplasm.[5] The apical blebs of these luminal epithelial cells are pinched off into the lumen, which is described as decapitation secretion. These secretions further drain into ducts that open into the hair follicles of the fine hairs in the cartilaginous canal and mix with the sebum of the sebaceous glands to create cerumen.[6]

Ceruminous adenomas are benign glandular neoplasms of ceruminous glands.[2] Wetli described ceruminous adenoma as "a well-differentiated, benign neoplasm, localized, occasionally showing cystic and papillary proliferation of glands histologically similar to normal ceruminous glands."[7]

Ceruminous adenomas are rare, accounting for less than 1% of all external ear tumours. They usually affect middle-aged patients with no sex predilection. Small ceruminous adenomas typically cause no symptoms, and patients present when the mass increases in size, causing canal obstruction.[5] Most tumours are small, with a size ranging from 1.15 to 1.2 cm.[2,5]

The macroscopic appearance of the tumour is variable. The most common is a polypoid mass that is tan or reddish in colour. The cut surface may show small cysts. Surface ulceration is quite rare.[8]

On microscopic examination, ceruminous adenomas show well-differentiated tubular, ductal, or papillary structures akin to normal ceruminous glands, with two distinct cell layers. The luminal cells contain pigment granules and display apical secretion by decapitation. Surrounding these luminal cells are the myoepithelial cells. Features suggestive of malignancy, such as invasion beyond the basement membrane, nuclear pleomorphism, mitotic activity, tumour necrosis, and haemorrhage, are usually absent.[2,6]

Complete excision is curative.[2]

Previously, irrespective of gland of origin, all glandular tumours of EAC had been termed ceruminoma.[9] Until recently, there has been some confusion in the literature with no agreed nomenclature system for these tumours.[5,10] WHO recognizes 2 categories of ceruminous neoplasms: malignant and benign, as shown in Table 2.[11]

Table 2: WHO recognizes 2 categories of ceruminous neoplasms: malignant and benign.

Benign/ ceruminous adenomas	**Ceruminous adenoma NOS, ceruminous pleomorphic adenoma, ceruminous syringocystadenoma papilliferum**
Malignant/ ceruminous adenocarcinomas	Ceruminous adenocarcinoma NOS, ceruminous adenoid cystic carcinoma, ceruminous mucoepidermoid carcinoma

CONCLUSION

Tumours of external auditory canal are uncommon. Ceruminous adenomas, as described here, are even rarer. Complete excision is usually curative, with no recurrence.

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