Case report

Pleomorphic adenoma of a minor salivary gland.- A case report and review of a rare entity.

Abstract:

Pleomorphic adenoma is the most common benign tumor of the minor salivary gland. It is also called as a mixed salivary gland tumor because of its dual origin from the epithelial and myoepithelial cells. The most common site of this tumor is the palate. 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 of pleomorphic adenoma is established according to the clinical examination, histopathology, and cytology, which are supported by radiographic findings of computed tomography and magnetic resonance imaging. Palatal Pleomorphic adenoma is best treated by wide surgical excision including its surrounding capsule, involving the periosteum and overlying mucosa and followed by curettage of the underlying bone. 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. It is also called a mixed salivary gland tumor because of its dual origin from the epithelium and myoepithelial cells. Majority of minor salivary gland pleomorphic adenomas occur in the second decade of life. There is a slight predilection for the female gender.[3]

The clinical presentation of pleomorphic adenoma of the hard palate is typically a firm or rubbery submucosal mass without ulceration of surrounding inflammation. Pain and tenderness are unusual.[4] The term pleomorphic denotes the embryogenic basis of the origin of these tumors, which

contains both epithelial and mesenchymal tissues histopathologically.[5] Diagnosis of pleomorphic adenoma is established based on history, physical examination, and histopathology. CT and MRI provide the imaging information of the location, size, and extension of tumor to the surrounding superficial tissues and deep structures. The treatment of choice for pleomorphic adenoma in minor salivary gland is wide local excision with the removal of periosteum or bone if they are involved. Simple enucleation of the tumor leads to high local recurrence rate and should be avoided.[6] Here we report a case of pleomorphic adenoma of the hard palate in a 21 years old female patient with a complaint of painless swelling in the left palatal region since nine months.

Case report:

A 21-year-old female patient reported to the oral medicine and radiology department with the 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 about left maxillary second molar till mid palatal region. The overlying mucosa was normal in color and ulcerated. On palpation, the swelling was firm and rubbery in consistency and appeared fixed to the underlying structure, sessile, 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. Differential diagnosis was a palatal abscess, odontogenic cyst, Kaposi's sarcoma, syphilitic gumma.

The Paranasal sinus view did not reveal any pathological changes in the bony structure. CT revealed lobulated soft tissue dense space-occupying lesion measures 30 × 20 mm 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:

The Pleomorphic adenoma is also known as a mixed tumor, enclavoma, branchioma, endothelioma, and endochondroma. It contains both epithelial as well as myoepithelial elements. Hence it is considered as a mixed tumor of the salivary gland.[7] It is the most common benign salivary gland tumor where as mucoepidermoid carcinoma is a most common malignant counterpart to be encountered in maxillofacial region. 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] The mixed minor salivary gland tumors occur mostly in fourth to sixth decades of life with a slight predilection for female gender.[9]

Pleomorphic adenoma arising from the minor salivary gland of palate presents as a firm, painless swelling with intact overlying mucosa. However, if the mucosa is ulcerated, it is mostly attributed to trauma or biopsy. If not, the mass should be considered for a malignancy. 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] In the present case, the patient complained of unilateral slow growing non tender swelling in the junction of hard & soft palate.

Different types of imaging modalities are helpful for salivary gland tumors. The noninvasive diagnostic aids are ultrasound, CT, and MRI.[12] Computed tomography is the most important diagnostic tool of these tumors as it determines the extension of the lesion. It cannot invade bone but may lead to cupped out resorption of bone due to pressure effect.[13] Magnetic resonance imaging is better to display soft tissue invasion and perineural spread. These help in determining the local spread, the extent of disease, and also help to some extent in determining the type of tumors. These should be considered to assess the presence of bony erosion or soft tissue and nerve involvement.[12]

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]

Histopathologically, it is an epithelial tumor of complex morphology, possessing epithelial and myoepithelial elements arranged in varieties of cell patterns such as cord-like and duct-like along with areas of epidermoid metaplasia embedded in mucopolysaccharide stroma. Formation of the capsule is a result of fibrosis of the surrounding salivary parenchyma which is composed of the tumor and is referred to as a false capsule. The tumor is composed of the island of stellate and spindle cells that are interspersed in myxoid background.[14]

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.[15] Recurrence if at all occurs can be attributable 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 minor salivary glands on the palate, buccal mucosa or lips sometimes lack encapsulations and may enter into normal host tissue as the tumor grows. Proper history, patient evaluation, histopathological, and radio imaging are necessary.. 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.[10]

Figure 1: Frontal view of the patient.

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 image showa no erosion or perforation of palatal bone.

