

CERUMINOMA REVISITED

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ABSTRACT

Ceruminoma is a catch-all term that has caused much confusion both in the literature and in clinical practice in regard to the specific histologic diagnosis and proper treatment for tumors arising from the ceruminous glands of the external ear canal. The term *ceruminoma* has been used in the past to refer to both benign and malignant lesions. To clarify the terminology and better determine appropriate treatment, two cases of benign adenoma of the ceruminous glands along with their histopathologic findings will be presented. The specific characterization of the individual types of ceruminous gland neoplasms, their clinical manifestations, histopathology, and recommended treatment will then be discussed. Finally, suggestions for the appropriate nomenclature for these rare tumors will be reviewed.

Adenoma of the ceruminous glands is a rare clinical pathologic entity in man.¹ The first such reported case was credited to Haug in 1894.² The term *ceruminoma* has been used in the past to describe tumors arising from the ceruminous glands of the external ear canal. Much confusion has resulted because of the varied clinical behavior and histologic manifestations of these tumors.³⁻⁵ In 1972, Wetli and colleagues⁶ recognized four distinct types of ceruminoma. In 1977, Pulec⁷ added mucoepidermoid carcinoma to the spectrum of possible tumors arising from the ceruminous glands.

Some authors have proposed that because the so-called ceruminous glands of the external auditory canal are modified apocrine sweat glands, similar to sweat glands found elsewhere, the designation *ceruminoma* is a misnomer and unjustifiable.^{4,5,8}

Instead, hidradenoma of the external ear canal was recommended.⁴ Cankar and Crowley⁵ were unable to demonstrate any specific histochemical differences between ceruminous gland tumors and sweat gland neoplasms arising elsewhere in the body.

However, because of the characteristic clinical features of ceruminous gland tumors in addition to histologic appearances, other workers have urged retention of the term.^{3,6,8}

To clarify some of the misunderstanding regarding the term *ceruminoma*, two case reports of benign adenoma of the ceruminous glands, along with their gross appearances and histopathologic descriptions, will be presented. In addition, a discussion will follow based on a review of the literature which will delineate the various types of neoplasms

originating from the ceruminous glands from the standpoint of histology, clinical course, and therapy. Finally, recommendations regarding the appropriate nomenclature for ceruminous gland tumors will be reviewed.

CASE ONE

A 52-year-old man presented to the Otolaryngology Clinic at the New York Eye and Ear Infirmary complaining of "something in the ear." He denied ear infections, hearing loss, pain, or discharge. Otoscopy revealed a round, smooth, 6-mm skin-lined mass in the left external auditory meatus that appeared to arise from the posterior canal wall (Fig. 1). The tympanic membrane was not visualized. The right ear was normal on otoscopy. There was no facial weakness or cervical adenopathy. The remainder of the examination was unremarkable. An audiogram revealed normal hearing for both ears. Mastoid films demonstrated well-aerated mastoids bilaterally with no destructive process noted. Biopsy of the mass showed normal squamous epithelium with "pseudoglandular formation" in the submucosa.

The patient was taken to the operating room where an excisional biopsy was performed with wide excision of the base. There was no evidence of deep invasion or bony destruction. The tympanic membrane was intact. Microscopic examination of the lesion demonstrated a benign adenoma of the ceruminous glands (*ceruminoma*) (Fig. 2). The patient

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Figure 1. Case 1. Intraoperative gross appearance of ceruminous adenoma of left ear. (Original magnification $\times 1$.)

has been followed for five months with no evidence of recurrence.

CASE TWO

A 48-year-old man presented to the Otolaryngology Clinic at the New York Eye and Ear Infirmary also complaining of "something in the ear." Otolaryngoscopic examination revealed a moderate amount of impacted cerumen and a round, 3-mm mass arising from the floor of the right ear canal. The remainder of the ear canal and tympanic membrane were within normal limits. The left ear canal and tympanic membrane were also normal. An excisional biopsy was performed in the clinic. Microscopic examination demonstrated benign adenoma of the ceruminous glands (Fig. 3). The patient was followed for six months postoperatively with no evidence of recurrence.

