CASE REPORT

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A rare case of pleomorphic adenoma of the tongue

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ABSTRACT

Pleomorphic adenoma of minor salivary glands accounts for less than 3% of all head and neck tumors. It is a benign tumor. Palate is the most common site followed by upper lips. Tongue is an unusual site, and there are only 29 reported cases in literature as per our knowledge. We present a case of a 38-year-old female patient who presented with mass in the anterior tongue that was diagnosed as pleomorphic adenoma and was treated by surgical excision.

Key words: Benign tumor, minor salivary gland, oral cavity, palate, tongue

INTRODUCTION

Pleomorphic adenoma is a benign neoplasm affecting the salivary glands. They are mainly seen in major salivary glands, especially in the parotid glands. Minor salivary glands are more commonly affected by malignant neoplasms than benign ones. Minor salivary glands can be found in the oral cavity, larynx, and trachea, and all these are potential sites for pleomorphic adenoma. Our case is rare of its type as it was present in an unusual site, i.e., the tongue.

CASE REPORT

A 38-year-old female patient presented to our outpatient department with a history of swelling in the tongue for the past 6 months [Figure 1]. It was slow growing, painless, not associated with difficulty in swallowing, or movement of the tongue. The patient does not report any incidence of trauma to the area and is not a known diabetic or hypertensive. There was nothing suggestive in the personal or family history. After getting informed consent, the patient was examined and, there was a 2 cm by 2 cm by 1 cm swelling in the ventral aspect of the tongue, mainly on the right side that was firm, non-tender, with smooth surface and no restriction of the tongue mobility. Mucosa over the swelling was freely mobile. Enlarged blood vessels were present on the surface. No other abnormality was detected in the oral cavity. No cervical lymphadenopathy was seen. Rest of ENT examination was within normal limits. Fine-needle aspiration cytology from the swelling came out as pleomorphic adenoma of minor salivary gland. Magnetic resonance imaging (MRI) showed homogenous mass of size 2.5 cm × 2 cm × 1 cm in the anterior aspect of the tongue, in the right side, not crossing midline. The muscles of tongue were not seen to be infiltrated and features were suggestive of benign lesion. After routine investigations, under general anesthesia, incision was given in the right lateral border of the tongue, and the mass was dissected off the tongue tissue [Figures 2 and 3]. The defect was primarily repaired. Post-operative period was uneventful. The histopathology report was confirmed to be pleomorphic adenoma of minor salivary gland [Figure 4]. The patient has been on follow-up for 4 months and there is no evidence of recurrence.

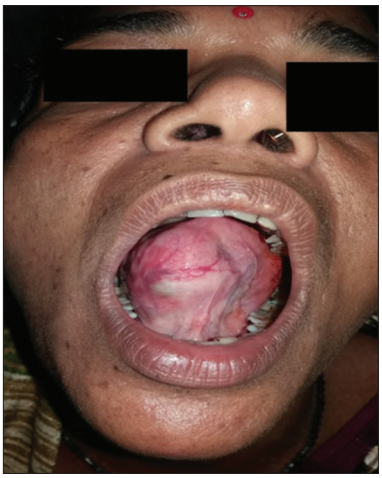

Figure 1: Swelling in the anterior aspect of the tongue

DISCUSSION

Pleomorphic adenomas constitute 3–10% of all neoplasms in the head and neck region.[1] They are more common in parotid and submandibular glands. Minor salivary gland pleomorphic adenomas are rare entities. The most common sites of these in the oral cavity are palate and lips. Unusual sites include cheek, retromolar trigone, and floor of the mouth.[2,3] The tongue is a rare site. They have female preponderance and usually occur in 40–50 years age group.[4,5] Of 143 intraoral pleomorphic adenomas studied by Eveson and Cawson, only 1 case involved tongue. As per article published by Tanigaki *et al.*, there are only 26 cases reported in the world since 1960 as pleomorphic adenoma in the tongue.[6] 3 more cases have been reported after that.

