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Intraoral Pleomorphic Adenoma: Report of A Case In A Young Patient

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Abstract:
Salivary gland tumours are relatively rare & morphologically diverse group of lesions. Intraoral minor salivary gland tumours comprises approximately <25% of all salivary gland tumours. Pleomorphic adenoma is a slow growing benign salivary gland neoplasm characterised by proliferation of parenchymatous glandular cells along with myoepithelial components. It is the most common type of salivary gland tumour and the most common tumour of parotid gland. It is also known as Mixed type salivary gland tumour. It refers to its dual origin from epithelial and myoepithelial elements.
A 16-year-old female patient presented with a swelling at the junction of the hard & soft palate on the left side. it was painless, firm in consistency & adherent to the overlying structures. USG showed a mixed echoic lesion predominantly hyperechoic & internal septations in some areas. Fine needle aspiration was done. Surgical excision and histopathological examination were followed.

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I. Introduction

Salivary gland tumours are clinically & morphologically diverse lesions that constitute less than 0.5% of the total head & neck malignancies of which 80% are benign[1]. Pleomorphic adenoma is the most common salivary gland tumour. It accounts for 50% of all salivary gland tumours of which 65% is in the parotid gland. It constitutes 40% of all intraoral salivary gland tumours & about 50% of these occurs in palate. It occurs more frequently in females than in males with a ratio of 6:4. The common age group to be affected by this tumour falls in the 4^{th}- 6^{th} decades of life with an average age of occurrence about 43 years. However, cases have also been reported in young adults & children[2]. Here we present a case report of Pleomorphic adenoma in a 16-year-old female.

II. Case Report

A 16-year-old female patient came to Government Dental College, Kozhikode demanding orthodontic treatment for her forwardly placed upper front teeth. During her intraoral examination a swelling was noticed at the junction of the hard & soft palate on the left side. Patient was totally unaware of the presence of swelling. The swelling was round with a diameter of about 3 cm extending anteriorly 1cm from the distal aspect of palatal marginal gingiva of 26, posteriorly up to two-third of the soft palate, medially from the midline extending distally up to the left maxillary tuberosity. It had a smooth & bosselated surface with a central area showing ulceration. Its margins were well defined & overlying mucosa was erythematous. On palpation it was painless, firm in consistency & adherent to the overlying structures. It was non pulsatile & non fluctuant. There was no associated bleeding or pus discharge. Based on these findings a provisional diagnosis of benign salivary gland neoplasm was made. The differential diagnosis includes Necrotizing Sialometaplasia, benign neural connective tissue tumours like Neurofibroma & Neurilemmoma

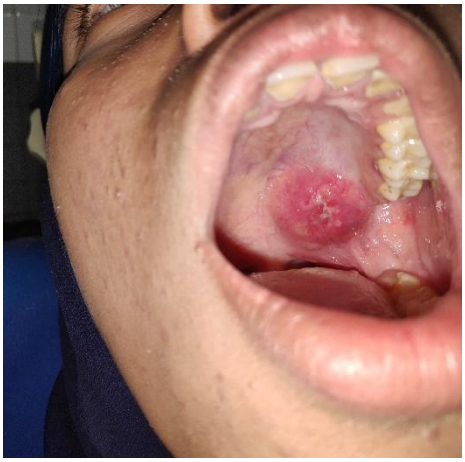
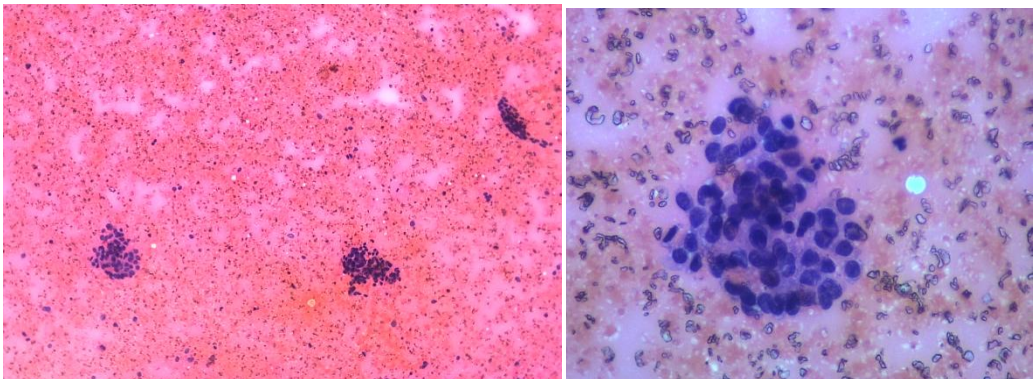


