

**Pleomorphic Adenoma of Oral Cavity Report of Five Cases with Emphasis on Causes of Recurrences – A Case series**

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**ABSTRACT**

Salivary gland tumors can be divided into three types: benign, locally aggressive, malignant tumors with potentiated to metastasize. Pleomorphic adenomas are the most common benign tumor of the major salivary glands. Infrequently, it may arise from the minor salivary glands and present as an intraoral mass over the palate or lip. We report a case series of five pleomorphic adenoma, with three cases involving hard palate and two involving of upper lip.

**Keywords:** palate, pleomorphic adenoma, reoccurrence, upper lip

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**INTRODUCTION**

Major salivary glands are more commonly affected by tumors than minor salivary glands. Pleomorphic adenoma happens to be the most common salivary gland tumor. Of the major salivary gland parotid gland is more commonly involved. Whereas palatal glands are more commonly involved in case of minor salivary glands, probably because of more number of glands are located on the palate as compared to other intraoral sites. But

majority of the tumour arising from these minor salivary glands are reported to be of malignant variety.

Pleomorphic adenoma usually presents as a slow growing, painless swelling, with intact overlying mucosa. Though benign, it requires wide surgical excision as it has high local recurrence rate. We are reporting five cases of pleomorphic adenoma out of which three cases involving the palate, two cases involving lip, with emphasis on clinical

presentation, causes of recurrence its management and differential diagnosis are discussed.

**CASE REPORT-1**

A 32 year old male patient reported with a chief complaint of swelling in the palate since 4 months. Swelling was sudden in onset, slowly increased in size, not associated pain. The swelling was totally asymptomatic except it causes difficulty in eating and speaking. Past

dental and medical histories were noncontributory.

Intraorally a solitary, dome shaped swelling was present at the junction of hard and soft palate on right side not crossing the midline. The swelling was well demarcated and measuring about 2x1cm in size with smooth surface. Overlying mucosa appeared slightly erythematous with normal surrounding mucosa.



**Dome shaped swelling on the right side of palate at the junction of hard and soft palate**

On palpation relevant inspeactory finding were confirmed. Swelling was non-tender on palpation, cystic in consistency with smooth surface. Swelling was not fixed either to overlying mucosa or underlying bone. Based on clinical examination a provisional diagnosis of minor salivary gland tumor was made.

Necrotizing sialometaplasia was considered under differential diagnosis.

Prior to excisional biopsy maxillary lateral topographic view and computed tomography was done which revealed no bone erosion. F.N.A.C. was done and suggestive of neoplasm of minor salivary gland, pleomorphic adenoma.

Surgical excision of the swelling was carried out under local anesthesia, and specimen was sent for histopathological examination. Microscopically section showed proliferation of epithelial & myoepithelial cells arranged in duct like structures with the presence of mucin myxomatous hyalinized areas were seen. Surface epithelium was stratified squamous with papillary areas. Capsule was seen covering the lesion.

**CASE REPORT-2**

A 68 year old female Patient reported with a chief complaint of swelling in the mouth since 4 months. Patient gave history of surgical excision of the swelling

three times in the past 10 years. There was difficulty in swallowing and speech and swelling was not associated with pain. No history of paresthesia. Past dental and medical histories were noncontributory.

On intraoral inspection a soft tissue growth was present on left side of palate measuring about 4X3 cm extending antero-posteriorly from 2 cm away from maxillary anterior alveolar ridge to soft palate area and mediolaterally involving the left maxillary alveolar ridge to 0.5 cm away from mid palatal suture. Overlying surface is smooth in the anterior region where as in the posterior part it is rough, erythematous and ulcerations were present.



**Soft tissue growth on the palate**

On palpation all inspectory findings were confirmed, it is firm in consistency, fixed

to the underlying bone and there was nontender on palpation. Based on history

and clinical examination provisional diagnosis of malignancy of minor salivary gland tumor was made.

Maxillary occlusal radiograph revealed deviated nasal septum and no bony erosion was noticed. Surgical excision was done and specimen was sent for histological examination which revealed pleomorphic adenoma.

### **CASE REPORT-3**

A 48 year old male patient reported to the department with a chief complaint of swelling in the palate for the past 12 years.

Earlier swelling was small in size & was not associated with any type of discomfort & pain. For the past one year swelling was growing & gained its present size. Past dental history revealed that he had undergone uneventful extraction of his teeth in the upper & lower back region of

the jaw 6 years back. His medical and family history was non contributory.

On intra oral examination a large well defined solitary swelling was present on the right side of the palate extending anteriorly to 1/3<sup>rd</sup> of the hard palate, posteriorly extending into the soft palate, laterally i.r.t to 13,14,15,16,17 region & medially crossing the midline of the palate. Dimension of the swelling was around 5x 4cm anteroposteriorly, and oval in shape. Its surface was smooth except one area where it was pointed. There was no discharge from the swelling, its border was distinct, and colour was normal. Mucosa adjacent to the swelling appeared normal. On palpation the following inspeactory findings of size, shape and number were confirmed. Swelling was firm in consistency, non compressible, nonfluctuant, non mobile & nontender.

