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## CASE REPORT

### PLEOMORPHIC ADENOMA OF THE PALATE: A CASE REPORT

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#### ABSTRACT

Pleomorphic adenoma is a benign tumor which commonly affects major salivary glands and rarely affects minor salivary glands. PA accounts for 53-77% of parotid tumors, 44-68% of submandibular gland tumors, and 33-43% of minor salivary gland tumors. We are presenting a case of pleomorphic adenoma of hard palate in a 45 years old male patient with complaint of pain less swelling in the mid-palatal region with a multinodular appearance of more than 15 years of duration. Despite the long standing history of the lesion there was no involvement of the bone, no lymphadenopathy, no malignant transformation though always has to be taken care of as malignancy is seen more commonly in minor salivary glands and the lesion was managed with wide surgical excision.

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#### INTRODUCTION

In head and neck tumors, salivary gland tumors account for less than 4%. Pleomorphic adenoma (PA) is the most common benign and Mucoepidermoid carcinoma is the most common malignant tumor of salivary gland (Gothwal A K *et al*, 2012). PA has been known with different names such as mixed tumor, enclavoma, branchioma and enchondroma. It is called as a mixed tumor of salivary gland origin because of its dual origin from the epithelium and myoepithelial cells. The tumor was named by Willis characterizing the unusual histologic pattern of the lesion (Rajendran S and Sivapathasundaram S, 2009). In 1972 WHO defined PA as "a circumscribed tumor characterized by its pleomorphic or mixed appearance clearly recognizable epithelial tissue being intermingled with tissue of mucoid, myxoid or chondroid appearance" (Verma P *et al*. 2014).

#### Case Report

A 46 year old male patient reported to the department of Oral Medicine & radiology with the chief complaint of a painless swelling on the palate since last 15 yrs. History revealed that initially the swelling was peanut sized which had gradually increased to the present size. There was no history of trauma to maxillofacial region. No evidence of pain, paresthesia and

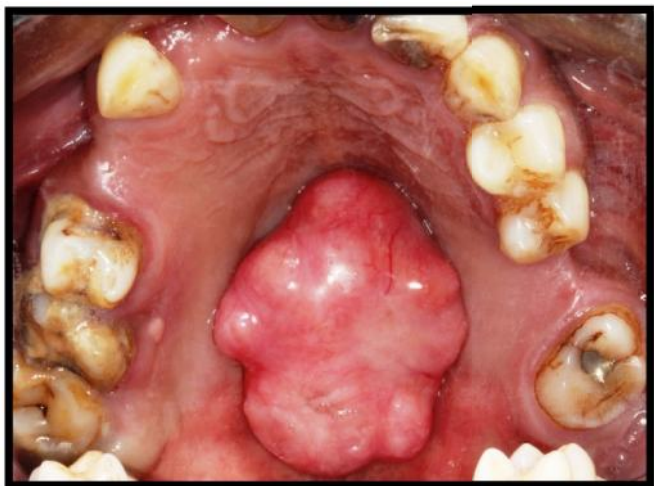
altered salivation. The past dental, medical and family history was noncontributory. Personal history revealed that patient was a chronic tobacco chewer. General physical examination was non-significant. There was no cervical lymphadenopathy.

Hard tissue examination revealed that there was advanced proximal caries with 11,21,27, moderate proximal caries with 12,34,35 and missing teeth with 16,24,36,37,42,46. Soft tissue examination revealed that there was Single, well-defined, sessile swelling with multinodular appearance in mid palatine region extending from anterior hard palate to the junction of posterior hard palate and soft palate, measuring 3 X 2 cm in size (Figure 1).

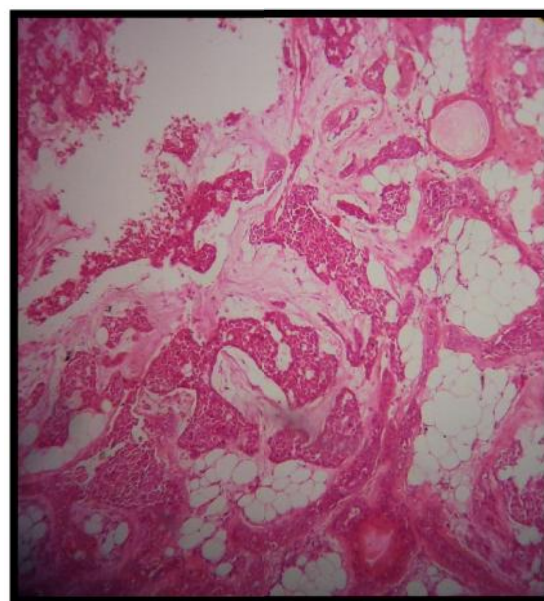
There was no change in the color of overlying mucosa and it was non-ulcerated. The lesion was firm in consistency, non-fluctuant, non-reducible, non-pulsatile and nontender on palpation. Based upon the history and clinical examination, the provisional diagnosis of benign minor salivary gland tumor was made and the differential diagnosis of torus palatine, fibroma, Neurofibroma, Fibrolipoma, Mucoepidermoid carcinoma, odontogenic and non odontogenic cyst were considered.

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**Figure 1** Intra-oral photograph showing swelling involving mid palatine region, extending from anterior hard palate to the junction of posterior hard palate and soft palate.



