INTRODUCTION

The adenoid cystic carcinoma (ACC) is a relatively rare epithelial tumor of the salivary glands. It accounts for about 5-10% of all salivary gland neoplasms, representing 2-4% of malignant occurrences of the head and neck area. Approximately, 31% of lesions affect minor salivary glands, particularly the palate, although they can also be observed in the sub-mandibular and parotid glands.[1] The frequency reported in the tongue is 19.8%, with 85% observed at the base of the tongue.[2] There had been only two cases reported on the dorsum of the tongue.[3] We report one such rare case of tongue neoplasm which turned out to be ACC in a middle aged lady.

CASE REPORT

A 45-year-old-female patient presented with an asymptomatic growth of the tongue, which was perceived just 2 weeks before consultation. The patient reported no significant medical history or any addiction. Laboratory blood studies were requested, which rendered normal results. Chest X-ray was also within normal limits. The intra-oral examination at that time revealed a mass in the dorsum of the tongue with light pain to pressure, without any evidence of cervical lymphadenopathy. The mass was firm, same color as that of the surrounding mucosa and asymptomatic otherwise [Figure 1]. As a pre-operative assessment of the lesion, a fine needle aspiration was done and the smear revealed a salivary neoplasm consisting of well delineated, tightly cohesive clusters of basaloid cells surrounding mucoid, hyaline globules, or clear spaces also forming honeycomb (cribriform) pattern [Figure 2]. At places dense aggregates of monomorphic small cells with uniform round to oval hyperchromatic nuclei and scanty cytoplasm were seen. Smears also showed individual tumor cells with high N: C ratio and nuclear moulding. Fine needle aspiration FNA findings were suggestive of ACC. Subsequently, the patient underwent surgery and the specimen was sent for histopathologic study. Macroscopically, the mass had firm consistency with an irregular form and surface, brown color and measured 2.5 × 1.5 × 1.0 cm. The histopathologic study revealed multiple pseudocystic spaces of variable sizes surrounded by cuboidal cells with scarce cytoplasm and oval nuclei, filled with eosinophilic material and hence was consistent with the diagnosis of ACC [Figure 3]. However, there was no evidence of perineural infiltration on serial sections.

DISCUSSION

Minor salivary gland neoplasms occur less commonly than the major salivary gland tumors and tongue is a relatively uncommon site for salivary gland neoplasms. However, those originating from the minor salivary glands are mostly malignant tumors. ACC is a malignant neoplasm that originates in both the minor and major salivary glands, characterized by slow growth, diffuse invasion, and potential to produce distant metastases, mainly to the lungs and bones.[4] It is an infrequent lesion, as it represents approximately 1-2% of all malignant neoplasms of the head and neck, and up to 10-15% of all malignant salivary gland neoplasms.[5] The most common intra-oral site for minor salivary gland tumors is the hard palate, followed by the base of the tongue[6] where up to 96% of all tumors are malignant, and ACC represents 30% of...
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them.[7] On the other hand, one of the least frequent sites of presentation for ACC is the mobile tongue, as several authors have reported an incidence of only approximately 3% of the cases.[8]

De Vries et al., analyzed 178 cases of salivary gland tumors, out of which only six cases were located on the tongue.[9]

Cytologically, cribriform variety of ACC can be diagnosed by hypercellular smears composed of clusters of small, relatively monomorphic epithelial cells with hyperchromatic nuclei. These cells are arranged around pseudocysts filled with globules of mucoid substances. These appear bright magenta in May-Grunwald Giemsa MGG stains and pale blue with Papanicolaou stain. Finger-like process of similar material can also be found in between the groups of cells in tubular variety. The solid variant of ACC also exhibits the same material and the cells resemble that of small cells of anaplastic carcinoma.

The globules of amorphous material surrounded by the monomorphic hyperchromatic cells was a clue to the diagnosis of ACC in our case, but since the hyaline globules are also found in other tumors like basal cell adenoma, pleomorphic adenoma, polymorphous low grade adenocarcinoma, epithelial myoepithelial carcinoma etc., they were considered as possibilities. It is important to distinguish the adenomas from ACC because of the conservative mode of management in case of adenomas. We ruled out the adenomas because of the nature of the globules and the cytological morphology. Unlike the adenomas, the hyaline globules were dense and stained intensely with MGG and the cells were relatively monomorphic, hyperchromatic with coarse chromatin and irregular nuclear membrane like that of ACC. Thus a provisional diagnosis of ACC was made.

Histopathological confirmation was carried out before institution of radical therapy. Of three histologic variants - tubular, cribriform and solid; in our case cribriform pattern was the dominant one without any evidence of perineural infiltration.

The main factors associated with patient survival were tumor location, clinical stage, and the observed histologic variable.

Conversely, Spiro et al.,[7] have not found histologic classification to be of any benefit, and deny a correlation between microscopic appearance and prognosis.

Due to the slow growth pattern of the tumor, there is much controversy regarding the treatment of ACC. However, due to local recurrence and late metastasis, surgery remains the mainstay of management with or without radiotherapy.

Early detection is recommended for ensuring a favorable prognosis and good quality of life. Fine needle aspiration can be a useful modality of investigation in this regard.
REFERENCES

1. Triantafillidou K, Dimitrakopoulos J, Iordanidis F, Koufogiannis D. Management of adenoid cystic carcinoma of minor salivary glands. J Oral Maxillofac Surg 2006;64:1114-20.

2. Spiro RH, Huvos AG. Stage means more than grade in adenoid cystic carcinoma. Am J Surg 1992;164:623-8.

3. Ishikawa Y, Ishii T, Asuwa N, Ogawa T. Adenoid cystic carcinoma originated from an anterior lingual minor salivary gland: Immunohistochemical and ultrastructural studies and review of the literature. J Oral Maxillofac Surg 1997;55:1460-9.

4. Umeda M, Komatsubara H, Nishimatsu N, Oku N, Shibuya Y, Yokoo S, et al. Establishment and characterization of a human adenoid cystic carcinoma line of the salivary gland which is serially transplantable and spontaneously metastasises to the lung in nude mice. Oral Oncol 2002;38:30-4.

5. Kim KH, Sung MW, Chung PS, Rhee CS, Park CI, Kim WH. Adenoid cystic carcinoma of the head and neck. Arch Otolaryngol Head Neck Surg 1994;120:721-6.

6. Khafif A, Anavi Y, Haviv J, Fienmesser R, Calderon S, Marshak G. Adenoid cystic carcinoma of the salivary glands: A 20-year review with long-term follow-up. Ear Nose Throat J 2005;84:662-7.

7. Spiro RH, Huvos AG, Strong EW. Adenoid cystic carcinoma of salivary origin. A clinicopathologic study of 242 cases. Am J Surg 1974;128:512-20.

8. Andersen LJ, Therkildsen MH, Ockelmann HH, Bentzen JD, Schiødt T, Hansen HS. Malignant epithelial tumors in the minor salivary glands, the submandibular gland, and the sublingual gland. Prognostic factors and treatment results. Cancer 1991;68:2431-7.

9. de Vries EJ, Johnson JT, Myers EN, Barnes EL Jr, Mandell-Brown M. Base of tongue salivary gland tumors. Head Neck Surg 1987;9:329-31.

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