Ceruminous Gland Adenoma of External Auditory Canal- A Case Report

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Abstract

Ceruminous Adenoma (CA) is a benign neoplasm of ceruminous glands. Developing exclusively in the external auditory canal (EAC) with benign clinical behavior and was first reported by Hang in 1894. It is rare in humans and poses a diagnostic problem for the clinician, due to the variety of clinical presentations. This tumor is mainly composed of the ceruminous gland cells (modified apocrine sweat glands). These glands are localized deep in the skin mostly in the cartilaginous part of the external auditory meatus. We are presenting a 45 year old female patient who was referred to our otology department with a two year history of right ear blockage with progressive hearing loss; her other medical history was unremarkable. History was negative for ear discharges, tinnitus, trauma and previous ear surgery. The physical examination and otoscopy showed soft mass of skin that fulfilled half of the posterior inferior portion of the external auditory canal (EAC) that was completely excised under general anesthesia utilizing a postauricular approach. The excised tumor specimen after proper staining confirmed the diagnosis of ceruminous adenoma. In conclusion the CA is a benign rear tumor raised from ceruminous gland in EAC slowly growing when getting bigger can block the EAC leading to hearing loss; it can be treated by complete surgical excision along with over line skin to prevent recurrence.

Keywords: Auditory Canal Adenoma; Ceruminous Gland

Introduction

Ceruminous Adenoma (CA) is a benign neoplasm of ceruminous glands. Developing exclusively in the external auditory canal (EAC) with benign clinical behavior and was first reported by Hang in 1894 [1]. The CA is located primarily in the cartilaginous portion of the EAC. Ceruminous tumors have been reported in patients as young as 12 years and as old as 92 years [2,3]. The mean age for patients with the most common benign tumor, CA is the sixth decade [4]. Ceruminous pleomorphic adenomas are less common than CA; the mean age for affected individuals is also in the sixth decade [5-8].

CA and pleomorphic adenomas have been reported in both sexes. The sex distribution for CA is nearly 50-50 [4]. A few more cases of ceruminous pleomorphic adenoma have been reported in men than in women, but the overall number of cases is too small to suggest a statistically significant difference [4-8].

They demonstrate a dual cell population of basal myoepithelial-type cells and luminal ceruminous cells [9]. The most often symptom of the CA of the EAC is the conductive hearing loss. It is treated by complete surgical excision with excellent prognosis [10]. It is rare in humans and poses a diagnostic problem for the clinician, due to the variety of clinical presentations [1]. We present herein a new case of CA to delineate the salient clinic-pathological features of this rare tumor.

Case Report

The present case report is about a 45 year old female patient who was referred to our otology department with a two year history of right ear blockage associated with mild hearing loss and on and off otolgia. Her other medical history was unremarkable. History was negative for ear discharges, tinnitus, trauma and previous ear surgery. The physical examination and otoscopy showed soft mass of skin that fulfilled half of the posterior inferior portion of the EAC (Figure 1) and Weber’s test was done to check the hearing loss of right ear. A Computed tomography (CT) scan of the temporal bone revealed a soft lesion occupying the EAC with no signs of bone destruction (Figure 2). Tympanic cavity and mastoid cleft were normal (Figure 3). There was no intracranial involvement. Intraoperative findings showed diffused cystic swelling and whitish cheesy fluid discharge. A specimen from the discharges was submitted for microbiological study, it was negative for microorganism. The lesion was excised completely along with over lining skin under general anesthesia utilizing a postauricular approach (Figure 4). The exposed bone covered with temporalis fascia graft.
Macroscopically the excised tumor specimen was a fusiform skin specimen measuring 1.5 cm in dimension (Figure 5). Microscopic examination showed that lesions consisted of two types of cells including inner cuboidal or columnar cells with eosinophilic cytoplasm and apical snouts and outer spindled myoepithelial cells with hyperchromatic nuclei (Figure 6). Mitotic figures, pleomorphism, necrosis, and invasiveness were not seen.

Immunohistochemistry (IHC) has greatly assisted surgical pathologists in the diagnosis of neoplastic diseases. Currently, IHC has a wide variety of uses, including determining diagnosis, determination of the primary site of origin for metastatic tumors, classification of neoplasms, and use as a prognostic and predictive biomarker. We used immunohistochemistry in our case by ultraview universal DAB detection kit method.

Immunohistochemical studies also confirmed the presence of two distinct cell types. CA demonstrate labeling of the luminal cells expressed cytokeratin 7 (CK7) (Figure 7) while peripheral (basal) cells expressed keratin 5/6, S100 protein and p63 (Figure 8). Cerumin is positive for PAS (Figure 9). Based on these histopathological and immunohistochemical findings the diagnosis of ceruminous gland adenoma was established.

The postoperative course was uneventful and the excision site was well healed with no evidence of recurrence until seven months follow up.

Discussion

Ceruminous glands are modified sweat glands, confined to the skin of the cartilaginous part of the EAC. Tumors arising from these glands are extremely rare and resemble those arising from sweat glands elsewhere in the body [4,11-13]. Wetli, et al, indicated that malignant tumors out-number benign (2.5:1) with equal male to

![Figure 3: Computed tomography scan of temporal bone showing the soft tissue mass in the outer 1/3 of the right External auditory canal.](image)

![Figure 4: Intra-operative photo showing the Right External auditory canal cystic mass (blue arrow) with normal tympanic membrane (green arrow).](image)

![Figure 5: The Excised Mass measuring 1.5 cm.](image)

![Figure 6: Microscopic Examination (H & E stain) of Inner cuboidal cells contain eosinophilic cytoplasm and apical snouts (arrow).](image)

![Figure 7: Immuno-histochemical staining of the inner luminal cells are strongly positive for CK7 (arrow).](image)
female distribution [14]. Clinically, these tumors may have a long history and many years may lapse before presentation [15-17] that has been confirmed by our case.

The differential diagnosis are ceruminous adenoma, ceruminous adenocarcinomas, neuro-endocrine adenoma of middle ear, parotid pleomorphic adenoma, meningioma and paraganglioma, which are difficult to diagnose at the initial stage, hence histology is only diagnostic [10].

CA demonstrates a dual cell population of basal myoepithelial-type cells and luminal ceruminous cells. Both diffusely and strongly immunoreactivity with CK7 of the luminal cells and with actin and S-100 protein of the basal myoepithelial-type cells, helps us to distinguish this tumor from other neoplasms that occur in the region [18]. The presence of a dual cell population and the absence of malignant features led us to a diagnosis CA. Immunohistochemical staining for cytokeratin (specifically CK7) which highlight the luminal cells and for basal/myoepithelial cell markers like CK 5/6, S-100 and p63 may be done to further demonstrate the dual cell population. Complete or adequate local excision is the treatment of choice; however, residual tumor often remains because of the difficulty of surgery at this location leading to recurrence [4,19,20]. However, there is no evidence of recurrence for these tumors in the literature, with a mean follow-up time of 15 years [11,12,14]. Our patient underwent total surgical excision of the mass along with the over lining skin and showed good healing process without signs of recurrence up to seven months later.

**Conclusion**

Ceruminomas are rare tumors arising from the ceruminous gland, which is a modified apocrine gland in the skin of the external canal. In our case, the lesion was excised completely using post auricle approach. After surgery the follow up of patient was uneventful. Ceruminous gland adenoma at hat are pedunculated rarely recur because of the ease of adequate surgical excision.

**References**


