Pleomorphic adenoma of a minor salivary gland– Report of a Case.

Abstract:

Pleomorphic adenoma is the most common benign tumor of the minor salivary gland which has elements of both epitrhelial and mesenchymal tissues. Majority of the salivary gland tumors occur in the second decade of life with a slight predilection for female. Clinically it presents as a firm or rubbery submucosal mass without ulceration or surrounding inflammation. Diagnosis is established according to the clinical examination, histopathology, and cytology, which are supported by radiographic findings of computed tomography and magnetic resonance imaging. Here, we report a case of pleomorphic adenoma of the hard palate in a 21-year-old female patient with a painless swelling in the left palatal region since nine months.

Keywords: *Pleomorphic adenoma, minor salivary gland tumor, palate*

Introduction:

Salivary gland tumors account for less than 3% of head and neck tumors.[1] Pleomorphic adenoma is the most common salivary gland tumor accounting for about 40 – 70% of all major and minor salivary gland tumors.[2] Pleomorphic adenoma is the most common benign tumor of minor salivary gland. The most common site of this tumor in the oral cavity is the palate (42.63%) followed by lip (10%), buccal mucosa (5.5%), and retromolar area (0.7%) lastly affecting floor of the mouth.[3] It is also called a mixed salivary gland tumor because of its dual origin from the epithelium and myoepithelial cells.[4] A case of Pleomorphic adenoma of the minor salivary gland of the palate has been discussed.

Case report:

A 21-year-old female patient reported to the Department of Oral medicine and radiology with a chief complaint of painless swelling over the left palatal region since nine months. The swelling was slow-growing, initially pea-sized, and increased to present size which is nontender not interfering with speech, mastication, and swallowing. There was no history of trauma, fever. Her past medical, past dental and family histories were noncontributory. On general physical examination, the patient was moderately

built, conscious with a normal gait. The vital signs were within normal limits. On extraoral examination, no abnormality was detected, and no lymph node involvement was noted.

Her intraoral examination revealed a solitary roughly oval-shaped, sessile swelling which approximately measures 2×2 cm extending from 5-6 mm from marginal gingival of left maxillary second molar till mid palatal region. The overlying mucosa was normal in color, not ulcerated. On palpation, the swelling was firm in consistency and appeared fixed to the underlying structure, nontender, nonpulsatile. There was no regional lymphadenopathy. Based upon the clinical findings and history, a provisional diagnosis of a Benign salivary gland tumor was considered. In the differential diagnosis, palatal abscess, odontogenic cyst, Kaposi's sarcoma, syphilitic gumma were included.

The Paranasal sinus view did not reveal any pathological changes in the bony structure. CT revealed lobulated soft tissue dense space-occupying lesion measures 3×2 cm noted in the regional soft palate on the left side with no calcific foci. MRI revealed a well defined oval-shaped mass measuring 2.8×2.0 cm pointed out in the region of the soft palate at the junction with hard palate, mildly to the left side extending up to midline. The lesion shows T2 and short T1 inversion recovery hyperintensities with few areas of hypo intensities in the center.

An incisional biopsy was performed under local anesthesia. The histopathological picture showed a tumor mass composed of epithelial and mesenchymal components with highly cellular and scanty connective tissue stromal cells. Areas of spindle cell proliferations resembling myoepithelial cells were evident. Myxomatous and chondroid areas were also seen. The patient was treated by wide local excision, and tumor was excised. There were no complications postoperatively and area healed well within six weeks.

Discussion:

Tumors arising from the minor salivary glands are uncommon clinical entities. Among them palate is the most commonly affected site followed by the upper lip and buccal mucosa respectively.[5] Pleomorphic adenoma is considered as the most common benign salivary gland tumor and literature search showed that eighty-four percent of pleomorphic adenomas occur in the parotid, eight percent in the submandibular gland and four to six percent in the minor salivary glands. Spiro RH et al. conducted a study on patients with salivary gland neoplasia in which he reported that twenty to forty percent of all salivary gland tumors arise from minor salivary glands.[8], mostly seen in fourth to sixth decades of life with a slight predilection for female gender. This case report was in concordance with this finding according to the

gender. Rahnama M et.al. reported that aetiology of Pleomorphic adenomais, in 70%, result of chromosome abnormalities involving pleomorphic adenoma gene 1 (PLAG 1) located on 8q12 and 12q15.[7]

Clinically it presents as a firm, painless swelling with intact overlying mucosa sometimes they have mucosal ulceration as was the case with this patient. Malignancy should be suspected in cases where ulceration of overlying mucosa is not result of trauma or biopsy[9]. Since pleomorphic adenoma of palate appears fixed to the bone, this is not caused by bony invasion but rather by the inelasticity of the palatal mucosa, which becomes distended by the tumor mass and may eventuate in cupped-out resorption of bone.[10] In other oral mucosal sites, it occurs as a freely movable, circumscribed mass.[11]

