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Case Report

Pleomorphic adenoma of the minor salivary gland in buccal mucosa: A case report

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ABSTRACT

Pleomorphic Adenoma (PA) is the most common tumor of the salivary gland, arising mostly from the parotid gland while small percentage from the minor salivary gland. The most common sites of pleomorphic adenoma of the minor salivary glands are the palate, followed by lips. The buccal mucosa or cheek is a rarely affected site by PA of the minor salivary glands. Here, we present a case report of PA of the buccal mucosa, presented with intraoral swelling, complete surgical excision was done with a follow up of 3.5-years.

1. Introduction

Salivary gland neoplasm accounts for 3–5% of all head and neck tumors.[1] Minor salivary gland tumor accounts for <25% of the total salivary neoplasms.[2] PA is the most common tumor of the major and minor salivary gland, 85% occur in the parotid gland and 7% in the minor salivary glands.[3] It is a benign mixed type of tumor as it has its origin from both epithelial and myoepithelial components. Among minor salivary gland, most common site of occurrence is palate (60-65%) and buccal mucosa is a rare site of occurrence (5.5%).[2,4] The present case report describes a rare case of PA arising in the right buccal minor salivary gland, excised under local anaesthesia with wide margins and followed for 3.5 years without reoccurrence.

2. Case Report

A 21-year-old female patient reported with the chief complaint of swelling over right side of face from last 2-3 years. The swelling was initially small in size and gradually increased to the present size of 3 cm × 3 cm. Patient had no significant medical, dental, family or surgery history. On general examination, the patient was apparently healthy. There was no regional lymphadenopathy and mouth opening was normal. On extraoral examination, facial asymmetry with obliteration of nasolabial fold due to swelling on the right side of face was noted. A solitary oval shaped swelling with normal overlying skin and smooth surface was present on right cheek region. Swelling was seen extending superio-inferiorly from ala-tragus line to 2 cm above the lower border of mandible. Antero-posteriorly it was seen extending 1 cm from the right corner of the mouth to the imaginary line running from the outer canthus of eye to the lower border of mandible angle (Figures 1 and 2). Intraorally, a well-defined oval shaped swelling was present on the right buccal mucosa extending anterio-posteriorly from the right retrocommissural area to second molar region and superio-inferiorly 1.5 cm above and below the occlusal plane. Swelling was having normal overlying mucosa with smooth surface. On palpation all inspectory findings were confirmed. Extraorally, swelling was afebrile with normal overlying skin. Intraorally, swelling was non-tender, mobile and firm in consistency, nonfluctuant, nonreducible, nonpulsatile with smooth surface (Figure 3). Based on history and clinical examination provisional diagnosis of cysticercosis was made with differential

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diagnosis of benign minor salivary gland tumour, dermoid cyst, lipoma, sebaceous cyst, fibrosed mucocele, benign connective tissue tumour, neurofibroma.[4] All the necessary blood investigations were carried out which came out to be within normal limit. Ultrasonography, revealed well-defined lobulated hypoechogenic lesion measuring 22 x 17mm in the right cheek with internal and peripheral vascularity with multiple foci of calcification. FNAC revealed spindle cell embedded in chondromyxoid stroma with hemosiderin laden cyst macrophages. Since the lesion was small in size, excision of the lesion was carried out (Figure 4). Excisional biopsy specimen revealed a well encapsulated tumor mass which exhibit spindly to cuboidal tumor cells arranged in nests, cords and sheets. Abundant areas of myxomatous material and keratin pearl formation within the tumor cells. Plasmacytoid like cells seen around the myxomatous material. Hyperchromatic tumor cells have spread out in the entire stroma and have obliterated its architecture making it more cellular and myxomatous at places (Figure 5). This confirmed the diagnosis of PA of salivary gland. Based on its location and not associated with the parotid duct or gland, it was considered to be of buccal minor salivary gland origin. The patient is under periodic review, and there is no evidence of recurrence after three and half years of follow-up (Figure 6).

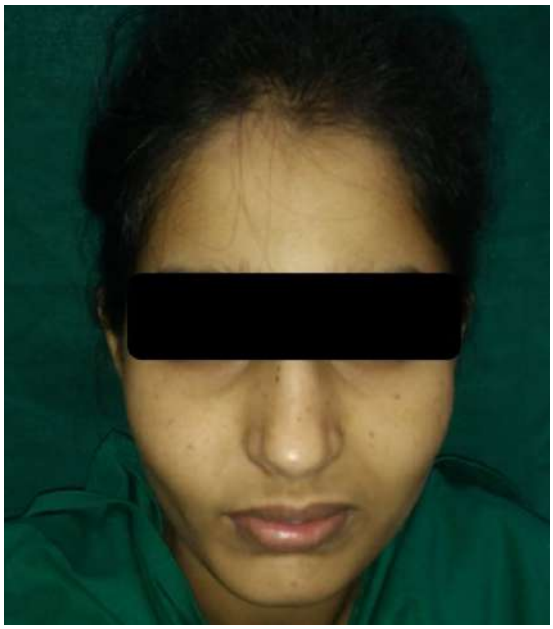

Fig. 1: Extraoral view of the patient with swelling on the right side of the face

