Rare Presentation Of Adenoidcystic Carcinoma Of External Auditory Canal With Subcutaneous Metastasis In Temporal Region

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Citation

Abstract
Adenoid cystic carcinoma of external auditory canal is a rare and specific variant of adenocarcinoma of the salivary gland. In this article we spot the case of a patient with adenoid cystic carcinoma of external auditory canal with subcutaneous metastasis in temporal region.

INTRODUCTION
Adenoid cystic carcinoma of external auditory canal is a rare and specific variant of adenocarcinoma of the salivary gland(1). Adenoid cystic carcinoma of the head and neck are usually found in the major salivary glands, as well as in the minor salivary glands such as the oral cavity, palate, nasal cavity, nasopharynx and the lacrimal glands. Adenoid cystic carcinoma of the external auditory canal arises from ceruminous glands, sweat glands or ectopic salivary gland tissue(1). Adenoid cystic carcinoma may result in distant metastasis or recurrences (2).

CASE REPORT
A 55 year old female presented to our ENT outpatient department with a 2 year history of external auditory canal stenosis with obliteration of meatus of canal with a smooth swelling arising from the ant canal wall. She also complained of moderate pain in the canal for the same duration of time. Along with biopsy of the tissue from the mass meatoplasty was done. Histopathological examination of the tissue revealed proliferating sheets of atypical cells which were monomorphic and basaloid in appearance. Cells were arranged in cribriform pattern. Some of the cells were also arranged in anastomosing cords and nests. These histopathological findings were consistent with adenoid cystic carcinoma of external auditory canal. (Fig 1)

She was sent for chemotherapy. She was given cisplatin 100mg/m² on day 1 and 5 FU 1000 mg / m² from day 1 to day 5. 5 cycles were given. After 1 year she presented with right temporal region swelling with stenosis of right external auditory canal (Fig2). Cytopathological findings were again consistent with adenoid cystic carcinoma in subcutaneous region of temporal region. Cytopathological examination revealed round to oval cells having scanty cytoplasm, high nuclear cytoplasmic ratio, uniform round to oval hyperchromatic nuclei and coarse nuclear chromatin. Hyaline spherical globule (arrow) is seen adherent to tumour cells. (Fig 3)
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Figure 2
Figure 2: Right temporal region swelling

Figure 3
Figure 3:H&E-400X

Figure 4
Figure 4: CT scan head showing metastasis in right temporal region which is extradural in nature.

CT head revealed metastasis to be extradural in nature (Fig 4). CT scan temporal bone axial view done after 2 months of CT scan head revealed thickening in the soft tissues of right ear external auditory canal. (Fig 5)

Excision of the swelling was done along with meatoplasty to maintain the patency of the canal (fig 6).

Figure 5
Figure 5: CT scan shows thickening in the soft tissue of the external auditory canal of right ear.
DISCUSSION

Malignant tumours of external auditory canal are rare and most are squamous cell carcinomas. Adenoid cystic carcinoma arising in the external auditory canal is exceedingly rare. Although adenoid cystic carcinoma is a rare tumour, it is relatively common in the salivary gland of the head and neck(3). Adenoid cystic carcinoma growth rate is slow and the nature of this carcinoma shows a slow malignant course(1). The main treatment is surgery. Post surgical recurrences and metastasis to the lungs, regional lymph nodes and bones occur over many years. Slow growth and subcutaneous metastasis in temporal region was observed in our case.

The true origin of adenoid cystic carcinoma in external auditory canal is controversial. It has been proposed that these tumours originate in ceruminous glands. Microscopic studies of these tumours and ceruminous glands demonstrate similar histological features. Some authors have suggested that these tumours arise from ectopic salivary glands of the external auditory canal although this opinion has not been proved(4). In general, ceruminous glands tumours are classified as ceruminous adenoma, pleomorphic adenoma, ceruminous adenocarcinoma and adenoid cystic carcinoma(3). Adenoid cystic carcinoma has three main histological patterns: tubular, cribriform and solid in the salivary glands, the prognosis of adenoid cystic carcinoma correlates with the predominant histological pattern. Tubular adenoid cystic carcinoma has the best prognosis where as solid adenoid cystic carcinoma has the worst prognosis(1). However in solid adenoid cystic carcinoma no significant correlation between these histological patterns and prognosis has been demonstrated. In this patient histopathology revealed cribriform pattern.

In our patient external auditory meatus stenosis with obliteration of meatus of canal was the main complaint. The chief presentation of adenoid cystic carcinoma of the external auditory canal is variable although ear pain and hemorrhagic or purulent discharge are the main complaints. Other complaints include facial paresis; tinnitus and hearing loss. Our patient had undergone meatoplasty meanwhile biopsy of the stenosis was also undertaken from the mass. Histopathological findings were consistent with adenoid cystic carcinoma of external auditory canal. Adenoid cystic carcinoma of the external auditory canal can be confidently diagnosed on the basis of cytology on which surgical management could be planned(5).

Adenoid cystic carcinoma may locally invade soft tissue and bone, extend into parotid gland and temporomandibular joint, invade by perineural and perivascular extension and metastasize too regional lymph nodes, lungs(6), liver, vertebrae(7). Contralateral metastasis to brain is also reported(4). This patient after chemotherapy returned with right temporal extradural region metastasis in subcutaneous tissue.

Treatment consists of radical excision of external auditory canal via a modified temporal bone resection(2). Radiotherapy and chemotherapy are not curative but can help in palliation and adjuvant therapy. In our patient excision of the extradural metastasis was done along with meatoplasty to maintain the patency of the canal.

CONCLUSION

Adenoid cystic carcinoma of external auditory canal is a rare tumour and should be considered among the most malignant tumours of external auditory canal. Along with regional lymph nodes, lungs, bones and liver head should also be evaluated for metastasis with CT. We like to emphasize the need for early detection of tumour along with necessary surgical management with long term follow up of these patients.

References

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