

Case Report

Polymorphous adenocarcinoma on the base of tongue: a rare case

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Abstract:

Polymorphous adenocarcinoma (PAC) is a distinct neoplasm of the minor salivary gland. Most patients present with an asymptomatic mass in the hard palate.” Rarely, “the mass can also occur in the” base of the tongue. A 56-year-old female patient, presented to ENT clinic, Teaching Hospital Jaffna with a history of voice change for 4 months. Her physical examination revealed fissured tongue, a mass of 1×1.5 cm in size with intact mucosa located at the left side of the base of tongue, which was crossing the midline and infiltrated into the deep tongue muscles. Deep biopsy revealed polymorphous adenocarcinoma. She was treated with radiotherapy. PAC is ‘unusual’ to arise at the base of the tongue. It is important to diagnose and treat it accordingly.

Keywords: Polymorphous adenocarcinoma, base of the tongue

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Introduction:

Polymorphous adenocarcinoma (PAC) is a rare malignant neoplasm with a low level of aggressive biological¹ behaviour when compared with other types of adenocarcinoma. About 75% of these tumours arise from minor salivary glands (MiSGs). It most frequently arises on the palate². In this article, we describe a case of PAC that arose in an unusual location-the base of tongue.

Case Presentation:

A 56-year-old female patient, presented to the ENT clinic of Teaching Hospital Jaffna (THJ) with a history of voice change for 4 months. She had no complaints of symptoms such as shortness of breath, odynophagia or dysphagia. The physical examination revealed a fissured tongue, a mass of 1×1.5 cm in size located at the left side of the base of tongue which was crossing the midline and infiltrating the deep tongue muscle but, the overlying mucosa was intact. (Fig. 1). No lymphadenopathy was detected on neck examination, and the patient was otherwise normal. Deep biopsy was taken from the mass under general anaesthesia. Histology revealed as polymorphous adenocarcinoma. After multidisciplinary team discussion (MDT), patient was referred to the oncology unit for further management.

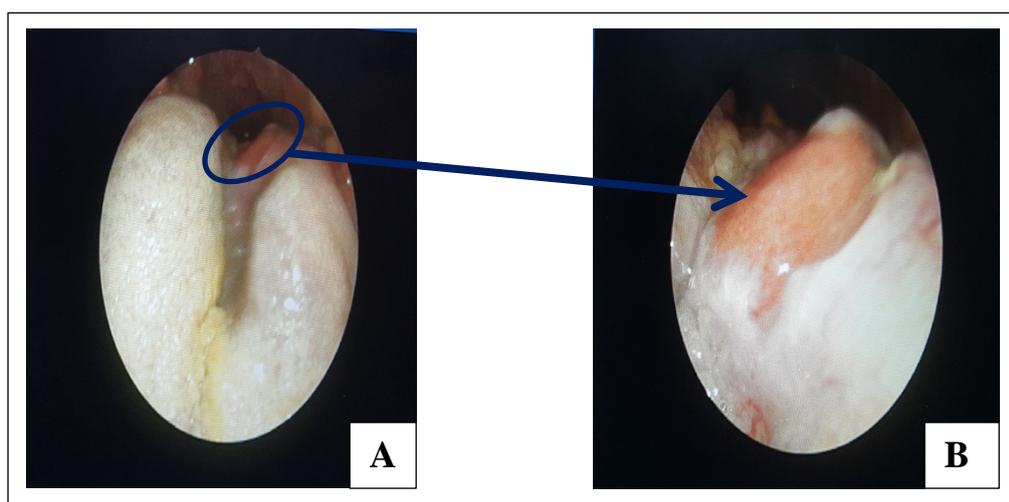


Figure 1 : A- fissure tongue with tongue base tumour, B- magnified view of tumour

Discussion:

Minor salivary glands (MiSG) can present along the whole upper aero digestive tract. Therefore, the signs and symptoms of MiSG tumours depend upon the anatomical site where the tumour is located. The majority of patients diagnosed are in the 5th to 6th decade. The most frequent histology's of MiSG tumours are Adenoid cystic carcinoma and Mucoepidermoid carcinoma (up to 60%), followed by Adenocarcinoma not otherwise specified².

Evans and Batsakis first defined polymorphous adenocarcinoma (PAC) arising in the minor salivary glands as a malignant tumour in 1984. Previously it was reported as terminal duct adeno carcinoma or lobular carcinoma^{2,3}. The palate is the most common site of PAC of the minor salivary glands followed by the buccal mucosa, lip, retromolar triangle and the cheek. Very rarely does the tumour occur in the tongue¹. Involvement of major salivary gland is relatively unusual². MiSG tumour classically presents as a painless submucosal swelling, which is fixed to the overlying mucosa.

There may be a small central area of ulceration.² Our patient presented with voice change. She had fissured tongue and base of tongue tumour with intact mucosa.

There are four histological patterns described: cribriform, tubular, lobular and papillary². Due to this morphologic pleomorphism, PAC has often been misdiagnosed as pleomorphic adenoma or adenoid cystic carcinoma¹ PAC differs from pleomorphic adenoma because it is characterized by infiltrative margins and an absence of chondromyxoid stroma¹. Differential diagnosis of adenoid cystic carcinoma is more challenging³ Immunohistochemical and molecular biological studies may help to differentiate these two lesions².

If feasible, wide local excision with tumour free margins is the treatment of choice for MiSG tumour. Surgical approach can vary. For smaller lesions, transoral robotic resections or transoral laser microsurgery and for larger tumours, suprahyoid release or mandibulotomy and free-flap reconstructions may be needed Pre-operatively, resectability of the tumour is determined by, anatomical site of origin, clinical and imaging finding, the histology and the availability of surgical expertise. These factors also determine the extent of necessary surgical resection and the functional implications.

Patients, who are inoperable or have an unresectable tumour, or who refuse surgery are treated with primary radiotherapy. Now newer techniques such as IMRT promise better results, where loco regional control seems to improve with limited toxicity. Neutron radiotherapy extends its local control up to 75% (5-years), to patients with unresectable disease. But this option is still unattractive due to its lacking survival benefit and severe delayed side effects, including sensorineural hearing loss, cervical myelopathy and necrosis of the soft tissues, the temporal and mandibular bone and the temporal lobe. In recent times, even better long-term local control is reported after giving a boost using the gamma knife at the end of neutron radiotherapy. For unresectable disease, good tumour control is reported on the “use of concurrent chemo radiation”, avoiding morbidity of a resection

When we consider our patient, her tumour was mainly found in left side but it crossed the midline and also infiltrated into the deep muscles of the base of the tongue. The ideal surgical option would be total glossectomy. But the inevitable functional deficits such as the impact on speech, deglutition and quality of life were the prime concern to decide against such a decision. As a result, primary radiotherapy was selected as the treatment option for our patient through MDT.

Conclusion:

PAC is an unusual tumour to arise at the base of the tongue. Therefore, the possibility of PAC needs to be considered in the differential diagnosis when dealing with base of tongue lesions.

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