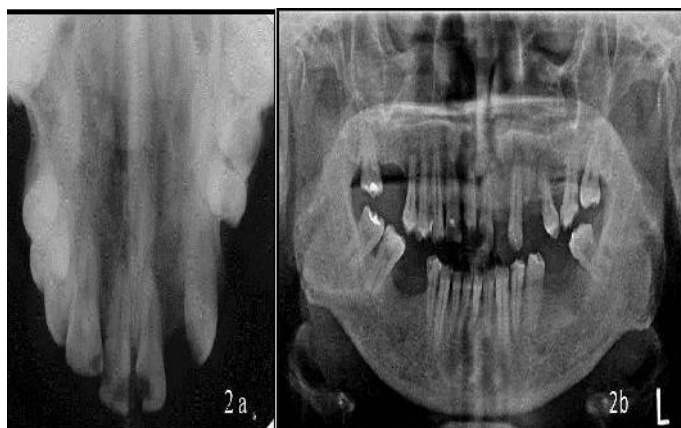
**Figure 3** Hematoxylin and eosin stained section (20X) demonstrating cuboidal, columnar, plasmacytoid cells are arranged in tubular, ductal pattern and solid sheets. Mucoid, myxomatous areas and keratin pearls are seen.

Various investigations were advised for the patient including hematological investigations (CBC, HIV, and HbsAg) which did not reveal any abnormalities. Radiographic investigations including maxillary occlusal radiograph (Figure 2a) and panoramic radiograph (Figure 2b) revealed that there was no involvement of bone. Incisional biopsy was done followed by histopathological examination with hematoxylin and eosin staining revealed connective tissue stroma consisting of cuboidal & plasmacytoid cells arranged in tubular, ductular, solid patterns. Large areas of keratin formation, myxomatous & mucoid tissue were seen (Figure 3). These histopathological features are suggestive of PA.

## DISCUSSION

PA accounts for 53-77% of parotid, 44-68% of submandibular, and 33-43% of minor salivary gland tumors (Lucas RB, 1984). Intraorally palate is the most common site followed by lips and buccal mucosa, retromolar area, floor of mouth, and alveolar mucosa (Gothwala K *et al*, 2012). Etiopathogenesis of PA is related to the myoepithelial cells and reserve cells in the intercalated duct. Neoplastically altered epithelial cells with the potential for multidirectional differentiation may be responsible for the tumor (Rajendran S and Sivapathasundaram S, 2009). 70% of tumors show karyotypic abnormalities, the most common of which are rearrangements involving chromosome 8q12-15 and 12q13-15. The target gene in PA with 8q12 abnormalities is putative pleomorphic adenoma gene (PLAG1) (Gneep D *et al*, 2009).

PA is predominantly seen in females in the age group of 40 to 60 years; in contrast to this present case was seen in a 45 year old male patient. Intraorally these tumors are slow growing, painless, smooth dome shaped, non-ulcerated, palpable rubbery masses and most commonly located on the postero-lateral aspect of hard and soft palate not crossing the midline (Ledesma-Montes C and Garces-Ortiz M, 2002). Whereas in present case the lesion was multinodular in appearance, located more anteriorly and crossing the midline. The most common adverse manifestations of tumors of this location are dysphagia, acute airway obstruction, and obstructive sleep apnea (Yoshihara T and Suzuki S, 2000). These tumors are able to invade and erode adjacent bone, causing radiolucency on the radiograph of maxilla, whereas in present case the lesion was 2X3 cm in size and long standing over duration of 15 years still there was no involvement of bone and patient did not have problems in speech and deglutition.



**Figure 2a:** Maxillary occlusal radiograph revealed that there was no involvement of bone. **Figure 2b:** Panoramic radiograph revealed that there was no involvement of bone.

Based on the above findings a final diagnosis PA of palate was made. Wide surgical excision of the tumor including periosteum was done with curettage of underlying bone in hospital setup (Figure 4). Specimen was sent for histopathology which was consistent with previous finding.



Considering the long standing history of the lesion and possibility of malignant transformation low grade malignant tumors of minor salivary gland such as carcinoma ex-pleomorphic adenoma, Mucoepidermoid carcinoma should always be ruled out. In present case patient did not show lymphnode involvement. Torus palatine and cystic lesions were ruled out based on clinical and radiographic findings.

Diagnosis of PA is always done on the basis of history, clinical examination, radiographic and histopathological investigations. Histopathological sampling procedures include Fine Needle Aspiration Cytology and core needle biopsy and the latter being more sensitive (Clauser L *et al*, 2004). Conventional radiographs like maxillary occlusal help in demonstrating bony invasion. Computed tomography can be used to see the bony invasion, erosions and magnetic resonance imaging (MRI) for soft tissue extensions of lesion, nerve involvement and perineural invasion. MRI is the preferred method of imaging for deeper tumors of the minor salivary glands (Kakimotoa N *et al*, 2009).

PA of the oral cavity lack a well-defined fibrous capsule, a feature associated with a high recurrence rate. Three histological subtypes include myxoid (80% stroma), cellular (myoepithelial cells predominating) and mixed (classic). Tumors with predominantly myxoid appearance are prone for recurrence (Chau MN and Radden BG, 1989). In present case cuboidal, columnar, plasmacytoid cells are arranged in tubular, ductal pattern and solid sheets. Mucooid, myxomatous areas and keratin pearls were also seen.

The management of PA of hard palate involves wide surgical excision of the tumor with removal of periosteum. Prognosis is excellent with cure rate of more than 95%. Reported recurrence rate was 6% and commonest reason being inadequate surgical technique such as simple enucleation leaving behind microscopic extensions (Kroll SO and Boyers RC, 2009). The risk of malignant transformation increase with duration of the tumor and propensity was documented to be 1.9-23.3% (Ethunandan M *et al*, 2006). In present case though the lesion was present for duration of 15 years there was no malignant transformation.

Majority of minor salivary gland tumors were reported to be malignant therefore complete history, careful clinical examination and appropriate investigations are recommended. Early detection is important to reduce surgical morbidity and to prevent malignant transformation. Long term follow up is important because of chances of recurrence.

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