Original Research Paper

Volume - 15 | Issue - 09 | September - 2025 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

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CERUMINOUS GLAND CARCINOMA OF EXTERNAL AUDITORY CANAL WITH ADENOID CYSTIC DIFFERENTIATION: A RARE ENTITY

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ABSTRACT External auditory canal tumours are exceedingly rare, accounting for less than 0.2% of all head and neck cancers. Among these, adenoid cystic carcinoma constitutes a small subset, often arising from ceruminous glands. Histologically, adenoid cystic carcinoma is characterized by perineural invasion, contributing to high recurrence rate despite aggressive treatment. The most common malignant tumors of external auditory canal are squamous cell carcinoma, adenoid cystic carcinoma, ceruminous adenocarcinoma. We report a case of 59year old male patient presenting with complaints of Right ear pain, associated with blood tinged discharge from right ear for 5 months. Examination under microscope revealed granulation tissue in right external auditory canal. HRCT TEMPORAL BONES suggested soft tissue density in right external auditory canal, which was biopsied and sent for histopathological examination. HPE suggested ceruminous gland carcinoma with adenoid cystic features and concurrent otitis externa. Lateral temporal bone resection, superficial parotidectomy along with removal of the tumor was done. Radiotherapy was given to the patient. Given the rarity and aggressive nature of this tumor, early recognition and timely intervention with multimodal therapy involving surgery, radiotherapy are essential for improving outcomes.

KEYWORDS : Temporal Bone Neoplasms, Ceruminous Gland Tumor, Adenoid Cystic Carcinoma, Radiotherapy

INTRODUCTION

Ceruminous glands are specialized apocrine glands predominantly found in the skin lining the cartilaginous or membranous portion of the external auditory canal. The true incidence, terminology, behaviour and histopathological classification of these tumors remain uncertain, leading to ongoing controversy regarding their management.

Here we present a case of right external auditory canal mass (small granulation) presenting as right otitis externa. Excisional biopsy, histopathological report turned out to be as malignant tumour of ceruminous gland with adenoid cystic features.

Case Report

A 59year old male patient, farmer, presented to OPD with complaints of excruciating right ear pain, blood tinged discharge from right ear since 5 months

Under Microscopic Examination:

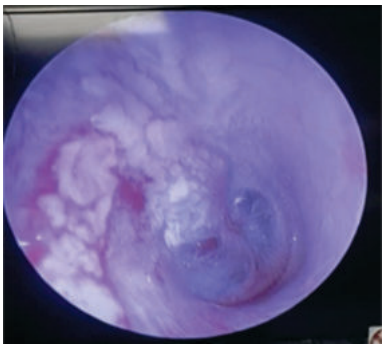


Figure Showing a Small Granulation in the Right External Auditory Canal

Examination Under Microscope: A small granulation of 1*2cm is seen in the cartilaginous part of right external auditory canal, no ulceration.

Tympanic membrane is Intact.

Palpation: Tragal tenderness present.

Investigations:

Routine Blood Investigations – within normal limits.
Patient is known Diabetic and is using medication, RBS-96mg/dl, HBA1C-6

- HRCT Temporal Bones - Soft tissue density noted in lateral 1/3rd of right external auditory meatus. Paucity of posterior and inferior groups of bilateral mastoid air cells noted.
- Biopsy – Ceruminous gland carcinoma of right external auditory canal with adenoid cystic features and concurrent otitis externa.
- Tuning Fork Tests:
  Rinne's test- Negative (BC>AC)
  Weber's test-Lateralised to right ear
- Puretone Audiometry – 45db Moderate conductive hearing loss

Provisional Diagnosis:

Ceruminous Gland Carcinoma With Adenoid Cystic Differentiation Planned For Lateral Temporal Resection And Superficial Parotidectomy Under Ga

Surgical Management

- Under GA, Endotracheal intubation
- Facial nerve monitoring electrodes placed.
- Infiltration with Injection LIGNOCAINE 2% with ADRENALINE and Modified Blair's incision is given extending superiorly into temporoparietal scalp

Soft Tissue Dissection

- Skin flaps elevated in subgaleal and subplatysmal planes.
- Temporalis muscle exposed and reflected anteriorly.
- The external auditory canal skeletonised
- Dissection carried down to the temporal bone, carefully identifying and preserving the Temporomandibular joint capsule and facial nerve trunk.

Lateral Temporal Resection

- Using high speed diamond burrs, bony external auditory canal, (to the level of bony-cartilaginous junction) tympanic membrane, mastoid air cell system, squamous and tympanic parts of the temporal bone removed en-bloc.
- Otic capsule- preserved.
- The tumour removed with 1-2 cm margin.

Superficial Parotidectomy:

- Parotid gland is mobilised superiorly from the Tragal cartilage and posteriorly from the sternocleidomastoid muscle and posterior belly of digastric muscles.
- Greater Auricular nerve is meticulously preserved.
- Following adequate superior-to-inferior exposure and gentle traction, the gland is carefully dissected away from the superficial surface of the Facial nerve.

Figure Showing Exposure of the Right Parotid Gland

Figure Showing Dissection Of Right Parotid Gland And Temporal Resection With External Auditory Canal With Underlying Bone

- Integrity of Facial nerve is checked with Facial nerve monitoring.
- Excised part of Parotid gland and lateral temporal bone specimen including external auditory canal with underlying bone is sent for Histopathological examination.
- Corrugated Drain is placed. Meticulous haemostasis is achieved using Bipolar cautery.
- Wound is irrigated and closed in multiple layers. Mastoid dressing is applied.