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

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 eosinophilic background.

Figure 6b: Excised specimen showing a well encapsulated mass

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Figure 1: Frontal view

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

Figure 3: Para nasal sinus view showing no perforation of maxillarv sinus.

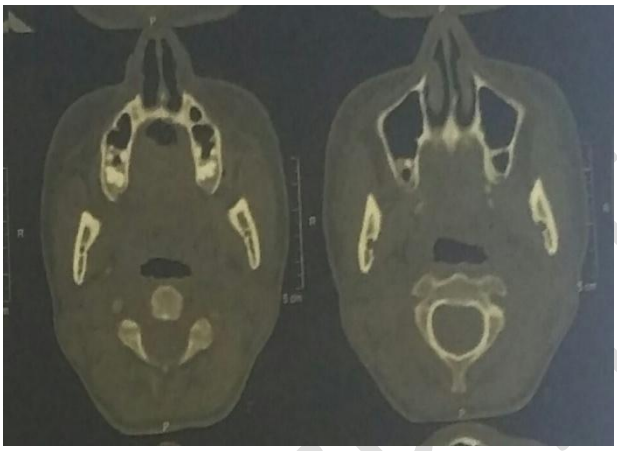


Figure 4a: CT image showa no erosion or perforation of palatal bone

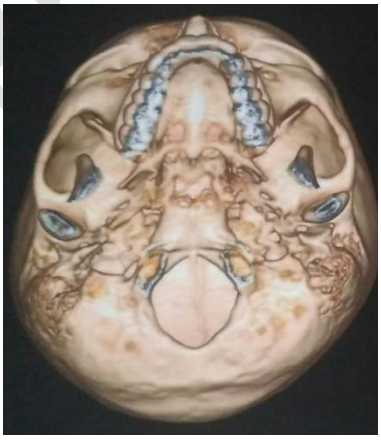
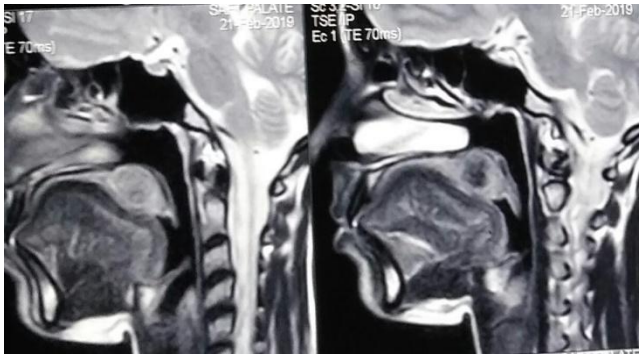


Figure 4b: 3d CT skull showing no pathological changes of palatal bone



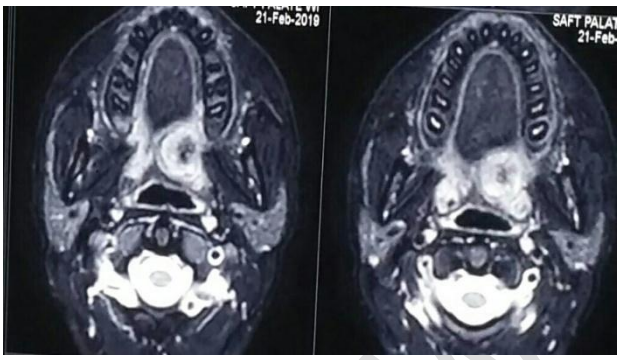


Figure5a : MRI sagittal

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

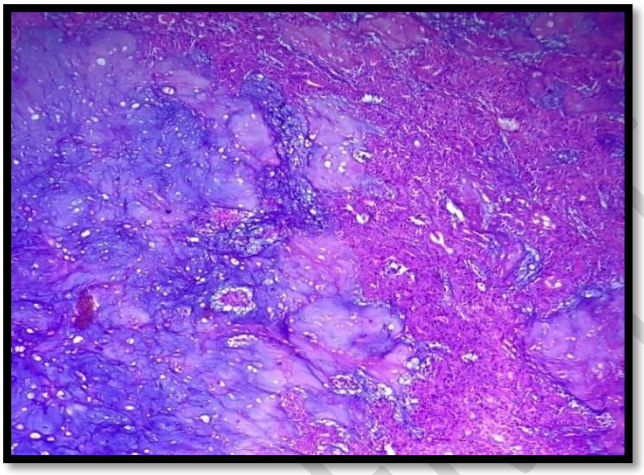


Figure 6a : Histopathological view showing ducts and myoepithelial cells surrounded by eosinophilic background

Figure 6b: Excised specimen showing a well encapsulated mass