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# Ceruminous adenoma mimicking otitis externa

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**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 transcanal 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

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## 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] We present a new case of CA mimicking external otitis and review the relevant literature on CA.

## 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, $3 \times 500$ mg/day) for 7 days.

The tumor was off-white in color with the dimensions of $20 \times 12 \times 15$ mm$^3$. Microscopic examination showed glandular structures which were composed of two cell layers: epithelial and myoepithelial. The inner layer consisted of

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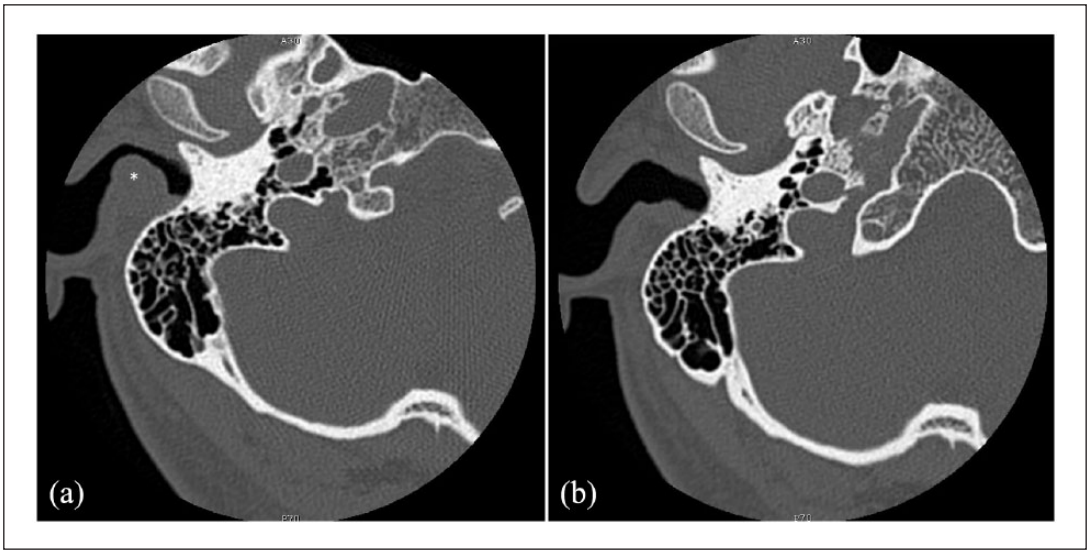


**Figure 1.** (a) CT imaging (horizontal plane) of CA (asterisk) in the right external auditory canal with no bone invasion. (b) A control CT scan was performed 1 year after surgery showing no recurrence.

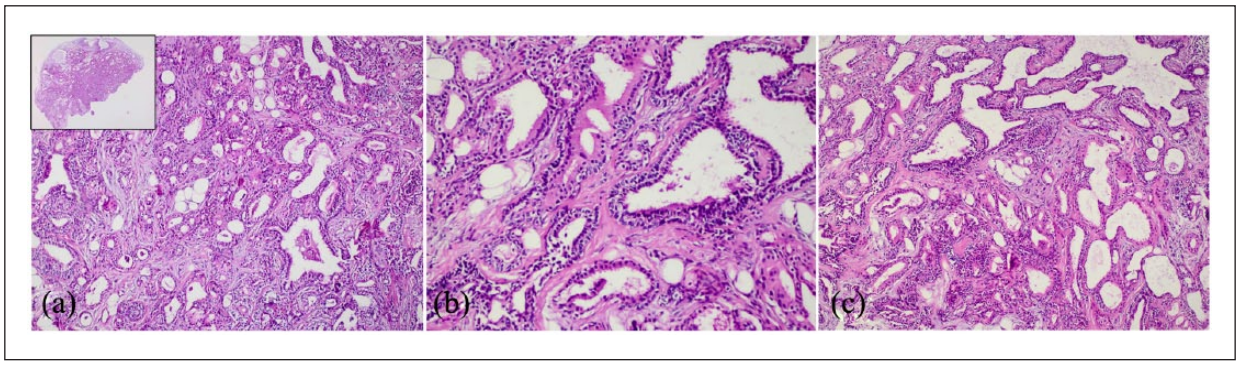


**Figure 2.** (a) The circumscribed but unencapsulated tumor (inset: H&E, 20×) of the ceruminous gland is composed of two cell populations: epithelial cells in the inner layer and myoepithelial cells in the outer layer (H&E, 200×). (b) Glandular structures show ceruminous secretion in the luminal cells (H&E, 400×). (c) Glandular structures are separated by fibrous connective tissue (H&E, 200×).

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-diethylenetriaminepentaacetic 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.[1,2] In the literature, those tumors have been excised using a transmeatal approach with a wide 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

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### Ethics approval

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