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Pleomorphic Adenoma of Soft Palate: Report of A Case With Brief Review of Literature.

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Abstract: *Salivary gland tumors are a relatively rare and morphologically diverse group of lesions. Pleomorphic adenoma is a benign tumor of the salivary gland that consists of a combination of epithelial and mesenchymal elements. The tumor most commonly arises from the parotid (60-70%) or submandibular glands. It develops less frequently in a minor salivary gland, presenting as an intraoral mass. We are reporting a rare case of benign pleomorphic adenoma of soft palate with a brief discussion and review of literature.*

I. Introduction

Salivary gland tumors account for less than 3% of the head and neck tumors **[1]**.Pleomorphic adenoma (PA) is the most common neoplasm of both major and minor salivary glands. It accounts for 60% of all salivary gland tumors. Palate is considered as the most common intra-oral site (42.8-68.8%), followed by upper lip (10.1%) and cheek (5.5%). Other uncommon sites include throat (2.5%), retromolar region (0.7%), floor of mouth and alveolar mucosa **[2-4]**. Most of the palatal pleomorphic adenoma arises from the hard palate. Pleomorphic adenoma arising from the soft palate is very rare; only a few cases have been reported so far in the literature.

Pleomorphic adenoma usually occurs as a painless, slow growing lesion in the fourth or fifth decade **[5].** The highest incidence is between 30 to 60 years of age with female predominance **[6].** Pleomorphic adenoma appears as a painless firm mass and, in most cases, does not cause ulceration of the overlying mucosa. Generally it is mobile, except when it occurs in the hard palate. Intra oral mixed tumors, especially those noted within the palate, lack a well-defined capsule. Lesions of the hard palate frequently involve periosteum or bone. Approximately 25% of benign mixed tumors undergo malignant transformation. Treatment for the PA is radical excision of the tumour. Inadequate resection leads to local recurrence **[7].**

Case report and observation

A 28 year-old married male, presented in the department of Otorhinolaryngology, RIMS, Imphal with chief compliant of slow growing swelling over the soft palate on the left side since last 5 months. Patient complained of mild difficulty in deglutition because of swelling. There was no history of pain, headache, vertigo, ear discharge, trauma or any other associated symptoms. There was no history of fever, weight loss, bleeding, pus discharge or any other type of discharge from the swelling. The personal history of the patient did not reveal any history of smoking, or any other addiction. On past history there was no history of similar illness in past neither there was any past history of significant medical or surgical illness. There was no family history of similar complaints. The patient did not take any treatment for the above complaints and presented for the first time in our hospital for the present complaints.

General examination reveals normal and stable vital parameters. There was no icterus or pallor. The examination of other salivary glands and neck did not reveal any other swelling. The systemic examination of other systems was within normal limits. On local examination there was a globular mass on left side of palate with normal overlying mucosa. The lesion appeared to arise from near the anterior border of soft palate, extending up to the posterior border till the region of upper pole of left tonsil. The hard palate was not involved by the lesion (**Figure 1**). It was non-tender, firm, and non-compressible and had well defined margins. Rest of otorhinolaryngological examination did not reveal any other significant abnormality. On the basis of history and clinical examination a diagnosis of solid non infectious mass of soft palate likely of benign etiology was made. The possibility of infective etiology (palatal abscess) was less likely as there was no fever, pain or tenderness, erythema or fluctuation.

Contrast enhanced computed tomography scan on a dual source 128 slice spiral CT with coronal and axial reformations revealed a well defined homogeneously non-enhancing hypodense soft tissue lesion of size 3cm x 1.8cm x 2 cm arising from left side of soft palate and abutting the hard palate anteriorly, uvula posteriorly, and nasopharyngeal wall superiorly. Laterally it was seen to be abutting the left lateral nasopharyngeal wall (**Figure 2(A), 2(B)**. There was no infiltration of the surrounding structures with well-defined surrounding planes. There was no non-enhancing area within to suggest central necrosis of secondary infection. The radiological features were suggestive of a benign neoplastic mass. The possibility of other clinical diagnosis of palatal abscess was completely ruled out. Transoral FNAC from the soft palate mass revealed features suggestive of pleomorphic adenoma.

