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Article in *Indian Journal of Otolaryngology and Head & Neck Surgery* · September 2024

DOI: 10.1007/s12070-024-05001-2

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Ceruminous Adenoma: A Case Report and Review of Literature

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Received: 26 July 2024 / Accepted: 16 August 2024
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Abstract

Ceruminous adenoma is a rare and benign tumor of the external auditory canal and present with a nonspecific symptom. Biopsy with histopathology examination is required to establish the diagnosis. Wide local excision of tumour shows favourable outcome with a low recurrence rate. Here, we present a case of ceruminous adenoma masquerading as a furunculosis.

Keywords Ceruminous Adenoma · Furunculosis · Endoscopic Transcanal Approach

Introduction

Ceruminous glands are modified apocrine glands found in the skin lining of the cartilaginous portion of the external auditory canal (EAC). The bony portion of the EAC is normally devoid of these cells. The function of these glands together with the sebaceous glands are to produce cerumen which act to clean and lubricate the EAC. Ceruminous

adenoma (CA) is a rare and benign tumor that arises from the ceruminous glands, usually seen as a mass in the outer half of the EAC [1]. Due to its rarity and non-specific clinical presentation, CA are sometimes preliminary treated as furunculosis or otitis externa [2, 3].

Case Report

A 48-year-old lady presented with left otalgia for 2 days in duration. She also had left ear fullness for the past 3 months. He had no ear discharge, hearing loss, vertigo, tinnitus, or fever. No facial nerve palsy or mastoid tenderness were elicited during examination. Otoscopy examination of the left ear showed a 1 × 1 cm sessile mass at the posterior wall of the EAC which was tender on probing (Fig. 1a). Tympanic membrane (TM) was intact. A preliminary diagnosis of left furunculosis was made and an incision and drainage performed. However, there was no pus seen. An ichthammol glycerin (IG) ear wick was inserted and she was started on ofloxacin eardrops. In the subsequent follow up, the mass persisted. A biopsy of the mass was performed and histopathological examination revealed features suggestive of ceruminous adenoma. High resolution computed tomography (HRCT) of the temporal bone showed soft tissue mass at posterior wall of outer half of the EAC, approximately 0.5 × 1.6 × 0.7 cm in size with no bony erosion. The middle ear cavity and mastoid air cells were normal.

The patient subsequently underwent an endoscopic wide local excision of the tumor. (Fig. 1b&c). Intraoperatively, a

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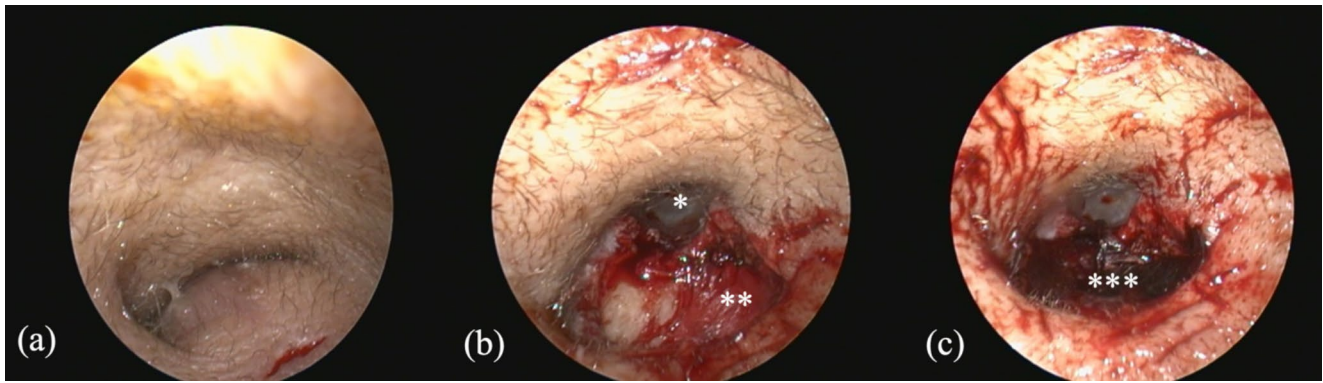


Fig. 1 (a) well-circumference mass arising from the outer 1/3 of the posterior wall of the left EAC. (b) view of the EAC post excision (*tympanic membrane, ** bony portion of the EAC). (c) temporalis fascia graft placed over the bony portion of the EAC (***) temporalis fascia)