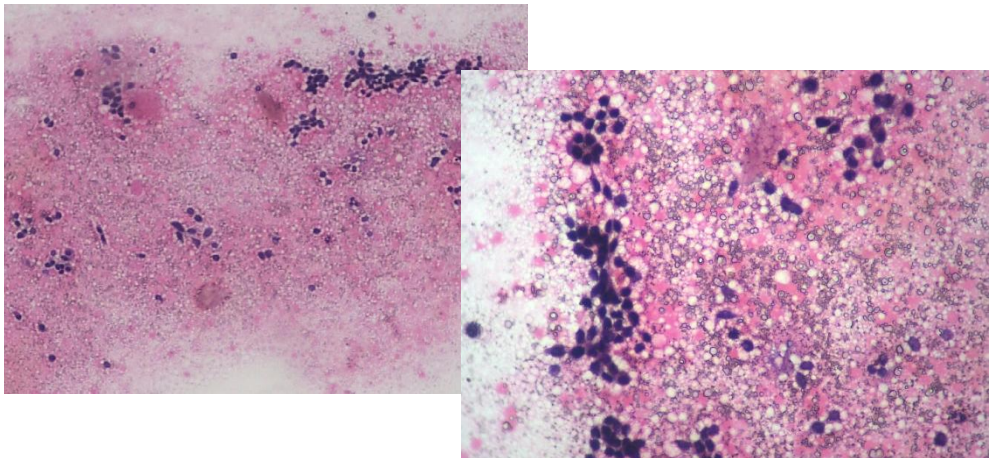
Figure 1: Clinical photograph of patient showing swelling in the palate

USG & MRI was advised. USG showed a mixed echoic lesion predominantly hyperechoic & internal septations in some areas. Margins were scalloped at some areas. Acoustic enhancement was present & vascularity was noted within the lesion. USG was suggestive of a benign minor salivary gland neoplasm probably pleomorphic adenoma. MRI showed a well differentiated T1, hypointense T2 & STIR heterogeneously hyperintense moderately enhancing mass lesion of size approximately 2.9*2.5*1.8 cm arising from the posterior inferior aspect of the left half of the hard palate extending to the soft palate. Features are consistent with benign soft palate lesion possibly minor salivary gland neoplasm.

Fine needle aspiration was attempted which showed scanty smear. Cytosmear showed numerous clusters of round to ovoid cells with hyperchromatic nuclei (ductal cells) & few spindle cells (myoepithelial cells)intermingled with minimal inflammatory infiltrate in a fibrinous background suggestive of pleomorphic adenoma.