Surgical excision of the lesion under general anaesthesia was planned. All preoperative routine investigations including blood and urine examinations were within normal limits. The patient underwent surgery under general anesthesia. A Boyle-Davis mouth gag was applied to access the mass transorally. Nasopharynx and oropharynx were packed with ribbon gauze. Packing of the nasopharynx helped stabilizing the soft palate during the dissection. Extra capsular excision with a flap elevation technique was done through a trans-mucosal curved incision with neural pathway preservation (**Figure 3**).The excised mass was 3.5 × 2.5 cm (**Figure 4**) and surgical wound was closed in two layers with advancement of adjacent mucosa. Histopathological examination revealed proliferating ducts and plasmacytoid myoepithelial cells in a cartilaginous myxoid stromal background along with other features of pleomorphic adenoma **Figure 5(A), 5(B).** The patient's postoperative course was uneventful. The healing was satisfactory with no scarification of the soft palate. No recurrence was observed after a follow-up period of 6 months (**Figure 6**).

II. Discussion

Pleomorphic adenoma is the most common tumor of major salivary glands, although approximately 80% of these are found in parotid gland. About 4-5% of pleomorphic adenomas are seen in minor salivary glands, amongst which palate is one of the sites **[8]**. Other sites include submandibular gland, lips, buccal mucosa, gingiva and tongue. It is composed of epithelial and myoepithelial cells arranged with various morphological patterns, demarcated from surrounding tissues by a fibrous capsule. It also ranks first as the most common tumor of the intraoral salivary glands of which palate is the most common intraoral site, followed by upper lip and buccal mucosa. Muco-epidermoid carcinoma is the most common malignant salivary gland tumor, while pleomorphic adenoma is the most common benign counterpart. Pleomorphic adenoma of the soft palate is rare [**9**].

Patients with pleomorphic adenomas of the minor salivary glands present mostly in fourth to sixth decades, with a slight predominance in female **[10]**. They usually present as a unilateral, painless, slow-growing mass in the parotid gland. However, when they originate in the hard and soft palate they present typically as a firm or rubbery submucosal mass without ulceration or surrounding inflammation.

The differential diagnoses for this case include palatal abscess, odontogenic and non-odontogenic cysts, and other soft tissue tumors. Abscess can be ruled out because of lack of signs and symptoms of inflammation, whereas cysts are not firm in consistency. FNAC should be performed as an adjunct to diagnosis prior to definitive surgical treatment. Computed tomography or magnetic resonance imaging should be considered when assessing for presence of bony erosion or soft tissue and nerve involvement [**11].**

The histological pictures of pleomorphic adenomas vary. Pleomorphic adenomas of the extra-major salivary glands are similar to those in the major salivary glands and are composed of a mixture of epithelial and stromal elements. Three main histologic subgroups have been identified: myxoid (80% stroma), cellular (myoepithelial predominant), and mixed (classic) type [**12].**

Surgical excision is the treatment of choice. Excision with a wide margin is recommended. In the palate, excision is usually done along with the overlying mucosa. In the present case, extracapsular excision was done with a flap elevation technique as there was a well-defined fat plane between the mass and surrounding tissues with no features of tumour infiltration. The flap technique is preferred because of less chance of cicatrization of the soft palate with its attending complications.

Longevity and recurrence are risk factors for malignant transformation. The propensity for malignant transformation is documented to be 1.9-23.3% [**13].** Recurrence rate of 2% to 44% in the pleomorphic adenoma has been reported in the literature **[14]**.

