Ceruminous adenoma: A rare tumor diagnosed on cytology with histological correlation

ABSTRACT
Ceruminous adenoma (CA) is a rare, benign tumor of the ceruminous gland found in the cartilaginous part of the external auditory canal (EAC). The tumor is diagnosed on histopathology and shows a characteristic dual population of luminal epithelial cells and basal myoepithelial cells. However, CA can be diagnosed on fine-needle aspiration cytology (FNAC) prior to surgery and the cytopathologist should be well aware of its cytological findings to avoid any misdiagnosis. The tumor shows an excellent prognosis with possible recurrence and malignant transformation. The present case emphasizes the cytological features of CA, which have been scantily described in the literature and highlights the role of cytopathologists in preoperative diagnosis.

Key words: Benign tumor; ceruminous adenoma (CA); ceruminous gland; external auditory canal (EAC); fine-needle aspiration cytology (FNAC)

Introduction
Ceruminous glands are specialized apocrine glands found in the cartilaginous part of the external auditory canal (EAC) and their function is to secrete cerumen. Primary tumors of EAC are extremely rare and ceruminous adenoma (CA) is an uncommon, benign tumor that arises from the ceruminous gland. The tumor can appear at any age ranging from 24 years to 85 years and is usually diagnosed on histopathology with an excellent prognosis after surgical excision.[1] Histopathological features of the tumor are well-established but the cytological features are not well-documented in the literature.

Here we present a case of CA, which was diagnosed on fine-needle aspiration cytology (FNAC). The present case highlights on the unique cytoarchitectural pattern, along with the cellular features, which describes the role of FNAC in preoperative diagnosis in cases of CA.

Case Report
A 45-year-old male came to our otorhinolaryngology department with complaints of earache and discharge from the right ear since the past 15 days. The patient had a habit of scratching his itching ear with a matchstick from childhood. Before 15 days, the tip of the matchstick remained inside while scratching his ear and gradually he developed pain and ear discharge.

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On examination, a mass was present on the cartilaginous part of the EAC, along with mucopurulent discharge in the EAC. The tympanic membrane was not visible. Otoendoscopy revealed a piece of matchstick present in the EAC and audiometry revealed mild conductive hearing loss.

The patient was advised FNAC. The aspirate was grossly gray-white fluid. On microscopy, the smears were cellular, revealing cohesive monolayered sheets as well as a few papillary fragments of epithelial cells having round to oval, bland-appearing nuclei and inconspicuous nucleoli [Figures 1a-c]. These cells displayed abundant eosinophilic cytoplasm. Spindle-shaped myoepithelial-like cells and a few plasmacytoid cells were also noted [Figures 1a and c]. Eosinophilic stromal fragments with intermixed fibroblastic spindle-shaped cells were present abundantly. At places, these fragments were admixed with the epithelial cells [Figure 1d]. Depending on the cytological and clinical findings, benign adnexal tumor, most probably CA was diagnosed and the patient was advised surgical excision with a wide resection margin.

Surgically, the removed specimen grossly measured 1.5 × 1.5 × 1.0 cm and was covered by skin at one side [Figure 2a]. Microscopically, the section showed unencapsulated tumor composed of proliferating cells arranged as tubular glands, solid sheets, papillae, and cysts. The tubules and glands were lined by a dual population of cells [Figures 2b and c]. Luminal epithelial cells were tall columnar to cylindrical with bland round to oval nuclei and abundant granular eosinophilic cytoplasm. These cells were showing decapitation secretion at places [Figure 2d]. The outer layer of the cells was basaloid in appearance. Adjacent stroma was fibromyxoid. Thus, the tumor was confirmed as CA on histopathology.

**Discussion**

CA is a rare benign tumor that arises from the ceruminous gland, found in the cartilaginous part of EAC, and is associated with chronic irritation. It is commonly found in felines and the Canidae family but is a rare occurrence in humans; it is seen in adults and its incidence is equal between males and females.[2] It usually appears as a polypoidal mass with a smooth or ulcerated surface with serosanguineous discharge with varying degrees of hearing loss, otalgia, and rarely bleeding; however, pain and facial nerve palsy are features of malignant transformation.[3]

The cytological features have not been extensively described in the literature to the best of our knowledge. Till date, only one case series of two cases was found where CA was diagnosed on FNAC.[3] The present case showed a unique spectrum of findings ranging from papillae formation, tightly cohesive epithelial and myoepithelial cells to the presence of scattered plasmacytoid cells and fibromyxoid stromal fragments. These cytological findings correlated very well with the characteristic histopathology of CA. Hence, knowledge of the cytological findings can improve the prognosis of the patient, prevent unnecessary surgical interventions and extensive surgery by a simple investigation such as FNAC.

Grossly, CA has been described to be a circumscribed solid mass covered by ulcerated or nonulcerated epithelium with its size variance of 0.4-2.0 cm. Rarely cystic change may
be seen. Microscopically, it is an unencapsulated tumor having a varied architectural pattern ranging from a closely packed gland, areas of cystic dilatation, solid areas, and papillary formation. The tumor is composed of two types of cells comprising luminal cuboidal to low columnar cells with intensely eosinophilic cytoplasm, having decapitation secretion. Epithelial cells are surrounded by a layer of myoepithelial cells with scanty cytoplasm and smaller hyperchromatic nuclei. The present case highlights almost all of the characteristic histopathological findings as has been previously described.

Immunohistochemically, CA shows positivity for CK7 by the luminal cells and positivity for S100, CK5/6, and p63 by the basal cells. Immunohistochemistry is not needed for its diagnosis but can be used to highlight its biphasic behavior.

CA should be differentiated from ceruminous adenocarcinoma, neuroendocrine adenoma of the middle ear, pleomorphic adenoma of the parotid gland, meningioma, and paraganglioma. A knowledge of the characteristic histopathological features is almost sufficient to exclude all of the above tumors. The tumor is treated by surgical excision with an adequate margin of normal tissue and recurrence has been reported with the presence of residual tumor tissue after surgical excision. It shows excellent prognosis after complete surgical excision and our case showed no sign of recurrence after 1 year of follow-up and also showed signs of improved hearing in the subsequent outpatient department visits.

Malignant transformation has been described in approximately 70% of the cases of CA with a possible intracranial extension and distant metastasis. Mansour et al. suggested that the tumor should be reported as “of uncertain malignant potential” and long-term studies are needed to determine the potential for malignancy in all ceruminous gland tumors.

Conclusion

To conclude, CA is a rare benign tumor of EAC with potential for malignant transformation. It can be diagnosed on FNAC and cytopathologists should be well aware of its cytological findings to make a preoperative diagnosis, which will affect the management plan of the tumor.

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Conflicts of interest

There are no conflicts of interest.

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