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Case Report

Adenoid cystic carcinoma at base of the tongue with rare cervical lymph node metastasis diagnosed on fine needle aspiration cytology

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ABSTRACT

Adenoid cystic carcinoma (ACC) is an uncommon malignant tumour of salivary glands. Occurrence at base of the tongue is rare. Distant metastasis is common, however, metastasis to regional lymph node is not seen commonly, but if occurs carries a bad prognostic value and lessens the average survival age of the patient. Fine needle aspiration cytology (FNAC) in such cases provides early diagnosis for rapid management of the patient. Review of literature shows that very occasional cases are reported with ACC at base of tongue with regional lymph node metastasis. Here, we present such a rare case of a 55 years old male patient with metastatic adenoid cystic carcinoma of cervical lymph node diagnosed on FNAC with primary at base of the tongue.

Keywords: Adenoid cystic carcinoma, FNAC, Base of tongue, Lymph node

INTRODUCTION

Adenoid cystic carcinoma (ACC) is an uncommon tumour that accounts for 1-2% of all malignant neoplasm of head and neck.[1] ACC mainly originates from minor and major salivary glands. It may also occur at trachea, chest, cervix and skin of face. Most common intraoral site is hard palate.[1, 2] Occurrence at base of the tongue is slightly uncommon.[1] The frequency reported in the tongue varies from 1% to 20.8%.[3] Patients usually present with slow growing painless mass. Metastatic spread to regional lymph nodes is rare but distant metastasis to lungs and bones is common.[2] Most of the cases are treated with surgical resection, however, postoperative radiotherapy is often recommended because ACC has infiltrative growth pattern and shows perineural invasion.[4] In this article, we present this rare tumour at rare site with rare metastasis diagnosed on fine needle aspiration cytology (FNAC), early diagnosis and treatment of which bears a prognostic value.

CASE REPORT

A 55 years old male patient presented in oncology department with dull pain in throat, difficulty in swallowing and hoarseness of voice. On clinical examination, a large mass at the base of the tongue was seen with enlargement of left cervical lymph node. Contrast enhanced computed tomography (CECT) neck revealed a large multicystic lesion measuring 5.5 × 5 cms at the base of the tongue more on the left side (Figure 1A). The mass was extending anterioly to anterior tongue, posteriorly to oropharynx, laterally to tonsillar fossa and inferiorly to geniohyoid-genioglossus muscles complex and to superior surface of hyoid bone (Figure 1B). Both neurovascular bundles were seen involved. Imaging suggested the presence of a malignant tumour arising from base of tongue with involvement of surrounding structures and left cervical lymph node.

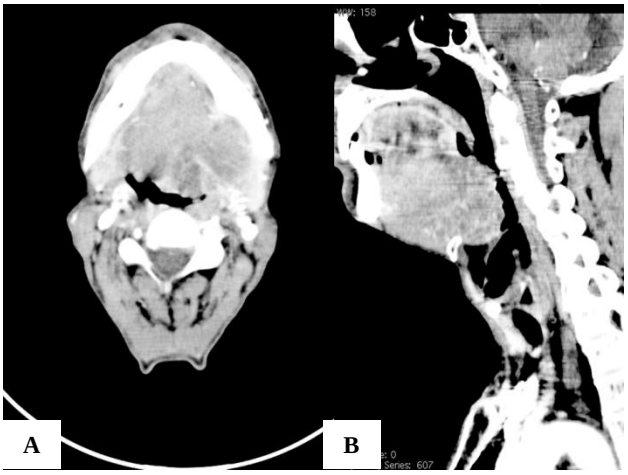

Figure 1 (A and B): CECT shows tumour arising from base of the tongue and invading the surrounding structures.

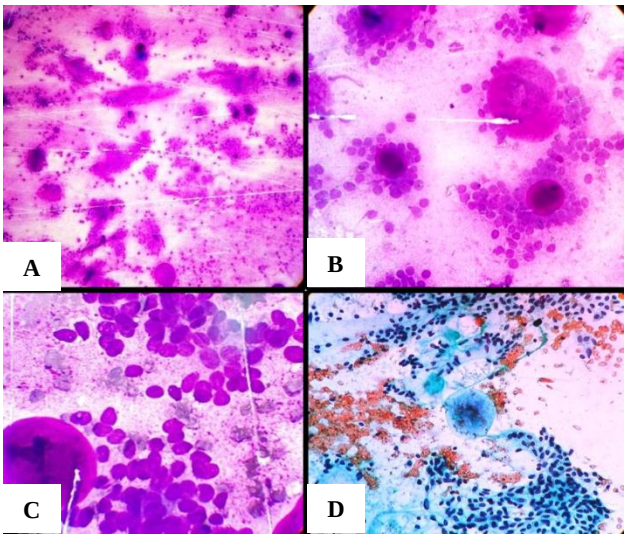

Figure 2: (A) Giemsa 10X, tumour shows high cellularity, hyaline globules/ matrix surrounding with tumour cells, (B) giemsa 20X, basaloid cells with angulated nuclei, inconspicuous nucleoli and scant cytoplasm, (C) giemsa 40X, single hyaline globule with sharp borders and surrounded by tumour cells and (D) pap stain 20X, tumour cells with lightly stained matrix.

