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Ceruminous Adenocarcinoma of the External Auditory Canal: A Case Report and Review of the Literature

H. Boudinar[1*], I. Horrane[1], Z. El hafi[1], Z. Arkoubi[1], R. Bencheikh[1], M. A. Benbouzid[1], L. Essakalli[1]

[1]Department of Otorhinolaryngology – Head and Neck Surgery, Ibn Sina University Hospital, Rabat, Morocco

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***Corresponding author:** H. Boudinar
Department of Otorhinolaryngology – Head and Neck Surgery, Ibn Sina University Hospital, Rabat, Morocco

Abstract | **Case Report**

Background: Ceruminous gland adenocarcinoma of the external auditory canal (EAC) is an uncommon malignant tumor with nonspecific clinical features and often delayed diagnosis. ***Case Presentation:*** A 72-year-old woman had purulent otorrhoea of the right ear and ipsilateral facial paralysis. Otoscopy revealed a mass occupying the right EAC. CT scan result was cholesteatoma. Histopathology showed adenocarcinoma of ceruminous gland. The patient underwent wide surgical excision with adjuvant radiotherapy and has been in complete remission at 3 years. ***Conclusion:*** Early biopsy is emphasized in this case in atypical chronic otorrhoea, and local excision with wide margins and adjuvant radiotherapy are advised as an effective treatment strategy.

Keywords: Cermuinous adenocarcinoma – EAC – biopsy- Radiotherapy.

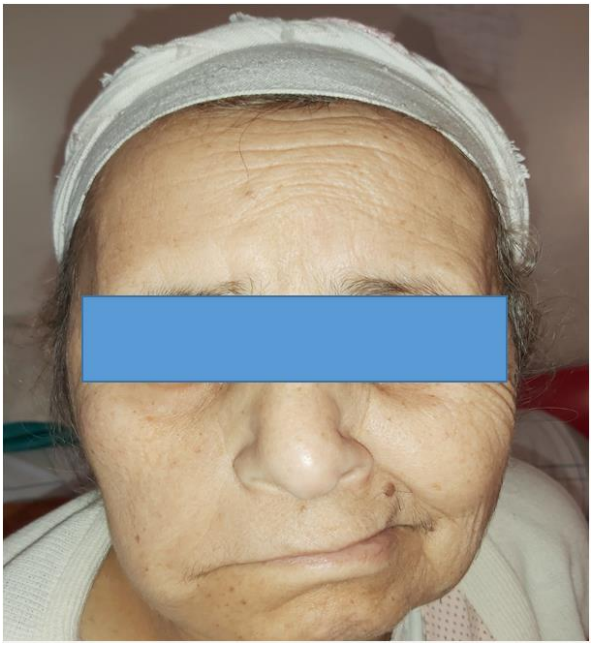
INTRODUCTION

Ceruminous gland tumors of the external auditory canal (EAC) are extremely rare, accounting for a negligible percentage of all ear canal neoplasms. Malignant subtypes, particularly adenocarcinomas, are even less common [1,2]. These tumors frequently present with symptoms that mimic benign EAC conditions such as cholesteatoma or chronic otitis media, due to overlapping clinical and radiologic features. This often results in delayed diagnosis and management [3,4].

Histopathological examination remains the gold standard for accurate diagnosis. We report a case of ceruminous gland adenocarcinoma in a 72-year-old woman, treated successfully with surgical excision followed by radiotherapy, achieving long-term remission of three years. Our aim is to highlight and discuss the diagnostic and therapeutic approaches to this rare malignancy.

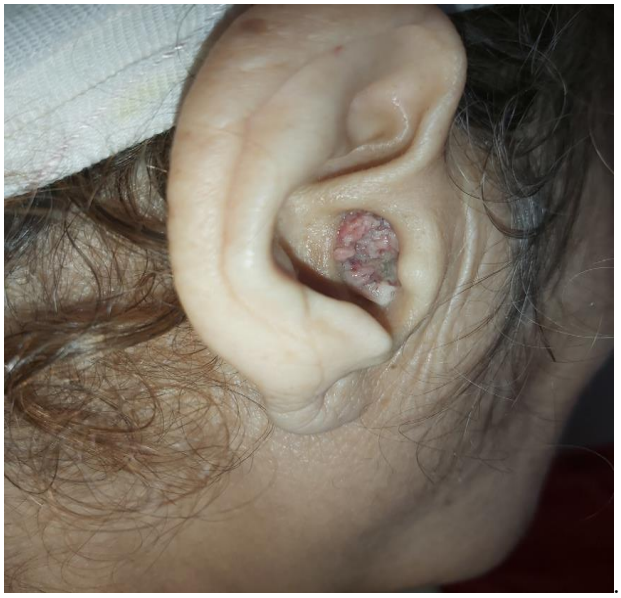
CASE PRESENTATION

A 72-year-old female presented to our department with right-sided otalgia, purulent otorrhea, auricular pruritus, hypoacusis, tinnitus, peripheral facial palsy (Grade IV, House-Brackmann), and cephalalgia, without associated vertigo. These symptoms prompted consultation at the ENT-CCF emergency unit at the Hospital of Specialties of Rabat.



