Ceruminous adenoma mimicking otitis externa

Uzdan Uz1, Ayca Tan2 and Onur Celik3

Abstract
Ceruminous adenoma is an extremely rare condition that arises in the external auditory canal. The right ear canal in a 32-year-old man was obstructed by a ceruminous adenoma mimicking otitis externa and its symptoms. The lesion was resected under microscopic view using a tran canal approach. There were no tumor-related symptoms postoperatively and he has been disease free for 1 year after surgery.

Keywords
Ceruminous adenoma, otitis externa, transcanal resection

Introduction
Ceruminous gland tumors of the ear canal are very rare clinical–pathological entities.1 Ceruminous adenoma (CA) arises from a modified apocrine sweat gland in the external auditory canal called a “ceruminous gland.”2 Mean age of onset is usually the sixth decade of life (range, 24–85 years) with no sex preference, and CA is more frequent in cats and dogs compared to humans.1,2 This benign tumor is difficult for clinicians to diagnose because of their varied clinical presentation, and diagnosis is made histopathologically; however, CA has a good prognosis after complete excision.2

Case presentation
A 32-year-old man was admitted to our clinic with ear discharge, pain, and hearing loss. From otoscopy, a mass was identified, covered by skin and protruding from the postero-inferior wall of the entrance to the external auditory canal. The passage of the canal was significantly narrowed by the tumor, and the tympanic membrane was not visible. After 1 week of topical treatment (ciprofloxacin, dexamethasone), the patient’s symptoms and clinical findings were observed to improve. Audiological tests and a computed tomography (CT) scan were performed. The pure tone audiogram indicated conductive hearing loss with a mean air-bone gap of 25 dB. The CT scan showed a soft tissue density mass protruding from the postero-inferior wall of the entrance to the external auditory canal and obstructing the canal with no invasion of other tissues (Figure 1a). Fine-needle aspiration biopsy was performed; however, an insufficient number of specialized cells were present in the pathological samples to permit diagnosis.

One month after the appearance of symptoms, surgical excision was scheduled under local anesthesia. Under microscopic view, after transcanal lateral circumferential skin incision, a skin flap was elevated and the encapsulated tumor with a diameter of approximately 20 mm was seen and excised en bloc. The skin flap was replaced to cover the surgical area and two Merocel® ear packs were placed in the external ear canal. After surgery, the patient was prescribed analgesics (paracetamol, 3 × 500 mg/day) for 7 days. The tumor was off-white in color with the dimensions of 20 × 12 × 15 mm3. Microscopic examination showed glandular structures which were composed of two cell layers: epithelial and myoepithelial. The inner layer consisted of

---

1 Department of Otorhinolaryngology, Bayindir Government Hospital, Bayindir, Turkey
2 Department of Pathology, Manisa Celal Bayar University, Manisa, Turkey
3 Department of Otorhinolaryngology, Manisa Celal Bayar University, Manisa, Turkey

Corresponding Author:
Uzdan Uz, Department of Otorhinolaryngology, University of Health Sciences, Izmir Bozyaka Training and Research Hospital, 35170 Izmir, Turkey. Email: dr.uzdan@uzdan.net
middle-sized cylindrical or cuboidal cells. Abundant eosinophilic cytoplasm, arranged in a columnar to cuboidal shape, was seen in the luminal cells. The outer layer consisted of spindle cells with elongated nuclei. Neither atypia nor mitotic activity was observed. All of the microscopic features were indicative of CA (Figure 2) and the surgical margin was clear.

The patient has been tumor free for 12 months postoperatively (Figure 1b), with no hearing loss or symptoms of external otitis in this period. Written informed consent was obtained from the patient.

Discussion

Ceruminous gland tumors of the external auditory canal are uncommon lesions originating in the outer third of the lateral ear canal (cartilaginous portion) and are absent in the bony part of the canal. Benign ceruminous gland tumors, called “ceruminoma,” were first reported by Haugh in 1894. Despite the fact that the definition of “ceruminoma” was removed from the World Health Organization (WHO) classification in 1991, this term is still in use today.

Ceruminous adenomas originate from modified apocrine glands and are classified as ceruminous gland tumors. They are painless, non-mobile, slowly growing semisolid masses, and symptoms of these tumors are caused by obstruction of the ear canal leading to conductive hearing loss, otorrhea, otitis externa, tinnitus, and aural fullness and, occasionally, the tumor has been found incidentally. It can also mimic furunculosis. In our case, the CA presented as a smooth round mass with discharge, hearing loss, and pain.