As a next diagnostic modality, FNAC from left cervical lymph node was planned. FNAC was done with 21 gauze needle under aspectic conditions. Both wet fixed and air dried smears were prepared and stained with Papanicolaou (Pap) and May Grunwald Giemsa stains respectively. On light microscopy, the smears examined were cellular and showed a tumour composed of basaloid cells, large hyaline globules/ metachromatic matrix spheres and lightly stained matrix spheres that have tumour cells (Figure 2A). The basaloid cells were uniform having coarse chromatin, angulated nuclei, inconspicuous nucleoli and scant cytoplasm (Figure 2B). These tumour cells were arranged in cohesive clusters and occasionly cup shaped fragments. Large hyaline globules/ metachromatic matrix spheres showed sharply defined borders (Figure 2C). At many places, the lightly stained matrix spheres were surrounded by tumour cells (Figure 2D). Naked nuclei were present in the background.

Considering the characteristic cytological features on FNAC smears and the malignant nature of the tumour on CECT, the diagnosis suggested was metastatic adenoid cystic carcinoma, left cervical lymph node. The primary of the tumour was suggested at the base of the tongue after correlating with imaging findings. The patient was planned for tumour resection with lymph node excision following cytological diagnosis.

DISCUSSION

Adenoid cystic carcinoma is an uncommon tumour of oral cavity, mostly arising from minor and some from major salivary glands of head and neck. The other extraoral sites noted are trachea, lacrimal gland, breast, skin, and vulva.[1] Most common intraoral site for this tumour is hard palate. Base of the tongue is relatively uncommon site.[2] 60% - 90% of the patients carry MYB-NFIB or MYBL1-NFIB fusion gene.[5] The clinical features depend largely on the site of origin of the tumour. Lesions that arise early usually present as painless slow growing mass of mouth or face. Advanced staged tumors may lead to pain and/or nerve paralysis, as this neoplasm has infilterative nature and shows invasion of peripheral nerves.

Histopathologically, ACC is a biphasic tumour having epithelial and myoepithelial components and grows in tubular, cribriform, and/or solid pattern. According to some authors, this tumour is graded as grade 1 and 2 according to the presence of solid component.[6] But the present WHO classification does not specify any grading system.[7] However, solid component is associated with adverse outcome.[8] Necrosis and vascular invasion may not be seen commonly. To highlight epithelial component, immunohistochemical (IHC) markers like CK7 and CAM5.2 may be used while myoepithelail component may be highlighted by SMA and S100 IHC markers. MYB overexpression is a sensitive but nonspecific immunomarker for adenoid cystic carcinoma.[6]

FNAC presents an effective tool for early diagnosis of head and neck tumors. In case of ACC, characteristic features on FNAC smears that may help to arrive at diagnosis are biphasic nature of the tumour, metachromatic matrix spheres/ hyaline globules, lightly stained matrix and basaloid cells having angulated nuclei, scant cytoplasm that surround the spheres.[7] At this site, the differentials which may be considered are basaloid squamous cell carcinoma (bSQCC), polymorphous low-grade adenocarcinoma (PLGA), pleomorphic adenoma with cribriform pattern and basal cell adenoma. In our

case, as there was not a single keratinised cell helped to rule out bSQCC.[9] PLGA could be ruled out by cellular and nuclear features. Additionally, radiological correlation showing invasion into surrounding structures and perineural invasion helped to rule out other benign tumorous conditions.

ACC shows distant metastatis to lung, liver, brain and bones, however, spread to regional lymph node is rarely seen as was present in our case.[2] Conley and Dingman in their study has shown that spread to regional lymph node is associated with poor prognosis.[10] In a recent study by Opletak et al[11] patients with positive lymph nodes at the time of diagnosis lived on average 52 months less than those with negative nodes. Also, in patients with positive nodes, the disease recurred on an average of 36 months earlier than those with negative node disease.[11] The other poor prognostic factors include female sex, older age, sinonasal location, lymphovascular invasion, high grade transformation, intraneural invasion, high staging, positive margin status and high mitotic index.[7] Surgical resection is the main mode of therapy. However, in some centres postoperative radiotherapy is preferred that helps in limiting the local failure.[1, 2] Recurrence for ACC following surgery is common due to its extensive tissue infiltrative nature and perineural invasion. Amit et al in their study supported the elective neck treatment in the patients of adenoid cystic carcinoma, with or without nodal metastasis due to more 5 years survival rate.[12]

CONCLUSION

Adenoid cystic carcinoma at base of the tongue is an uncommon presentation and should be considered with other common differentials at this site. Radiological correlation in addition to cytological features may help to reach this diagnosis early and to rule out other tumors. Metastatic spread to regional lymph node is also rare that carries a poor prognostic value and affects the median survival age of patients.

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