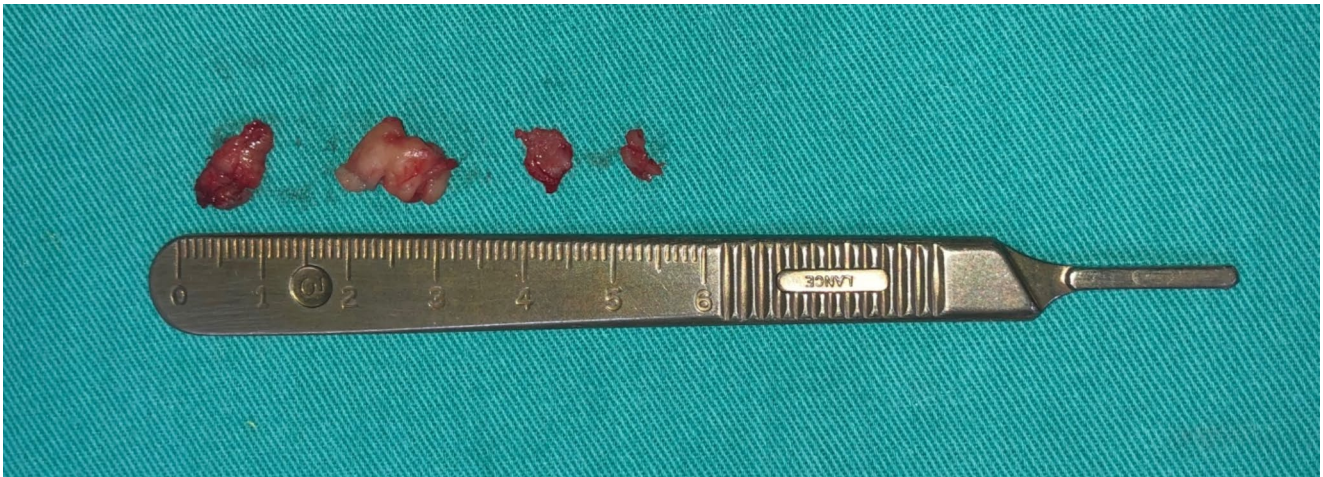


Fig. 2 Lesion measuring approximately 2.5 cm x 1 cm

1.5 × 1.5 cm firm greyish mass at the inferior-posterior part of the outer one-third of the EAC was removed until the bony part of the EAC (Fig. 2). A left temporalis fascia graft was harvested to cover the bony base of the EAC. Postoperatively period was uneventful. Histopathological examination revealed well circumscribed glandular proliferations in a solid, cystic pattern. Immunohistochemical staining was positive for CK7. (Fig. 3a&b). No malignant features were seen. At her last follow-up visit, she was well and there was no clinical evidence of recurrence.

Discussion

According to the WHO classification, ceruminous neoplasm can be classified into benign (ceruminous adenoma, ceruminous pleomorphic adenoma, ceruminous syringocystadenoma papilliferum) and malignant (ceruminous adenocarcinoma, ceruminous adenoid cystic carcinoma, ceruminous mucoepidermoid carcinoma) [6, 7]. Ceruminous adenoma (CA), one of the benign type of ceruminous

neoplasm, accounts for <1% of all external ear tumor [1]. CA usually affects the middle-age groups (mean age, 55 years) with no gender predilection [1, 4]. In our case, the patient is a 48-year-old lady.

CA can present either as a smooth mass or an ulcerated mass located on the posterior wall of the outer half to the outer one-third of the EAC [3, 4, 6, 7]. The most common symptom is conductive hearing loss followed by otorrhea, otalgia, tinnitus, aural fullness and neural symptoms., [4–6] In our case, the patient presented with otalgia which was likely due to a superimposed infection of the EAC, mimicking a furunculosis/ otitis externa [2, 3, 7]. A painful mass that does not respond to antibiotic treatment should raise the suspicion of a more sinister diagnosis [4]. In our case, a biopsy was taken from the EAC mass as the patient did not show any clinical improvement after the initial treatment.