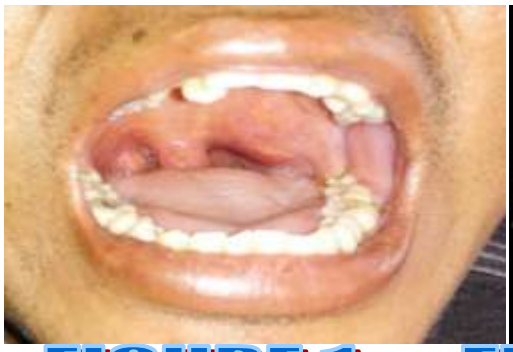

FIGURE 1

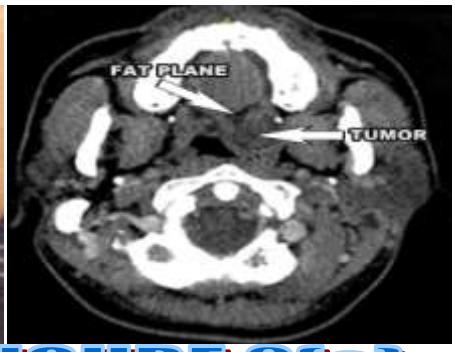

FIGURE 2(a)

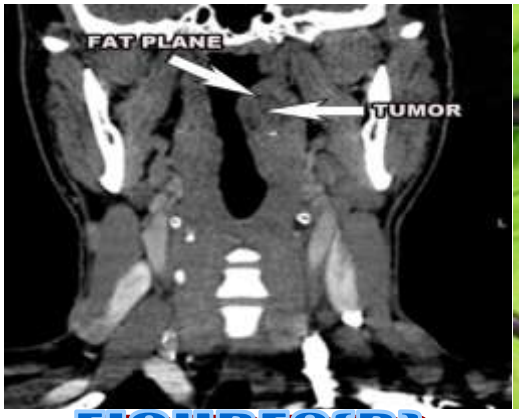

FIGURE2(B)

FIGURE 3

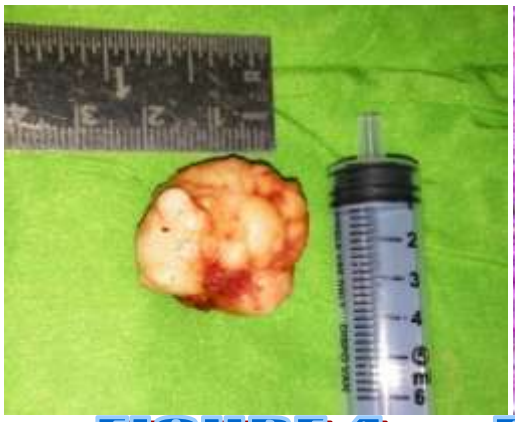

FIGURE 4

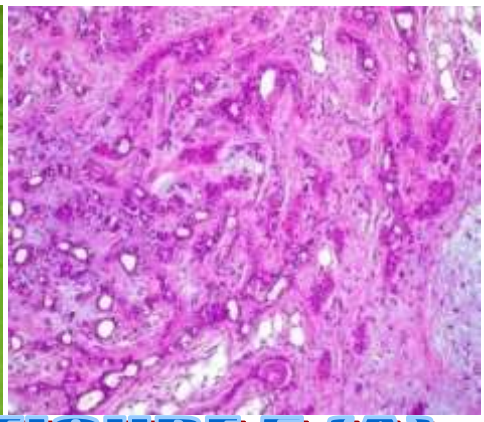

FIGURE 5 (A)

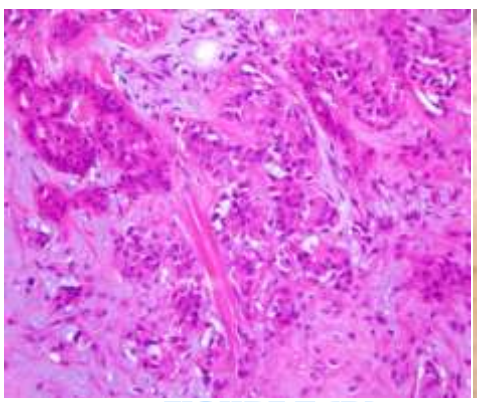

FIGURE 5 (B) Higher Magnification

FIGURE 6

1) **FIGURE 1:-** Shows the tumor at the time of presentation.
2) **FIGURE 2** (A) (B):- Shows the tumor with a well defined fat plane in axial and coronal cuts respectively.
3) **FIGURE 3:-** Intraoperative wound closure after excision.
4) **FIGURE 4:-** Shows the specimen after excision.
5) **FIGURE 5** (A) and (B):- Shows the histopathological picture with epithelial and mesenchymal components in normal and high magnification respectively.
6) **FIGURE 6:-** Shows postoperative photograph after 6 months.