Diagnosis of Pleomorphic adenoma is established according to the physical examination, histopathology, which are supported by radiographic findings of computed tomography (CT) and magnetic resonance imaging (MRI).[12] Depending on the location and size of tumor, imaging with CT scan or MRI is helpful in setting the diagnosis and planning the treatment.[13]

Differential diagnosis of Pleomorphic Adenoma includes odontogenic and non-odontogenic cysts, soft tissue tumors, palatal abscess, mucoepidermoid carcinoma, adenoid cystic carcinoma, and salivary gland tumors. Palatal tissues contain components of soft tissue and harbor minor salivary gland tissues. As a result, soft tissue tumors such as neurofibroma, fibroma, lipoma, neurilemmoma as well as salivary gland tumors can also be considered in the differential diagnoses.[6]

As the name suggests, mixed histology which consists of three components: an epithelial, a myoepithelial elements and a mesenchymal component, which are arranged in varieties of cell patterns such as cord-like and duct-like along with areas of epidermoid metaplasia embedded in mucopolysaccharide stroma. Its microscopic diversity can exists in different areas of the same tumor and from one tumor to the other. The tumor is composed of the island of stellate and spindle cells that are interspersed in myxoid background.[14] It is typically encapsulated and well circumscribed tumor, but incomplete encapsulation is more common for tumors of minor glands, especially palatal lesions

Simple enucleation of the tumor has been reported with high recurrence. Therefore the treatment of benign minor salivary gland tumors is wide surgical excision along with the removal of periosteum and under lying bone if found to be involved. Many authors had advocated wide surgical excision with curettage of the underlying bone with a surgical curette or bur to avoid recurrence.[15] Recurrence of the lesion may be due to inadequate surgical techniques such as simple enucleation leaving behind

microscopic pseudopod-like extensions, capsular penetration, and tumor rupture with spillage of tumor cells.[12] Reconstruction of the palate should be considered for functional and aesthetic point of view. The soft tissue defect of the palate can be left to granulate, whereas the hard tissue defect can be corrected with the help of obturator. In the present case, the patient did not require any reconstruction as the palatal mucosa regenerated without any formation of a fistula.[15]

Conclusion:

Pleomorphic adenomas of palate is a most common tumor of minor slivary glands. It is a challenging entity to diagnose and to treat. Proper history, patient evaluation, histopathological, and radio imaging are necessary because of its clinical diversity. Early diagnosis and wide local surgical excision result in complete removal of the pathology with no recurrence. By adequate surgical excision, the tumor usually does not recur, but most recurrences can be due to inadequate surgical technique. A long term follows up is needed because of the recurrence even after several years of initial excision.

Consent Disclaimer:

As per international standard or university standard, patient's consent has been collected and preserved by the authors.

Figure 1a: Frontal view

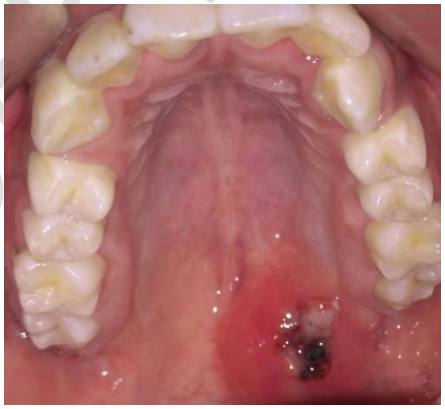


Figure 2: Intraoral Clinical picture showing swelling on the left side of the hard palate

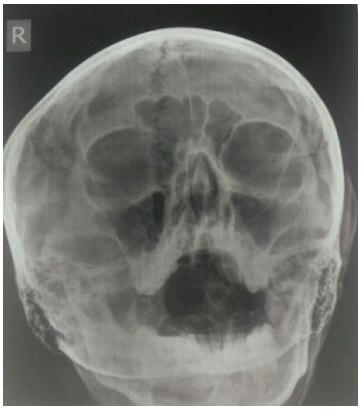
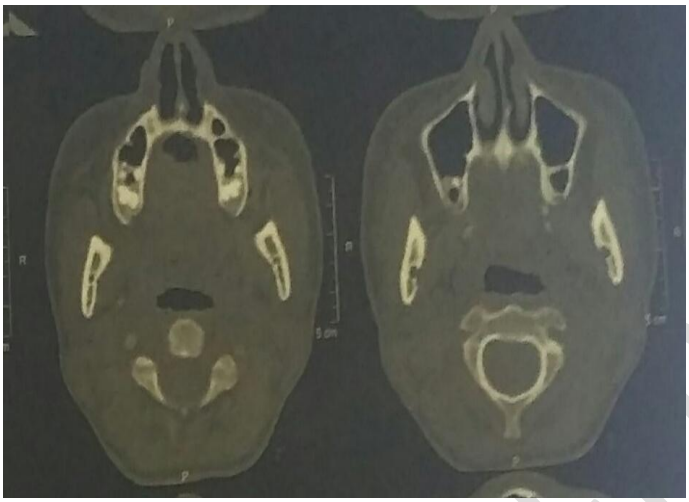