DISCUSSION

Ceruminous glands are modified apocrine sweat glands which lie deep in the dermis of the cartilaginous portion of the external auditory canal.⁹ As seen with hematoxylin and eosin stain under light microscopy, ceruminous glands are large, irregular, and apocrine. There is an inner layer of cuboidal to columnar secretory cells and an outer layer of elongated myoepithelial cells similar to apocrine sweat glands found elsewhere in the body.^{2,9,10}



Figure 2. Case 1. Ceruminous adenoma showing glands with two-layered epithelial cells—an inner, eosinophilic and columnar layer, and an outer layer of elongated myoepithelial cells. (Hematoxylin-eosin; original magnification $\times 40$.)

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Figure 3. Case 2. Ceruminous adenoma showing lipochrome pigments on its mucosal surface (dark areas). (Hematoxylin-eosin; original magnification $\times 60$.)

The histologic features of tumors arising from the ceruminous glands exhibit considerable variation, although all manifest to a greater or lesser degree a two-layered epithelial structure analogous to that of normal sweat glands: an inner oxyphilic columnar layer and an outer myoepithelial layer. The tumors also exhibit a variable degree of interglandular stroma.³ In the past the histomorphologic classification of ceruminous gland tumors had been implicit and inexact.²⁻⁵

Much confusion concerning these tumors resulted from use of the catch-all, ambiguous term "ceruminoma," which included the full spectrum of ceruminous gland tumors ranging from a benign adenoma to metastatic adenoid cystic carcinoma.^{3-5,9}

More recently, authors have identified distinct patterns of ceruminous gland neoplasms^{6,7,9} and have encouraged a more exact nomenclature based on the specific histopathology of the lesion.

CLASSIFICATION

Ceruminous adenoma is a well-differentiated, benign, localized proliferation of glands histologically similar to normal ceruminous glands which resembles apocrine tumors located elsewhere.⁶ Adenoma of the ceruminous glands produces symptoms related to obstruction of the ear canal from the mass. Pain is seldom present unless there is an associated otitis of the ear canal. Otologic examination reveals a polypoid mass in the external auditory meatus usually not associated with a history of otorrhea or previous ear infection.³ The mass is covered with squamous epithelium or granulation tissue.

Accurate pathologic diagnosis is essential. Often the pathologist will have limited or no experience with them. Thus the clinician should be alert to the possibilities and ascertain whether the lesion is benign or malignant. Conservative excision is adequate to produce a lasting cure.⁷ Including this tumor with the malignant varieties under the catch-all term "ceruminoma" has created confusion in the literature regarding the behavior of this neoplasm and the appropriate treatment.

Johnstone and colleagues⁴ questioned why some tumors appear invasive and tend to recur while others do not. Cankar and Crowley⁵ concluded that even in those tumors appearing histologically benign, an adequate margin of uninvolved tissue should be included in the block resection. O'Neil and Parker¹¹ believed that most of the apocrine gland tumors of the external auditory canal should be regarded as malignant and recommended radical treatment when first diagnosed. However, this has been shown to be incorrect, since the adenoma is a benign lesion, requiring only local excision.⁷

Primary adenoid cystic carcinoma in the external ear canal (frequently called "cylindroma" in the older literature) is an uncommon neoplasm. Patients invariably complain of pain. The natural history of the disease is characterized by a long duration, ranging from ten to thirty years. It has a propensity for perineural invasion and metastasis. Death usually results from local invasion and distant metastatic spread, often to the lungs. The tumor has small, dark cells that are arranged in a solid pattern or with cystic spaces rendering a "swiss cheese" pattern.^{6,7} Pulec⁷ recommends wide excision of the entire external auditory canal; extensive radical mastoidectomy, 487

including, if necessary, dura; total parotidectomy; and excision of the mandibular condyle and involved surrounding structures. Radiation has little to offer the patient.

Ceruminous adenocarcinoma may occur in any degree of malignancy.⁷ The neoplasm of low malignancy with well-differentiated tumor cells may be difficult to discern from normal ceruminous glands. Wetli and associates⁸ emphasized that invasion is the crucial histologic feature, as the tumor may be cytologically indistinguishable from the benign form. There may be extensive local infiltration involving the middle ear, mastoid, base of skull, parotid gland, etc. Pulec⁷ described five patients with ceruminous gland adenocarcinoma, one of whom was lost to follow-up. The remaining four died within four years of onset of the disease. All had pain and two developed facial paralysis. Wide surgical excision as described for adenoid cystic carcinoma should be performed, combined with radiation.