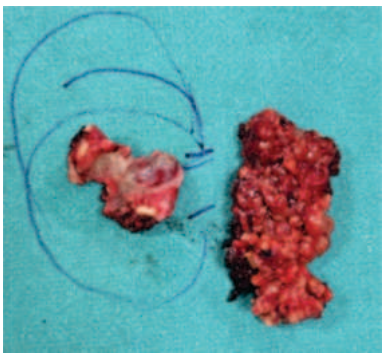


Figure Showing Excised Parotid Gland and Lateral Temporal Bone Specimen Including External Auditory Canal with Underlying Bone

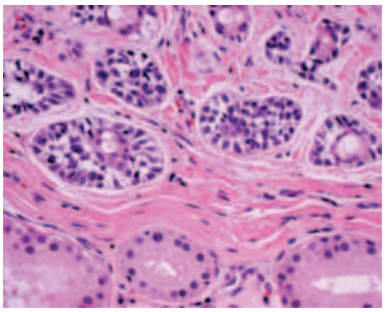
Post-operatively patient developed Facial nerve palsy, Intravenous DEXAMETHASONE at a dose of 8mg administered twice daily for 5 days, then tapered to once daily for the next 5 days.

Injection PIPTAZ 4.5g thrice daily, Injection VANCOMYCIN 1g twice daily, are given intravenously for a period of 7 days.

Drain was removed after 4 days. Mastoid dressing removed after 5 days.

Histopathological Report:

Excised lesion revealed an infiltrative neoplasm originating from the ceruminous glands. Tumour displayed dual cellular populations characteristic of adenoid cystic carcinoma, including basaloid epithelial and myoepithelial cells arranged in cribriform and tubular patterns, embedded in a myxoid stroma. Perineural invasion was noted. Consistent with aggressive behaviour of adenoid cystic differentiation. Foci of chronic inflammatory infiltrates and granulation tissue were present in the adjacent dermis, indicative of concurrent otitis externa. Histological picture is that of "CERMUMINOUS GLAND CARCINOMA OF RIGHT EXTERNAL AUDITORY CANAL WITH ADENOID CYSTIC DIFFERENTIATION".



Histopathological Picture of Ceruminous Gland Carcinoma

Final Diagnosis- Ceruminous Gland Carcinoma Of External Auditory Canal With Adenoid Cytsic Features.

Radiotherapy- External beam radiotherapy was given to the patient with a dose of 70Gy via conventional fractionation for a period of 6weeks. Patient continues to be on regular follow up at 3-month intervals.

DISCUSSION

- Ceruminous gland carcinoma of external auditory canal is an extremely rare malignancy derived from modified apocrine sweat glands and accounts for less than 0.0002% of all head and neck neoplasms and require a high index of suspicion for early diagnosis due to their insidious and non specific clinical presentation.[1]
- Histopathology confirmed ceruminous adenocarcinoma with adenoid cystic differentiation, showing typical cribriform and tubular patterns with prominent perineural invasion—a hallmark of adenoid cystic carcinoma and a key factor associated with local recurrence and poor prognosis [2]
- Given the tumour's infiltrative behaviour and location within the temporal bone, lateral temporal bone resection (LTBR) was performed to achieve oncologic clearance. LTBR remains the surgical standard of care for malignant tumours confined to the cartilaginous and lateral bony EAC [3]
- Involvement or proximity to the parotid gland warranted superficial parotidectomy to address potential regional spread and achieve clear margins.
- Parotidectomy is particularly indicated in tumours abutting the anterior canal wall or with radiological suspicion of parotid involvement [4]
- Perineural invasion, as seen in this case, significantly increases the risk of local recurrence, even after apparent complete resection.
- Therefore, adjuvant radiotherapy is recommended in such settings. Radiotherapy, typically delivered using IMRT techniques, allows conformal dosing to the tumour bed while sparing adjacent neurovascular structures.
- A total dose of 60–66Gy in standard fractionation has been associated with improved local control in ceruminous ACC [5]
- Perineural invasion, positive margins, and solid histologic patterns are associated with worse prognosis[7].
- 5-year overall survival for ceruminous ACC ranges from 50% to 70%, with disease-free survival (DFS) significantly lower, often around 40–50%, reflecting the high rate of local recurrence and delayed metastasis[8,9].
- Notably, distant metastases (commonly to lungs, bone, or liver) may occur several years after initial treatment, underscoring the need for prolonged surveillance.

CONCLUSION

- Ceruminous gland carcinoma with adenoid cystic differentiation is an uncommon malignancy of the external auditory canal, often diagnosed at a late stage due to its subtle clinical presentation and resemblance to chronic otitis externa.

- Histopathology remains crucial for definitive diagnosis, particularly in identifying the biphasic cellular architecture and characteristic perineural invasion of adenoid cystic patterns.
- Complete surgical excision with histologically clear margins remains the mainstay of treatment. Adjuvant radiotherapy may be considered
- Additionally, molecular profiling and immunohistochemical studies may aid in refining diagnostic accuracy and developing targeted therapies.

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