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Recurrent Adenoid Cystic Carcinoma of External Auditory Canal - A Rare Entity in Head and Neck Cancer

Veer Ramanlal Shah[1], Nikunjkumar Naranbhai Patel Lakhavje[2], Venkata Joga Prasanth Mokhasanavisu[3], Shashank Pandya[4]

Abstract: ***Primary adenoid cystic carcinoma (ACC) of the external auditory canal (EAC) is extremely rare in literature and accounts to 5% of malignancies of EAC. Characterized by recurrances and distant spread, its management requires adequate wide surgical resection with or without post op radiotherapy. Therefore we present a rare case of recurrant ACC of EAC which was managed by surgical excision followed by post op radiotherapy.***

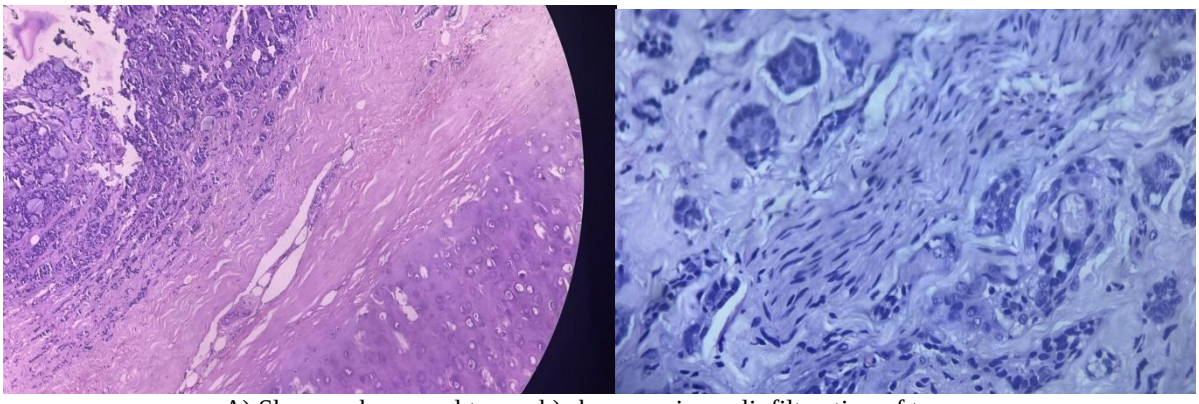
Keywords: Adenoid cystic carcinoma, external auditory canal, recurrant

1. Introduction

Malignancies of external auditory canal are very rare among head and neck malignancies with squamous cell carcinoma constituting around 80% of them. Of the remaining only about 5% are due to Adenoid cystic carcinoma (ACC) [1 - 4]. Adenoid cystic carcinoma in head are usually seen in oral cavity, palate, nasal cavity, nasopharynx [5] The characteristic feature of ACC is its aggressive clinical behavior with high rates of recurrances and distant metastasis [6 - 10]. This is attributed to its propensity for perineural spread and and direct bone invasion [3, 11, 12].

2. Case Report

A 76 yr old male presented to our OPD with history of earache and decreased hearing since 2 years for which he was operated 3 years ago of which reports are not available. He was diagnosed to be a case of Adenoid cystic carcinoma of external auditory canal. He had recurrence of symptoms since 2 months and on examination was found to have a lesion in the superior wall of cartilaginous canal which was sent for histopathological examination, which confirmed the recurrence of disease. CT scan of temporal bone showed a polypoidal soft tissue lesion of 8mm x 9mm x 7mm in the superior wall of cartilaginous canal. Middle ear, osscicles, mastoid, internal acoustic meatus was found to be normal with no bony erosion. Facial nerve was clinically and radiologically normal. Patient underwent sleeve resection of the lesion with modified radical neck dissection type III with canaloplasty with fascia lata graft. Post operative healing was good and patient was referred for post op radiotherapy and has been on follow up since then. Its unique histological characteristics are presented below.



A) Shows submucosal tumor b) shows perineuralinfilteration of tumor

3. Discussion

The origin of ACC is not clearly known but is thought to be arising from ceruminous glands or sweat glands of EAC [13]. It is commonly seen in the 5th decade with slightly higher prevalence in females [14]. Early detection of ACC of EAC is often missed due to its presentation with nonspecific otologic symptoms like earache, otorhea, hearing loss and therefore being mistreated as otitis externa and chronic otitis media [13, 15]. Histologically 3 patterns are seen namely solid, tubular, cribriform. Immunohistochemistry demonstrates positivity for cytokeratin and S - 100 which indicates origin from ceruminousglands [13, 16].

Due to its low incidence rates there is some ambiguity regarding its management and with this report we aim to share our clinical experience and contribute towards it.

Once diagnosed surgical resection is the main treatment of choice with options being En bloc resection, Lateral resection includes excision of osseous and bony EAC, ossicles of middle ear. In subtotal resection inner ear is

included excluding petrous apex which is removed in total temporal resection [17]. In addition to this parotid gland, temperomandibular joint, duramatter and ITF can be included foe adequate margins. There are few recommendations which suggest lateral resection for T1 & T2 lesions and larger resections for T3, T4 disease [18]. Such extensive resection are done for clear margins as positive margins demonstrated poor prognosis as suggested by gardan et al [19].

In a study of 23 clinical cases by xiaojunjiang, they have recommended parotidectomy even in early stage of disease as it showed low recurrence rates [20]. Surgical management of neck is indicated when found positive for nodes or in recurrant cases. There are several studies which proved the role of adjuvant radiotherapy in advanced disease in attaining low recurrence rates [21, 22]. Role of chemotherapy is controversial and some studies show no improvement in survival [23].
Metastatic lesions are seen most commonly in lung followed by bone, kidney and brain [24].

4. Conclusion

Elderly patients with persistant refractory otologic symptoms should be suspected of malignancy as early detection is an important factor for survival. Adenoid cystic carcinoma once diagnosed, should be treated with surgical resection with adequate margins along with post op radiotherapy where necessary to prevent recurrances.

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