Pleomorphic adenoma or mixed tumor of the ceruminous glands consists of nests of epithelial cells in a myxoid, pseudocartilaginous or hyaline stroma which contains mucin.⁶ Pulec⁷ described for the first time mucoepidermoid carcinoma of the ceruminous glands.

Malignant tumors of the ceruminous glands outnumber benign by about 2.5 to 1—a sharp contrast to the rarity of malignancy arising in cutaneous apocrine tumors.⁶

CONCLUSIONS

Confusion regarding the appropriate terminology for these rare tumors is based on the controversy regarding recommended treatment. Also, the term "ceruminoma," referring to both benign and malignant neoplasms, has been misleading. Since the various histologic types of ceruminous gland tumors have been identified and characterized, appropriate therapy must be individualized based on the specific

histology of the lesion. In addition, there appears to be no advantage to retaining ceruminoma as a generic term, since it has been established that past references to ceruminoma in fact represent a heterogeneous group of neoplasms.¹

If therapeutic strategies are to be defined based on specific histologic diagnoses, then specific terminology, such as "ceruminous gland adenoma," "adenocarcinoma," and so on should be used. The term "ceruminoma" should be limited to benign adenoma of the ceruminous glands, and the other, more specific terms should be used when describing the actual histopathology. Treatment should then be determined based on the exact identification of the histology of the lesion in question.

REFERENCES

1. Michel RG, Woodard BH, Shelburne JD, et al: Ceruminous gland adenocarcinoma: A light and electron microscopic study. *Cancer* 41:545-553, 1978
2. Adler HJ, Sommer I: Adenoma of the ceruminous glands. *Arch Otolaryngol* 99:533-535, 1944
3. Batsakis JG, Hardy GC, Hishiyama RH: Ceruminous gland tumors. *Arch Otolaryngol* 86:92-95, 1967
4. Johnstone JM, Lennox B, Watson AJ: Five cases of hidradenoma of the external auditory meatus: So-called ceruminoma. *J Pathol Bacteriol* 73:421-427, 1957
5. Cankar V, Crowley H: Tumors of ceruminous glands: A clinicopathological study of seven cases. *Cancer* 17:67-75, 1964
6. Wetli CV, Pardo V, Millard M, et al: Tumors of ceruminous glands. *Cancer* 29:1169-1178, 1972
7. Pulec J: Glandular tumors of the external auditory canal. *Laryngoscope* 87:1601-1612, 1977
8. Arora YR: Ceruminoma of the external auditory meatus. *J Laryngol Otolaryngol* 569-572, 1964
9. Althaus SR, Ross JAT: Cer Arch Otolaryngol 92:40-42, 1970
10. Leitner MJ: Adenoma of the ceruminous glands. *Am J Clin Pathol* 22:466-471, 1952
11. O'Neill PB, Parker RA: Sweat gland tumors ("ceruminoma") of the external auditory meatus. *J Laryngol* 71:824-831, 1957

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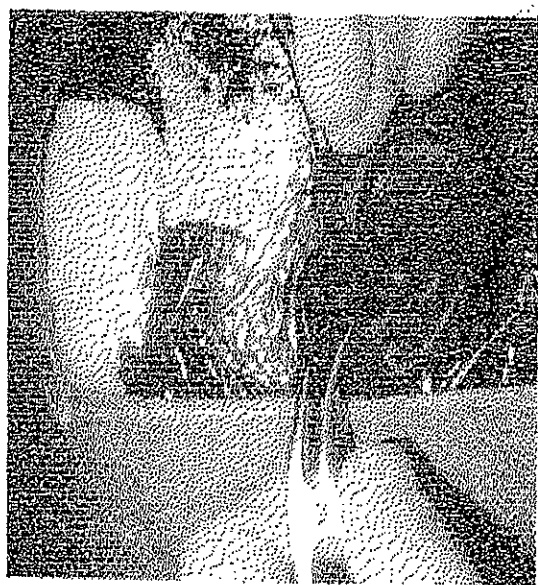


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DISCUSSION

Ceruminous glands are modified apocrine sweat glands which lie deep in the dermis of the cartilaginous portion of the external auditory canal.⁸ As seen with hematoxylin and eosin stain under light microscopy, ceruminous glands are large, irregular, and apocrine. There is an inner layer of cuboidal to columnar secretory cells and an outer layer of elongated myoepithelial cells similar to apocrine sweat glands found elsewhere in the body.^{2,9,10}