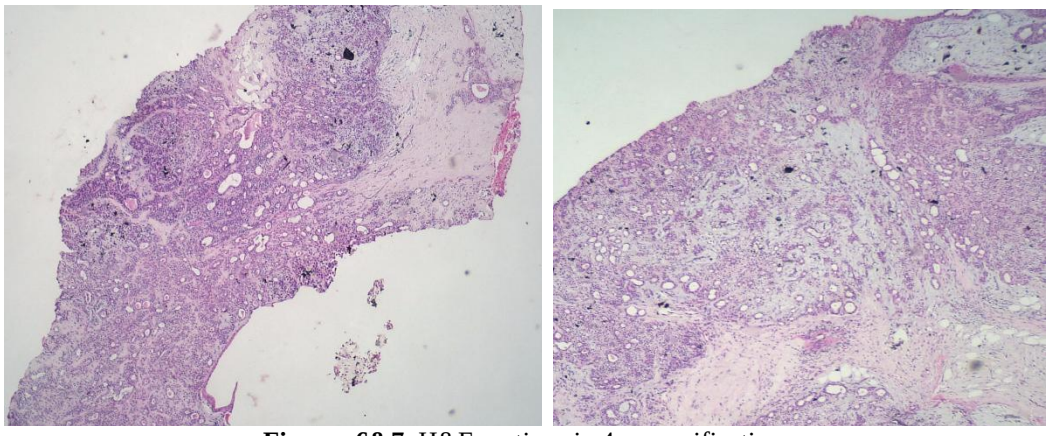


Figures 2&3: Cytosmear in 10x and 40x magnification respectively

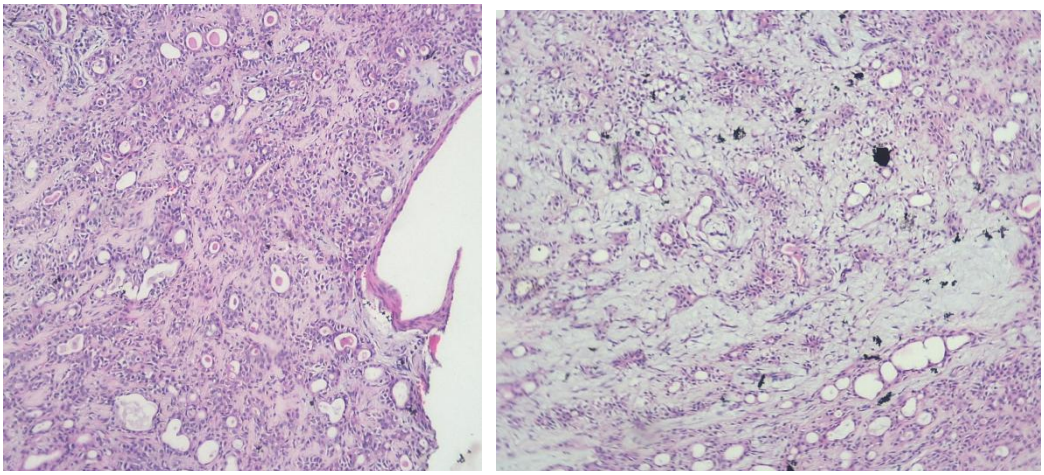


Figures 4&5: Cytosmear in 10x and 40x magnification respectively

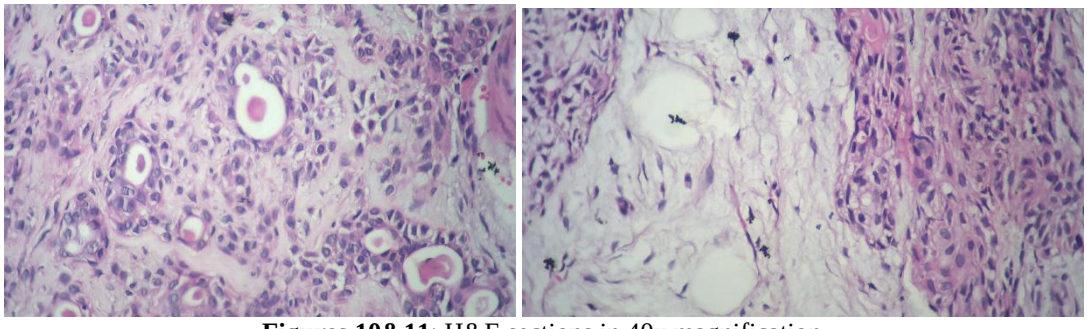
An incision biopsy followed by excision biopsy was done. Microscopically H&E stained sections of excision biopsy showed tissue composed of tumour cells proliferating as duct like structures filled with eosinophilic coagulum and a few nests & cords. Tumour cells are of cuboidal, spindle & plasmacytoid morphology. Areas showing squamous metaplasia & keratin pearl formation are seen. Myxoid & hyaline like areas are also present. Intervening connective tissue stroma is moderately collagenous with a few lobules of adipocytes & endothelium lined vascular spaces. Thus a confirmatory diagnosis of pleomorphic adenoma was made.