According to the literature, ceruminous gland tumors may be classified into seven categories (Table 1) with either benign nature: (1) ceruminous adenoma, (2) pleomorphic...
Table 1. Classification of benign and malignant ceruminous gland tumors.

| Benign nature                  | Malignant nature                  |
|--------------------------------|----------------------------------|
| Ceruminous adenoma             | Ceruminous adenocarcinoma         |
| Pleomorphic adenoma            | Adenoid cystic carcinoma         |
| Cylindroma                     | Mucoepidermoid carcinoma         |
| Syringocystadenoma papilliferum|                                  |

adenoma, (3) cylindroma, (4) syringocystadenoma papilliferum, or malignant nature: (5) ceruminous adenocarcinoma, (6) adenoid cystic carcinoma, and (7) mucoepidermoid carcinoma.1

Ceruminous adenoma is a well-differentiated, localized benign neoplasm that shows immunohistological characteristics similar to papillary proliferation of those glands to normal ceruminous glands. The precursor of pleomorphic adenoma of the ear canal is considered to be from myoepithelial cells of the ceruminous glands and histopathological findings are comparable to the appearance of pleomorphic adenoma of the salivary gland. Cylindroma, also called benign eccrine cylindroma, is an uncommon benign entity arising from the eccrine glands located anywhere in the head and neck region including the external auditory canal. The difference between CA and benign cylindroma is that the cylindroma originates entirely from the eccrine glands, whereas CA is thought to originate from the apocrine glands. Syringocystadenoma papilliferum is a rare benign adnexal tumor occurring in the head, neck, scalp, and also in the external auditory canal, and has been thought to be derived from the eccrine or apocrine sweat glands.3 Adenoid cystic carcinoma is the most common malignancy of the ceruminous gland and is involved in local, perineural, and parotid gland invasion.4 Mucoepidermoid carcinoma is a rarer malignancy of the ear canal than adenoid cystic carcinoma. The potential origin of mucoepidermoid carcinoma is the ceruminous gland; however, the actual pathogenesis is not known.5

Differential diagnosis of CA includes pleomorphic adenoma, meningioma, exostosis, osteoma, eosinophilic granuloma, cholesteatoma, choristoma, branchial cleft cyst, meningioma, neuroendocrine adenoma, paraganglioma, and also malignant tumors such as ceruminous adenocarcinoma.2 The diagnosis is made histologically. Imaging techniques such as CT and magnetic resonance imaging (MRI) do not give sufficient information to permit a diagnosis of benign ceruminous gland tumor. However, showing no signs of invasion is an important pointer to the benign nature of these tumors. In addition, MRI with gadolinium-diethyleneetriaminedipentaaetic acid (DTPA) contrast is convenient for differentiating between malignant and benign gland tumors.6

The current treatment of choice for all benign ceruminous gland tumors of the ear canal is en bloc surgical resection with a sufficient margin including canal skin in order to prevent recurrence and reconstructed with a free skin flap.7,8 In our case, the tumor was excised using a transcanal approach with a skin flap and no recurrence has been observed for a year after surgery. In addition, if the tumor was well encapsulated and under microscopic view, a transcanal approach with a skin flap should be the preferred surgical option. In appropriate cases, minimally invasive techniques instead of wide resection could reduce the risk of complications and increase the quality of a patient’s life postoperatively. In addition, although the recurrence rate is low, long-term follow-up is recommended.

Conclusion

In this case of ceruminous adenoma, a routine surgical procedure with skin flap elevation, en bloc tumor resection with sufficient margin, and flap replacement was performed. The importance of this case report is not the surgical procedure used but the observation that a rare tumor may mimic ordinary external ear canal diseases.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

ORCID iD

Uzdan Uz https://orcid.org/0000-0001-8973-3590

References

1. Thompson LD. Update from the 4th edition of the World Health Organization classification of head and neck tumours: tumours of the ear. Head Neck Pathol 2017; 11: 78–87.
2. Das R, Nath G, Bohara S, et al. Ceruminous adenoma: a rare tumor diagnosed on cytology with histological correlation. J Cytol 2017; 34: 168–170.
3. Behera M and Chatterjee S. A case of syringocystadenoma papilliferum of eyelid with literature review. Indian J Ophthalmol 2015; 63: 550–551.
4. Liu H, Zhang Y, Zhang T, et al. Correlation between the pathology and clinical presentations in patients with adenoid cystic carcinoma of the external auditory canal. Head Neck 2017; 39: 2444–2449.
5. Magliulo G, Ciniglio Appiani M, Colicchio MG, et al. Mucoepidermoid carcinoma of the external auditory canal. *Otol Neurotol* 2012; 33: e21–22.

6. Prasad V, Shenoy VS, Rao RA, et al. Adenoid cystic carcinoma—a rare differential diagnosis for a mass in the external auditory canal. *J Clin Diagn Res* 2015; 9: MD01–2.

7. Lassaletta L, Patron M, Oloriz J, et al. Avoiding misdiagnosis in ceruminous gland tumours. *Auris Nasus Larynx* 2003; 30: 287–290.

8. Niemczyk E, Niemczyk K, Maldyk J, et al. Ceruminous adenoma (ceruminoma) arising in a nevus sebaceus of Jadassohn within the external auditory canal of a 3 year-old boy—a case report. *Int J Pediatr Otorhinolaryngol* 2015; 79: 1932–1934.