Review of literature

1) Lomeo P et al. in 2001 reported a18-year-old female presented with dysphagia and difficulty breathing due to a mass in her throat. On examination a 3-cm mass of the soft palate was noted obstructing the nasopharynx and impinging on the oropharynx. The mass was completely removed under general anaesthesia. The frozen section diagnosis was a pleomorphic adenoma. The soft palate was reconstructed. The postoperative course was uneventful, and there was no evidence of velopharyngeal insufficiency. **[15]**

2) Kurokawa H et al in 2008 reported a 34 year old male with two year duration of slowly growing tender mass in the right soft palate .On examination mass was 3*3 cm in dimension mildly tender and elastic hard. CT revealed a well-defined nodular mass in the right soft palate with a peripheral rim of enhancement and low attenuation. Magnetic resonance imaging (MRI) showed a mass lesion in the right soft palate with well defined margins. The entire mass was removed under general anaesthesia. Gross examination of the surgical specimen showed a brown, entirely necrotic, well-encapsulated lesion measuring 37* 35* 27 mm. Histologic and immune-histologic examinations led to the diagnosis of pleomorphic adenoma with extensive necrosis. There was no recurrence in the 13 months follow up period.**[16]**
3) Chen HH et al. in 2010 reported a case of 60-year-old male had a 3-month history of a small soft palatal mass with progressing left cheek numbness, proptosis, and disturbed vision. On examination an area of numbness in the distribution of 2nd division of trigeminal nerve was found, proptosis, and disturbed visual acuity at the level of counting fingers, left esotropia which was due to traumatic left abducens nerve palsy by saw dust 10 months ago. Biopsy of the tumor revealed Pleomorphic Adenoma. CT revealed asymmetry with some bulging contour at palatal region. MRI revealed irregular soft tissue mass involving left maxilla, and extended from pterygopalatine fossa, inferior orbital fissure to Cavernous Sinus with bony destruction of the skull base. Under general anaesthesia total maxillectomy with free flap reconstruction intending to radically excise the tumor was performed, pterygopalatine fossa was explored but complete excision of tumor was infeasible at the skull base. After excision of the tumor, HPE and immunohistochemistry revealed a diagnosis of carcinoma ex pleomorphic adenoma with perineural and lymphatic spread. Concurrent chemo-radiotherapy (CCRT) was given after surgery due to positive resection margin and advanced tumor stage. The tumor status was completely remitted without deterioration of visual acuity, proptosis and slightly improvement of left check numbness. Five months later, the patient complained of deteriorated visual acuity to the level of light sensation. Follow-up MRI revealed the tumor recurred at left Cavernous Sinus. CCRT for the recurrence was implemented and the disease had been in a stable condition. However, the vision in his left eye was completely lost after the 2nd CCRT.**[17]**
4) In 2011 Sharma Yogesh et al. reported a 45 year female with one year history of painless progressive swelling over left palatal region, on evaluation with CT scan no bony erosion was seen .Under local anaesthesia excision was carried out with 1 cm clear peripheral margin. HPE reported as pleomorphic adenoma with no post operative recurrence.**[18]**
5) Shetty CK et al in 2012 reported a female of 65 years presenting with a slow-growing 4 * 3 cm painless swelling on the soft palate without any complaints of dysphagia. There were no enlarged cervical lymph nodes. Fine needle aspiration cytology of the mass showed features suggestive of Pleomorphic adenoma. CT scan of the neck was done with contrast, which showed a well-defined, mildly-enhancing mass measuring about 4 cm by 3 cm on the right soft palate with areas of calcification. Excision of the mass was done under general anaesthesia. The mass was excised with preservation of the overlying mucosa. No evidence of recurrence or any other lesion in a 5 month follow-up period was noted.**[19]**
6) Daryani D et al. In 2012 reported a 40-year-old male patient with a complaint of difficulty in swallowing, decreased appetite and hoarseness of voice since 1 week due to swelling in the palate since 1 week. On examination, a solitary, 3 × 3 cm, well-defined, roughly oval swelling was noted on the right soft palate. A FNA biopsy revealed Spindle-shaped mesenchymal cells in the stromal matrix suggestive of pleomorphic adenoma. Conventional occlusal radiographs revealed no bony changes to the hard palate. There was no evidence of calcification. Surrounding bone was normal. Wide excision of the lesion was performed along with curettage of underlying bone. An encapsulated mass measuring approximately 2.4 cm × 1.9 cm was recovered from the soft palate. HPE revealed a final diagnosis of Pleomorphic adenoma, myoepithelial cell