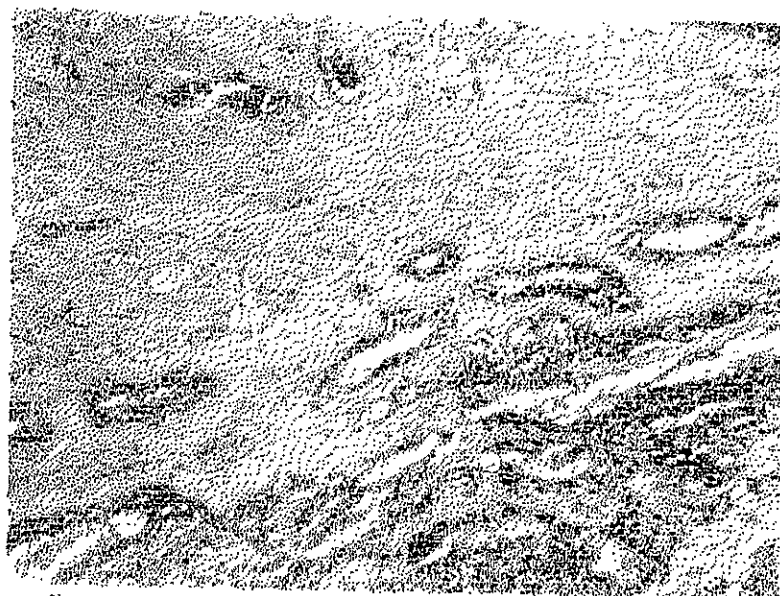


Figure 2. Case 1. Ceruminous adenoma showing glands with two-layered epithelial cells—an inner, eosinophilic and columnar layer, and an outer layer of elongated myoepithelial cells. (Hematoxylin-eosin; original magnification $\times 40$.)

The histology of the ceruminous gland, although it differs from that of normal apocrine sweat glands, is similar to that of other sweat gland tumors. The histology of the ceruminous gland is characterized by the presence of a large, irregular, and apocrine glandular structure with a two-layered epithelium. The inner layer consists of cuboidal to columnar secretory cells, and the outer layer consists of elongated myoepithelial cells. The glandular structure is surrounded by a fibrous stroma. The histology of the ceruminous gland is similar to that of other sweat gland tumors, such as the eccrine sweat gland tumor and the apocrine sweat gland tumor. The histology of the ceruminous gland is characterized by the presence of a large, irregular, and apocrine glandular structure with a two-layered epithelium. The inner layer consists of cuboidal to columnar secretory cells, and the outer layer consists of elongated myoepithelial cells. The glandular structure is surrounded by a fibrous stroma. The histology of the ceruminous gland is similar to that of other sweat gland tumors, such as the eccrine sweat gland tumor and the apocrine sweat gland tumor.

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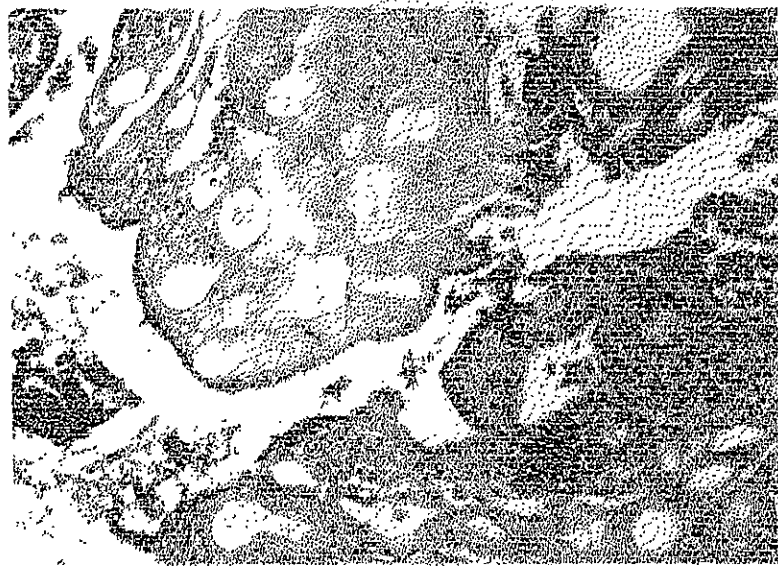


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