Figures 6&7: H&E sections in 4x magnification



Figures 8&9: H&E sections in 10x magnification



Figures 10&11: H&E sections in 40x magnification

III. Discussion

Pleomorphic adenoma was first described by Billroth in 1859 & it was Minssen who introduced the term mixed tumour for this entity in 1874. In 1953, Willis coined the term Pleomorphic adenoma for these histologically complex tumours[3]. Pleomorphic adenoma still continues to be the most common intraoral salivary gland neoplasm. Hayashi et al in their retrospective observational study done in the Japanese population

observed Pleomorphic adenoma to be the most frequent intraoral minor salivary gland tumour accounting for 41.2% of all tumours with majority of cases occurring in the palate[4].

Pleomorphic adenoma is a benign neoplasm consisting of cells with epithelial (luminal) & myoepithelial (abluminal) differentiation accompanied by variable amounts of characteristic stroma. The epithelial & mesenchymal coexistence with in the lesion resulting in amalgamation of cellular & stromal components is responsible for its mixed appearance[5]. Numerous theories have been put forward explaining the histogenesis of this tumour. The most accepted concept in its origin from myoepithelial cells reserve cells in the intercalated duct[2]. Overexpression of genes PLAG1 & HMGA2 is held responsible for the pathogenesis of Pleomorphic adenoma[6,7].

The most common age group for Pleomorphic adenoma is between fourth & sixth decade. Its occurrence in the second decade as in our case is rare. Literature reports numerous cases of Pleomorphic adenoma in children below 18 years of age. Jorge et al reported five cases of intraoral Pleomorphic adenoma in patients below 18 years[8]. Literature suggests that Pleomorphic adenoma at a younger age is associated with higher recurrence rate[9]. The other factors contributing to increased recurrence rates are enucleation alone, rupture or spillage during removal, presence of protuberences beyond the main tumour & abundance of chondromyxoid stroma[10].

The role of FNAC in diagnosis of salivary gland tumours has been emphasized extensively in literature. Singh et al diagnosed 64 cases of minor salivary gland tumours based on cytological findings of which 50% were Pleomorphic adenoma[14]. However, a few malignant lesions like epithelial myoepithelial carcinoma, carcinoma ex pleomorphic adenoma & mucoepidermoid carcinoma were diagnosed as Pleomorphic adenoma in FNAC[11,12]. Hence Pleomorphic adenoma is a known mimicker of numerous benign, malignant & reactive conditions, a final diagnosis must always be confirmed on the basis of histopathology[13].

Macroscopically they appear as irregular or ovoid mass with well-defined borders. Tumours of minor salivary glands are commonly not encapsulated. The cut surface may be rubbery, fleshy, mucoid or glistening with a homogenous tan or white colour. The microscopic hallmark of Pleomorphic adenoma is the variable & diverse histologic pattern brought about by the glandular epithelium & mesenchyme like tissue. The epithelial component is arranged as ducts or small cysts that may contain an eosinophilic coagulum or in the form of islands & sheets. Squamous metaplasia with keratin production may be seen. The myoepithelial cells may be angular, spindle or plasmacytoid in appearance. They contribute to variable stromal pattern which may be chondroid, Myxoid, osseous or hyaline like areas. Melting of myoepithelial cells into the stroma is a prominent finding[5]. Depending on the proportion of cellular & stromal component, Foot & Frazell have categorized Pleomorphic adenoma into four categories; (a) principally Myxoid (b) Myxoid & cellular in equal proportion (c) Predominantly cellular (d) Extremely cellular. The myxiod variants are associated with increased chances of recurrence& the cellular varaiant has a greater tendency to undergo malignant change[2].

Pleomorphic adenoma is commonly managed by conservative surgical excision. In the current patient the tumour was excised & closed by sutures. She had an uneventful post-operative healing period. Post excision of palatal Pleomorphic adenoma an obturator or splint may be placed for its proper healing. Surgeons also use another simple & relatively inexpensive technique inspired by "Bagota bag" method used by General Surgeons[15].

IV. Conclusion

Pleomorphic adenoma is the most common salivary gland tumour. The feature that makes this case stand out is its age of presentation. As younger age is associated with increased risk of recurrence, the current patient requires strict follow up. For many salivary tumours diagnosis is straightforward but the wide range of morphological diversity between and within tumour types means that a diagnosis may not be possible on small incisional biopsies and careful consideration of the clinical and pathological features together is essential.

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