The etiology is not known clearly. There are evidences to suggest that exposure to radiation and viruses (simian virus) have a role to

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Figure 2: Peroperative picture

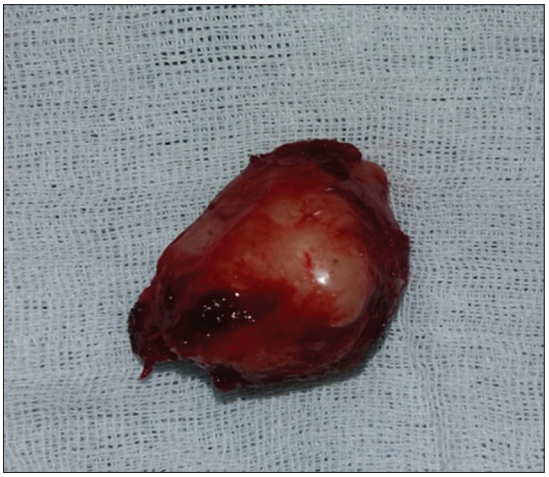

Figure 3: Resected specimen

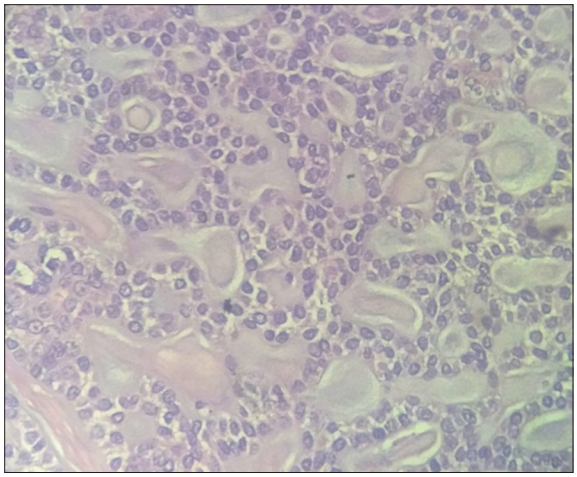

Figure 4: Histopathological picture

play. Abnormalities in the chromosome loci 8q12 and 12q15 are also implicated. These tumors usually present as smooth masses that are slow growing and painless.[7] The symptoms depend on site and size - may be asymptomatic, or can cause dysphagia if large and located posteriorly in the oral cavity, respiratory difficulty, etc.

Histologically, pleomorphic adenomas are mixed tumors, i.e., they have both epithelial and mesenchymal elements. Their proportion can vary, depending on which the consistency of the tumor also changes.[8] There are variants such as pleomorphic adenoma with lipomatous change, myxolipomatous pleomorphic adenoma, and pleomorphic adenoma with squamous differentiation.[9] Studies show that there is the presence of S-100 protein and glial fibrillary active protein-related antigen in these tumors and may be beneficial in diagnosis.[10] MRI is the investigation of choice when the extend of tumour is to be known in the tongue, because of better soft tissue delineation. However, in palate, computed tomography is more helpful as it shows bone erosion better. Biopsy is avoided for fear of seeding. However, fine-needle aspiration is safe and is the investigation that is most commonly done.

The definitive treatment of pleomorphic adenoma is surgery. In oral cavity, it can be removed with a safe margin. Care should be taken that no tissue is left out as recurrence can occur. Spiro *et al*. in his study stated that recurrence rate of pleomorphic adenoma in minor salivary glands was around 6%.[11] Tumors in the anterior tongue, palate, and others in oral cavity can be removed transorally. Those of base of tongue and soft palate may require combined transoral-transcervical, transpharyngeal, or transmandibular approaches according to the size and extension. Lasers and coblators are also being used. Radiotherapy may be indicated postoperatively when the lesion was in a difficult area or if the surgeon doubts that he has left back some tissue, like in parapharyngeal space. However, this is usually not required in majority of cases. Wherever the lesion and whatever the approach and technique used, a pleomorphic adenoma of the oral cavity should always be removed.

CONCLUSION

Pleomorphic adenoma of the tongue is a rare neoplasm. However, once diagnosed, it can be easily excised. This entity should always be kept in mind as a differential diagnosis in cases of tongue swelling so that timely treatment can be provided.

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