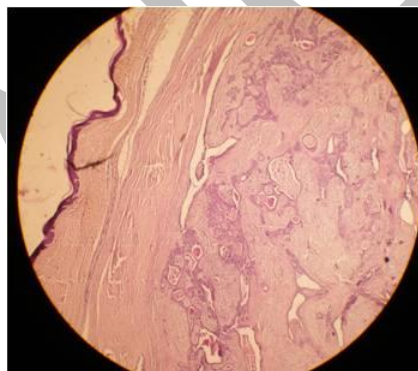


**Well defined swelling on the right side of the palate**

Aspiration was tried; it did not yield any content. Vitality test revealed all the teeth were vital.

Based on the history given by the patient and the clinical examination carried out, a provisional diagnosis of pleomorphic adenoma i.r.t palate was made. Differential diagnosis considered for the swelling was myoepithelioma, cystadenoma, muco epidermoid carcinoma & adenoid cystic carcinoma. Maxillary occlusal radiograph revealed no bony involvement. Paranasal sinus view showed no sinus involvement.

Surgical excision of the swelling was carried out under general anesthesia, and specimen was sent for histopathological examination. Microscopically section showed proliferation of epithelial & myoepithelial cells arranged in duct like structures with the presence of mucin myxomatous hyalinized areas were seen. Surface epithelium was stratified squamous with papillary areas. Capsule was seen covering the lesion. Based on histopathologic findings a final diagnosis of pleomorphic adenoma was made.



**Epithelial and myoepithelial cells arranged in duct like structures**

**CASE REPORT-4**

A 16 years old male patient reported to department with a chief complaint of swelling on right side of lip since 3 years. Swelling was small in size and not associated with any discomfort and pain. From last one year swelling was growing and gained present size. Past

dental, medical and family histories were non-contributory.

On extraoral examination a swelling was seen on right side of upper lip extending from nasal filtrum to right corner of the lip and from right nostril to upper lip measuring about 2.5 X 2 cm. Overlying skin was normal. On palpation

all inspectory finding were confirmed. No firm in consistency. rise in temperature, non tender, mobile and



Extra-oral swelling on right side of upper lip

On intraoral examination a well defined swelling was present, extending from upper labial frenum to upper right canine and was obliterating the labial vestibule. Overlying mucosa appeared normal. On palpation all inspectory finding were confirmed, non tender with firm in consistency. Swelling was mobile and was not fixed to underlying mucosa or overlying skin.



**Intraorally well defined swelling**



Based on the history given by the patient and the clinical examination carried out, a provisional diagnosis of odontogenic fibroma was made.

Surgical excision of the swelling was carried under general anesthesia, specimen was sent for histopathological examination. Microscopically section showed proliferation of epithelial & myoepithelial cells arranged in duct like structures with the presence of mucin myxomatous hyalinized areas were seen. Surface epithelium was stratified squamous with papillary areas. Capsule was seen covering the lesion. Based on histopathologic findings a final diagnosis of pleomorphic adenoma was made.

**CASE REPORT-5**

A 65 years old male patient, reported with the swelling of upper lip of 25 years duration. The swelling was very small to begin which gradually increased

in size. The swelling was totally asymptomatic since its appearance. No history of the swelling undergoing any secondary changes. But from past 2-3 months, patient felt that the swelling has suddenly increased in size causing disfigurement of face and forcing him to seek medical attention.

On examination, the entire lip was involved by the swelling and causing gross asymmetry of lower third of the face. The swelling also causing the obliteration of the patency of oral aperture. It was measuring about 3.5 X 2.5 cm, the surface was smooth, except at centre of the swelling, where it was ulcerated due to impingement of the mandibular anterior teeth.

On palpation swelling was firm in consistency, non tender and surface was smooth except in the center of the swelling, where it was nodular.

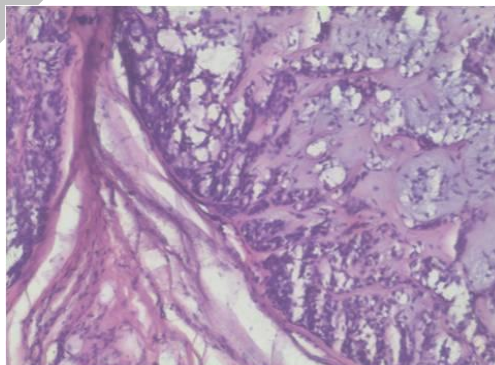


**Swelling on the lip**

Provisionally it was diagnosed as pleomorphic adenoma, considering its benign nature and slow growth. However, conditions like lymphangioma were considered in differential diagnosis.