Figure 3: Para nasal sinus view showing no perforation of maxillary sinus



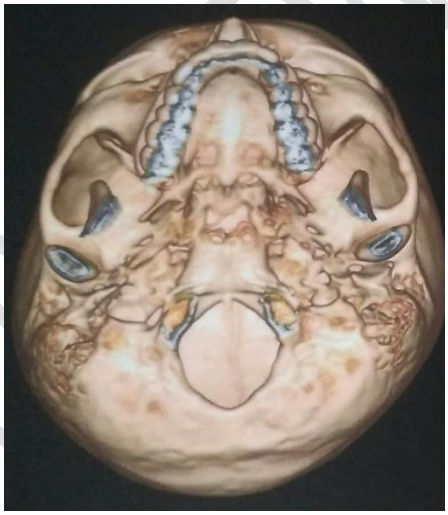
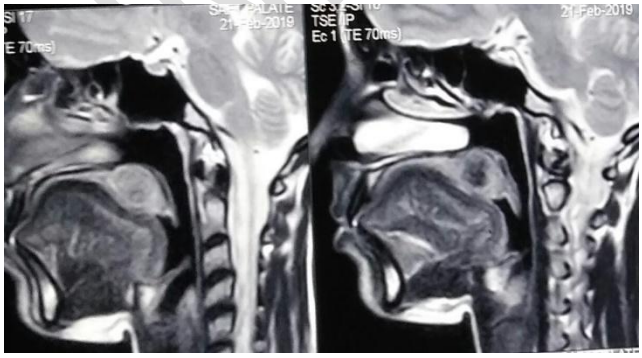


Figure 4a: CT reporting no erosion or perforation palatal bone

Figure 4b: 3d CT skull showing no pathological changes of palatal bone



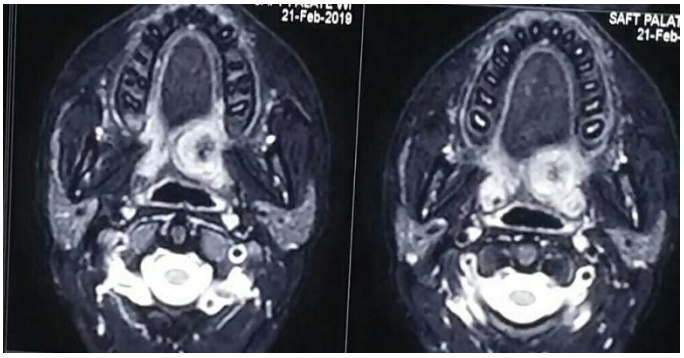


Figure5a : MRI sagittal view **Figure5b** : MRI axial view

Figure 5a, 5b showing well circumscribed lesion at the junction of hard and soft palate with hyperintensities and few areas of hypointensities in the center

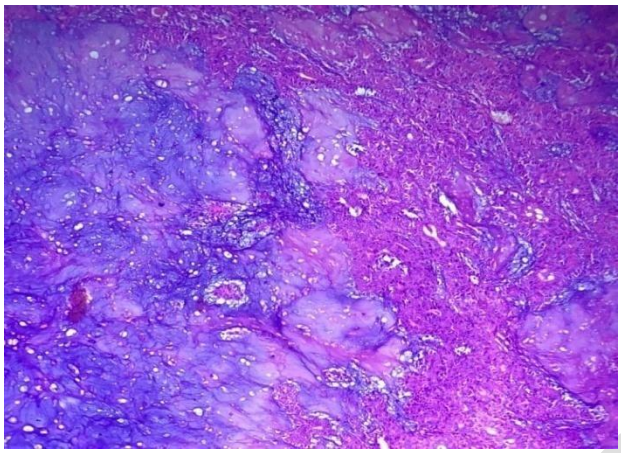


Figure 6a : Histopathological view showing ducts and myoepithelial cells surrounded by a hyalinized eosinophilic background

Figure 6b: Excised specimen showing a well encapsulated mass

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