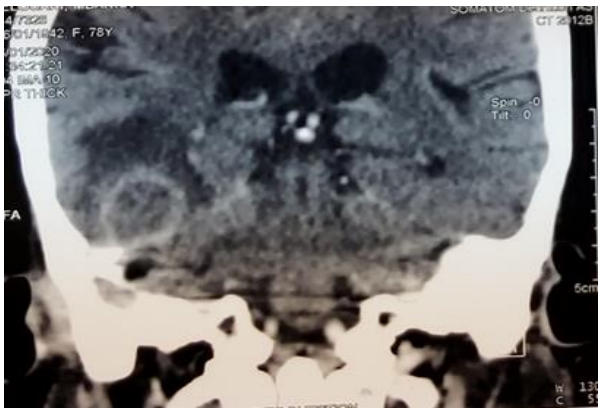
Clinical photograph of our patient showing House-Brackmann grade IV facial paralysis

Otoscopic examination revealed a polypoid, budding mass completely obstructing the right EAC, along with purulent discharge. The facial nerve was involved, while other cranial nerves remained intact.

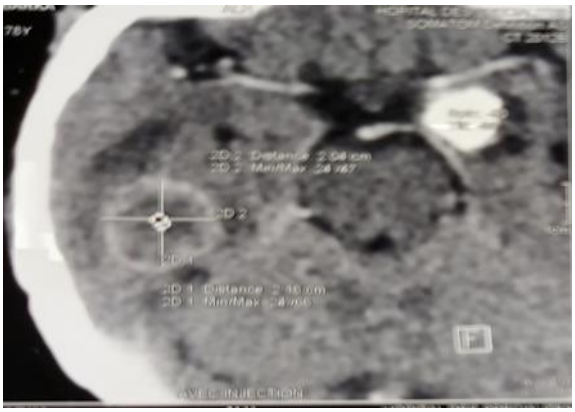


Clinical image showing macroscopical aspect of the EAC tumor

Computed tomography (CT) of the temporal bone demonstrated a homogeneously enhancing isodense lesion involving the right EAC, with lysis of its bony walls and extension into the middle ear. There was complete erosion of the ossicular chain, tegmen tympani, and mastoid cavity, with endocranial extension into the basitemporal region. The lesion also encased the second segment of the facial nerve and destroyed the lateral semicircular canal, initially suggesting an aggressive cholesteatoma.



CT scan in coronal section showing the EAC tumor and its extension



CT scan in axial section showing the endocranial extension of the tumor

Histological evaluation of a deep biopsy revealed infiltrative glandular structures composed of cuboidal to columnar epithelial cells within a desmoplastic stroma, consistent with ceruminous gland adenocarcinoma. Based on the 1991 Pittsburgh TNM classification, revised in 2002, the tumor was staged as T4cN0M0 [5].

Surgical management consisted of en bloc resection with tumor-free margins. This was followed by intensity-modulated radiotherapy (IMRT) at a total dose of 60 Gy. Given the absence of clinical lymphadenopathy, cervical lymph node dissection was not performed. The patient has remained disease-free for three years based on clinical and imaging follow-up.

DISCUSSION

Malignant tumors of the EAC and middle ear are rare and include both epithelial and glandular types. Adenoid cystic carcinoma (ACC), when arising in the external ear, is often termed ceruminous adenocarcinoma. ACCs more commonly occur in salivary glands, respiratory mucosa, and lacrimal glands. Rare locations include the breast, cervix, Bartholin's and Cowper's glands, and the skin [1,6,7].

The histogenesis of ceruminous adenocarcinoma remains under debate, though it is generally accepted that these tumors originate from modified apocrine sweat glands [1]. Diagnosis is often delayed due to nonspecific symptoms and slow tumor progression. Refractory otalgia is the most common presenting symptom [8].

Histopathological confirmation with deep biopsy and immunohistochemistry is essential. Imaging, particularly CT and MRI, plays a key role in evaluating local extension. These tumors may radiologically mimic benign lesions, as seen in our case [3].

The Pittsburgh TNM staging system is commonly used for classifying EAC malignancies:

- T1: Tumor limited to EAC without bony erosion
- T2: Tumor with bony erosion <5 mm
- T3: Bony erosion >5 mm or middle ear/facial nerve involvement
- T4a: Invasion of adjacent soft tissues (auricle, retroauricular skin, parotid, TMJ)
- T4b: Invasion of inner ear or petrous apex
- T4c: Dural or intradural invasion [5]

Our patient was classified as T4c due to the intracranial extension.

Treatment choice depends on:

- Tumor type and location (ear or temporal bone)
- Tumor grade and stage
- Patient's age and overall health status
- Family and medical history
- Patient preference

The cornerstone of treatment is wide en bloc surgical excision with or without adjunctive therapy. ACCs of the EAC often necessitate petro-mastoidectomy, sometimes extending to subtotal petrosectomy. Parotidectomy is critical due to potential perineural spread along the facial nerve [9,10].

Cervical lymph node dissection is reserved for selected cases based on clinical and radiological findings. In our patient, the tumor board recommended surgery followed by adjuvant radiotherapy without lymphadenectomy due to the absence of nodal disease.

ACC located in the EAC tends to behave aggressively, with local recurrences and distant metastases occurring in up to 30% of cases. The most common metastatic sites include the lungs, bones, kidneys, and brain [4,6].

Diagnosis of primary cutaneous ACC should only be made after excluding secondary involvement from adjacent structures or metastases from salivary gland primaries [4].

CONCLUSION

Ceruminous gland adenocarcinoma of the external auditory canal is a rare but aggressive malignancy that may clinically mimic benign conditions. Early biopsy and histopathological evaluation are crucial for timely diagnosis. A multidisciplinary approach involving radical surgical excision and adjuvant radiotherapy offers the best chance for long-term control.

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