Ultrasonography of upper lip revealed the swelling to be mixed echoic soft tissue mass with few cystic areas. These features were favouring neoplastic growth. After carrying out routine blood

investigations, an incisional biopsy was done and sent for histopathological examination which revealed lobulated mass of tumor cells of glandular epithelium surrounded by fibrous capsule showing no invasive area and reported as Pleomorphic adenoma.



**Lobulated mass of tumour cells surrounded by capsule**



Surgical excision of the tumor mass with an adequate margin was done. After the excision of the lesion, the enlarged upper lip was reconstructed by excising tissue on either side of the lip. The lip contour and symmetry was maintained. The wound healing was uneventful.



**After excision and reconstruction of enlarged lip**

### **DISCUSSION**

The term pleomorphic adenoma was suggested by Wills due to its unusual histological pattern. It is also known as benign mixed tumor, iceberg tumor, endothelioma, branchioma, enchondroma and the most common tumor of salivary glands. It accounts 60-70% of parotid, 40-60% of Submandibular and 40-70% of minor salivary gland tumors. Pleomorphic adenoma is more prevalent in 4<sup>th</sup> to 6<sup>th</sup> decade of life supporting this out of our five patients three belongs to 4<sup>th</sup> to 6<sup>th</sup> decade and two belongs to 2<sup>nd</sup> and 3<sup>rd</sup>

decade respectively. Intraorally most frequent site is palate (64.4%) followed by upper lip (18.8%), buccal mucosa (12.7%) and lower lip (2.7%). Out of our five cases three are of palate and two of upper lip. There is slight female predominance with a male to female ratio 1:1.17 to 1:2.4.

The term pleomorphic describes the embryogenic basis of origin of these tumors, which contains both epithelial and mesenchymal tissues.<sup>11</sup> It has been postulated that these tumors arise from intercalated and myoepithelial cells.<sup>9</sup>

Intraoral pleomorphic adenoma appears as slowly growing, painless, firm and non-tender mass<sup>10</sup>. In early stage the mass is mobile and may have intermittent growth. Tumour is having smooth surface in early stages, becomes more irregular, lobulated and may ulcerate in late stage. Local discomfort increases as the size increases and may be associated with pain when overlying mucosa ulcerates.

The tumor presents morphologically diverse features; however, both epithelial and mesenchymal elements must be present for diagnosis. Histopathology reveals a tumor composed of islands of stellate and spindle cells that are interspersed in a myxoid background. The pleomorphic nature is determined by an inner layer of epithelial cells and an outer layer of myoepithelial cells arranged in a variety of patterns associated with scant or abundant stroma. Variation may include squamous metaplasia, calcification, cartilage-like tissue, oxyphilic cells and rarely malignant transformation.<sup>9,10</sup>

Although it is a benign tumor, it has a high recurrence rate and in a small number of cases, benign pleomorphic adenoma may degenerate into a malignant

tumor.<sup>9,10</sup> Pleomorphic adenomas of the oral cavity lack a well defined fibrous capsule (incomplete capsule means tumor tissue merging with fat or salivary gland tissue), a feature associated with a high recurrence rate.<sup>10</sup> histologically capsule is subdivided into myxoid, classic, cellular type. In myxoid type proportion of stroma is 80%, classic type contains 30-50% of stroma whereas in cellular type proportion of stroma is 20-30%. Characteristically capsule is divided into incomplete capsule, capsule penetration, pseudopodium and satellite nodule.

These tumors are also able to invade and erode adjacent bone, causing radiolucent mottling on the x-ray of the maxilla<sup>12</sup> as compared to this none of our three cases involving palate had bone erosion.

The diagnosis of pleomorphic adenoma is established on the basis of history, physical examination, cytology and histopathology as done in our cases. F.N.A.C. is widely accepted tool for the diagnosis of salivary gland tumor with diagnostic accuracy ranging from 80-95%. Occlusal radiography, C.T. reveals bone erosion, which usually occurs in late stage of disease. In late stage margins are more

clearly evident in MRI as compared to CT. Ultrasonography revealed hypoechoic area.

Most common causes of recurrence is (a) implantation recurrence, due to rupturing of the capsule. (b) islands of tumor tissue left behind as a result of surgery. (c) multicentricity of pleomorphic adenoma.

The treatment of pleomorphic adenoma of the hard palate is surgical excision with a surrounding cuff of normal tissue.<sup>9,10</sup> The excision should include periosteum or bone if these are included<sup>10</sup>. These tumors usually do not recur after adequate surgical excision. Most recurrences can be attributable to inadequate surgical techniques such as simple enucleation leaving behind microscopic pseudopod-like extensions.<sup>10</sup>

Treatment of pleomorphic adenoma of lip is under great consideration because of aesthetics consideration. While doing surgical excision facial asymmetry and scar after surgery should be considered during treatment plan.

### **CONCLUSION**

Pleomorphic adenoma is a rare benign salivary gland tumor affecting the palate, lip. Although benign, but can undergo

malignant changes if diagnosed in late stage. So, early diagnosis and treatment result in complete cure leading to less morbidity.

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