3. Discussion

Salivary gland tumors accounts less than 5% of all head and neck tumors.[3] PA is the most common salivary gland tumor affecting both major and minor salivary glands.[4]

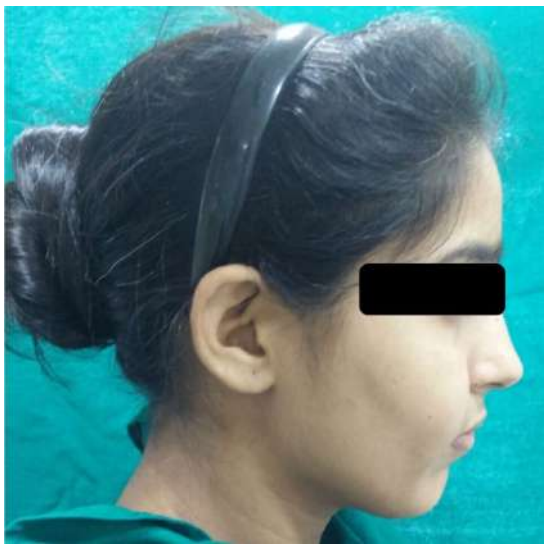

Fig. 2: Extraoral view of the patient with swelling on the right side of the face

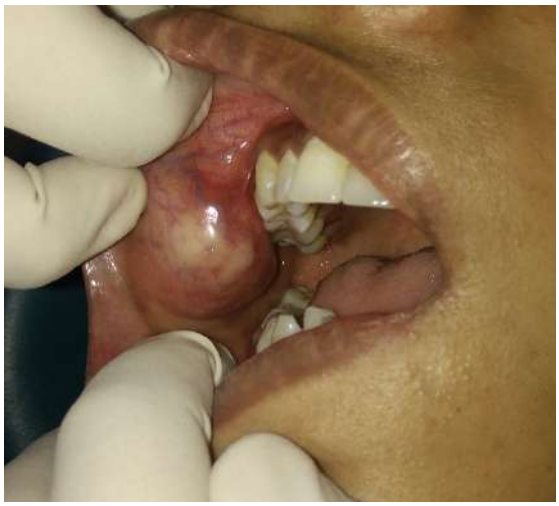

Fig. 3: Intraoral view of the patient with swelling on the right buccal mucosa

Most commonly involved gland is parotid (85%), followed by submandibular gland (8%), and intraoral (7%) salivary glands.[3] Among the minor salivary glands, palatal glands are the most commonly affected (43% to 70%) followed by the upper lip (10.1%) and cheek (5.5%) while rarely involve the throat (2.5%), retromolar region (0.7%).[2,3,5] The term PA was proposed by Willis because of its pleomorphic histology consisting of the epithelial and mesenchymal cells and intercellular matrix composed of fibrous, myxoid, mucoid, hyalinised, chondroid or osseous tissues. WHO in 1972 defined PA as "a circumscribed tumour characterized by its pleomorphic or mixed appearance clearly recognizable epithelial tissue being intermingled with tissue of mucoid, myxoid or chondroid appearance".[6]