CA must be differentiated from ceruminous adenocarcinoma as the mode of treatment is very different [9]. It is however, very difficult to differentiate CA from ceruminous adenocarcinoma clinically as they present with similar clinical features. A histological examination from

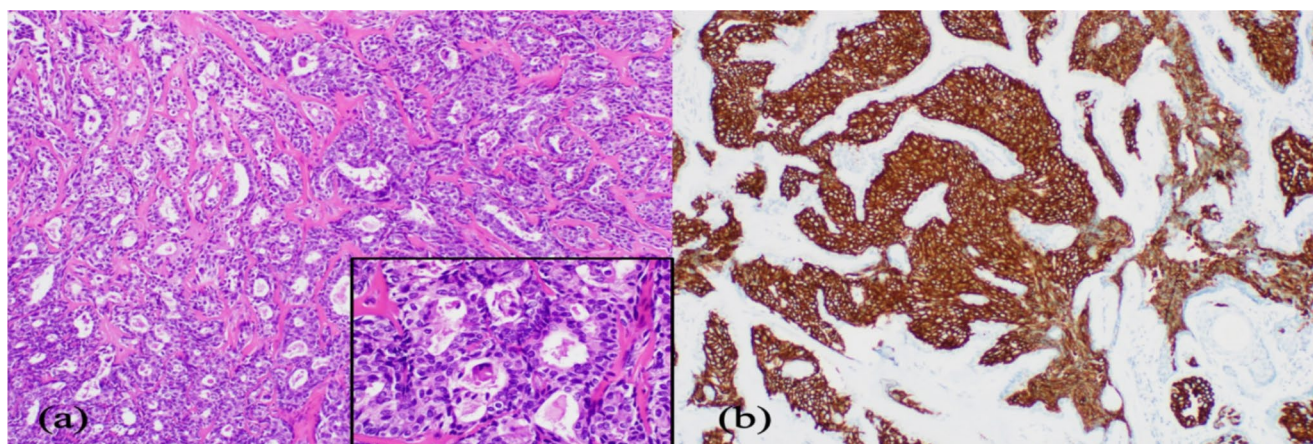


Fig. 3 (a) Fragments of tissue composed of well circumscribed glandular proliferations in a solid, cystic pattern. The glands are composed of inner cuboidal cells with eosinophilic cytoplasm and outer spindled myoepithelial cells with hyperchromatic nuclei embedded within hya-

linized stroma. No prominent pleomorphism or mitotic figures. No invasion or necrosis. (Inset: H&E,40X). (b) Immunohistochemical staining highlights the inner cuboidal cells (positive for CK7)

a tissue biopsy is the gold standard to confirm the diagnosis. Macroscopically, CA are well-circumscribed but non-encapsulated tumours. It appears as grey to pink polypoid masses with smooth and firm in consistency, covered by a non-ulcerated epithelium [4, 6, 7, 10]. The mean size of CA masses reported were 1.1 cm with a range from 0.4 to 2.0 cm [4]. In our case, it appeared as shiny, rubbery, grey to pink mass. Unfortunately, the mass was fragmented during manipulation with the size of the fragments ranged from 0.3 to 1.2 cm.

Microscopically, CA is composed of dual cell population, arranged in glandular/cystic pattern. The inner luminal cells consists of cuboidal to columnar cells with abundant eosinophilic cytoplasm and contain golden-brown coloured ceroid pigment [4–6, 10]. These cells were surrounded by an outer layer of cells called myoepithelial cells. The inner luminal cells is labelled by cytokeratin 7 (CK7) and outer myoepithelial cells stained with S100, CK5/6 and p63 [4, 5, 10]. Ceruminous adenocarcinoma on the other hand, shows an absent of frank cellular pleomorphism, prominent nucleoli, atypical mitotic features and necrosis. Perineural or lymphovascular invasion and absence of ceroid pigmentation could be identified in cases of ceruminous adenocarcinoma [4, 6, 8]. In our case, the histopathological report showed glandular structure lined by inner cuboidal to columnar cells with eosinophilic cytoplasm (positive for CK7 stain) and outer spindled myoepithelial cells (positive for p63 stain) with no nuclear pleomorphism, prominent nucleoli, necrosis or mitotic figures seen which confirmed the diagnosis of CA in our patient.

Radiological features of CT scan such as tumour invasion/infiltration or bony erosion will suggest malignancy [3, 6]. In our patient, the CT scan showed no bony erosion and no evidence of local invasion/infiltration of tumour which

indicate a benign EAC tumour. Other differential diagnosis of CA includes osteoma, neuroendocrine adenoma of the middle ear (NAME), pleomorphic adenoma of the parotid gland, meningioma, and paraganglioma [3, 5, 7, 10]. Osteoma can be diagnosed based on its hyperintense appearance at tympanosquamous or tympanomastoid suture on CT scan. A mass originating from the middle ear cavity might suggest of NAME. A pleomorphic adenoma of parotid gland could extend into EAC due to its close anatomy with the floor of EAC. A thorough otoscopy examination and CT scan could be used to locate the origin and extension of the mass in order differentiate CA from NAME and pleomorphic adenoma of parotid gland [4].