(plasmacytoid type) predominant, of the soft palate. There were no postoperative complications and no recurrence was seen during the 1 year follow-up period.**[20]**

7) In 2013 Sahoo NK et al. reported a 45-year-old female who presented with a painless slow growing swelling of palate over the last 20 years. The mass was extending to oropharynx causing mechanical obstruction of airway. MRI imaging depicted an oval-shaped mass occupying oropharynx and displacing the tongue inferiorly. Fine needle aspiration cytology (FNAC) was suggestive of Pleomorphic Adenoma. The entire tumor mass was excised along with overlying mucosa. Histopathological examination confirmed diagnosis of Pleomorphic Adenoma of minor salivary gland. There was no recurrence of the lesion since 1 year.**[21]**

8) Rout RM et al. in 2015 reported a 50 years old female with swelling in the mouth since one year. It was associated with bilateral complete nasal block, difficulty in swallowing since 2 months and progressive change in the voice since 2 months. On examination a smooth globular mass in the region of the soft palate more towards the right side. Visible size of the mass was about 8 × 6 cm nearly obliterating oropharynx. On DNE, the mass extended to the nasopharynx and blocking it completely. CT scan picture showed a heterogeneous mass extending up to the right pterygoid plates without any evidence of bone erosion. FNAC was suggestive of pleomorphic adenoma. Tracheostomy was done to provide general anaesthesia as tumor obliterated oropharynx. Tumor tissue was dissected out by finger dissection. Soft palate was repaired in two layers. HPE was confirmative of cellular pleomorphic adenoma. There was no recurrence in the post operative period.**[22]**

9) Kumar VM et al. in 2015 reported a 30-year old female with a painless swelling over the left side of soft palate of 3 months duration. FNAC of the swelling was suggestive of an inflammatory lesion with inconclusive diagnosis. CT scan revealed no bony involvement. Wide local excision with clear margins was performed under general anaesthesia. HPE confirmed it to be pleomorphic adenoma of minor salivary gland in the soft palate. Follow-up was done for a period of 10 months without any recurrence.**[23]**

10) Hmidi Mounir et al. in 2015 reported a 45 year-old female, presenting with painful deglutition and slow growing swelling over palate since 6 months. CECT revealed no infiltration of the surrounding structures with well defined surrounding planes, no non enhancing area within to suggest central necrosis of secondary infection. Excised mass was 5 × 4 cm and surgical wound was closed with advancement of adjacent mucosa. HPE was conclusive of pleomorphic adenoma. There was no post operative complication after 1 year of follow-up period.**[24]**

III. Conclusion

Pleomorphic adenoma of the soft palate is very rare with only a few reports in the literature. They are usually asymptomatic and, so, may present late in the course of the disease process with possible malignant transformation. Early diagnosis and timely intervention is key to a successful management of this rare but potentially malignant lesion.

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