Fig. 4: Excised tissue

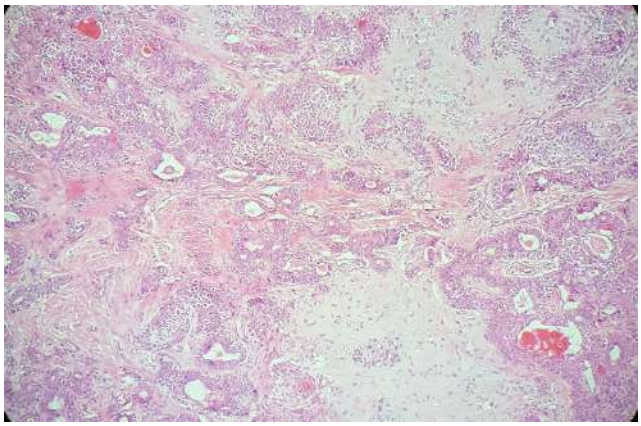

Fig. 5: Histopathological view of the tumor

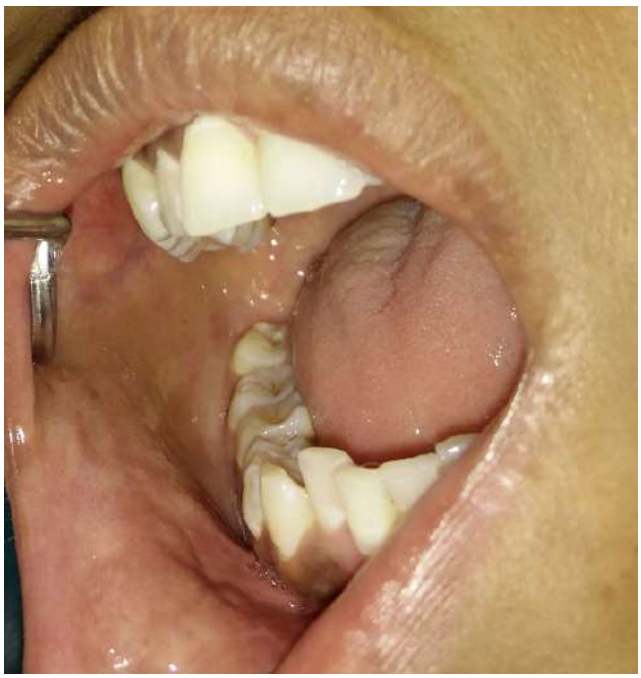

Fig. 6: Post operative intraoral view of the patient

PA of minor salivary glands usually occurs in the fourth to sixth decades of life with a slight female predilection, most commonly affecting the facial region.[2,3,7] In the present case also female was affected which was in accordance to the previous literature but patient was quiet young in age which was not in concordance of the previous studies. Etiopathogenesis of PA is not yet clear, however, described to have an association with radiation and chemical exposure, viral infection, use of tobacco and genetic predisposition with chromosomal aberration of 8q12 and 12q15.[3,7,8] The PA of minor salivary glands usually present as solitary, well defined, painless, firm, nodular submucosal masses showing intermittent growth with rare ulceration of the overlying skin or mucosa.[1] PA range from 1-7 cm in diameter or even larger.[1,9] The noticeable facial deformity prompts the patient to seek treatment. In our case patient reported with well defined, painless, firm mass which grew in considerate size and caused the facial deformity.

Fine needle aspiration cytology is the preferred diagnostic modality to see the basic nature of the lesion.[9] Ultrasonography, CT scan and MRI determines the size and extent of lesions and its effect on the surrounding tissues.

The differential diagnosis of minor buccal salivary gland pleomorphic adenoma includes buccal space infection, hemangioma, dermoid cyst, lipoma, sebaceous cyst, fibrosed mucocele, benign connective tissue tumor, neurofibroma, mucoepidermoid carcinoma. Buccal space abscess was ruled out due to the absence of signs of inflammation. Lack of bluish discoloration and negative diascopy test rules out hemangioma. Dermoid cyst was ruled out because of solid nature of PA and lack of tissue showing three germ layers. Sebaceous cyst shows punctum and fixed mass. Histologically, mucoepidermoid carcinoma was ruled out as both epithelial and myoepithelial cells were seen. Negative slip test and absence of fluctuance and histologically lack of lipomatous component rules out lipoma.[2,3,9]

Histologically, PA have epithelial cells arranged in cord or duct-like cell patterns, and mesenchymal cells with intercellular matrix showing fibrous, hyaline, myxoid, cartilaginous, and osseous areas.[9] Myoepithelial cells have pleomorphic morphology with spindle or round shaped cells and are responsible for such pleomorphic extracellular matrix production. Foote and Frazell categorized tumor into the principally myxoid, myxoid and cellular components in equal proportions, predominantly cellular and extremely cellular.[1]

PA has recurrence of 6% in benign minor salivary gland tumors.[2] PA is known to produce recurrence either due to inadequate surgical removal or due to the spillage of tumour cells while excision. PA have a capsule either thin or incomplete with microscopic pseudopod-like extensions into the surrounding tissue.[6,8] Therefore, treatment of choice for PA is wide local excision with safety margins. PA

has malignant potential, accounts for about 3% of salivary tumor, hence regular follow-up for at least 3–4 years has been recommended.[3,10]

4. Conclusion

PA of buccal mucosa is a rare neoplasm and presents as a challenge to even the most experienced maxillofacial surgeon, physician or pathologist. It should also be considered in the differential diagnosis of slow-growing, painless cheek swellings. Early diagnosis and proper surgical excision with wide margins will give excellent prognosis but it requires a long term follow-up to rule out recurrence or malignant transformation.

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6. Conflict of Interest

The authors declare that there is no conflict of interest.

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All authors contributed equally in the study. They have read and approved the content of the manuscript.

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