Wide local excision with free margin is the recommended treatment of CA [2, 4–7, 10]. Surgical approaches include endaural, postauricular and transcanal approaches [2, 3, 5, 7, 10–13]. The rationale of different surgical approaches mainly depends on the degree of external canal obstruction and the extent of the lesion. [14]. Postauricular approach provide the optimum surgical visualisation especially in the situation where the medial end of the tumour is unable to appreciate through EAC. The disadvantages of postauricular approaches is its cosmetic worry of the scar behind the ear and carry risk of facial nerve injury especially in young children. For small benign tumours, both endaural or transcanal approaches are viable options. Compared with transcanal approach, endaural approach provides a better access and visualisation which can facilitate a complete excision with a small drawback of leaving a neglectable scar near the EAC. A summary of different surgical approach of wide local excision of CA based on our literature reviews are shown in Table 1, all of which carries equivocal good surgical outcome. A complete excision of CA is important as it shows good long-term prognosis [4, 5, 10]. In spite of

Table 1 Comparison case report of different surgical approaches in EAC ceruminous adenoma. N/A: no available

Authors	Journal and year of publication	Age	Size of tumor	Surgical approach	Outcome
Elsürer C et al.	Eur Arch Otorhinolaryngol. 2007 (2)	37y/o	1 × 1 cm	Wide local excision	No recurrence
Uz, U et al.	SAGE open medical case reports, 2018 (3)	32y/o	2 × 1.5 cm	Transcanal approach	No recurrence
Varshney H et al.	Indian J Otol., 2014 (5)	38y/o	N/A	Endaural approach	No recurrence
Psillas G et al.	Case Rep Med, 2015 (7)	87y/o	1.9 × 1.4 cm	Transcanal approach	No recurrence
Das et al.	Journal of cytology, 2017 (10)	45y/o	1.5 × 1.5 cm	Wide local excision	No recurrence
Yeo et al.	Kosin Medical Journal, 2018 (11)	55y/o	1.1 cm	Transcanal approach	No recurrence
Niemczyk et al.	International journal of pediatric otorhinolaryngology, 2015 (12)	3y/o	N/A	Postauricular approach	No recurrence
Giuseppe et al.	Otology & neurotology, 2011 (13)	5y/o	0.6 cm	Transcanal approach	No recurrence

this, residual/recurrence CA post wide local excision have been reported in a previous case study, as far as 4 years from the initial presentation [4, 5, 10]. In our patient, endoscopic transcanal approach was selected as it was able to provide an adequate and complete surgical view the ear canal without leaving any scar externally. Its high resolution image and its ability to magnify the view, permits a better view of the tumor margin, which is imperative in facilitating the complete excision of the tumor. Its minimal invasiveness nature also help to provide shorter postoperative pain and healing period. Endoscopic transcanal approach, however, requires single handed surgery training and practice [15]. Even through in our patient, the EAC mass fragmented into few pieces during the procedure, we were still able to excised it completely from the EAC. At her last follow-up, no recurrence was seen.

Conclusion

A biopsy for histology is important in any case of painful EAC mass which shows no response to treatment. Other possible differential diagnosis of EAC mass needs to be considered. A wide local excision with a clear margin and long-term follow-up post excision is highly recommended given the recurrence nature of CA.

Author Contributions Weijie Boon and Boon Han Kevin Ng conceived the original idea. This was also discussed with Ing Ping Tang. Histopathological picture provided by Nur Shazwaniza Awang Basry Eventually, all the authors discussed and agreed with the focus and ideas of this paper. The main text of the paper was written by Weijie Boon and edited by Boon Han Kevin Ng, Leong Wai Yee, Foong Seong Kin, Ing Ping Tang. All authors revised the manuscript and contributed equally.

Funding No funding is involved in this paper.

Data Availability No applicable.

Declarations

Ethics Approval and Consent to Participate Not applicable. Reference number is not available.

Consent for Publication Explicit verbal consent was obtained from the patient for the publication of this case report.

Competing Interests The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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