PLEOMORPHIC ADENOMA OF A MINOR SALIVARY GLAND- REPORT OF A CASE

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ABSTRACT

Pleomorphic adenoma(PA) is the most common benign salivary gland neoplasm which affects the major salivary glands (90% occur in the parotid gland) and infrequently arises from minor salivary glands(10% of them occur in the minor salivary glands). Also called benign mixed tumor, this salivary gland neoplasm has elements of both epithelial and mesenchymal tissues. This case report highlights the varied clinical and histopathological features of this benign tumor which could be misdiagnosed as a malignancy.

Key words *- benign tumor, minor salivary gland, palate.*

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INTRODUCTION

Pleomorphic adenoma (PA) is a benign salivary gland tumor which represents about 3-10% of the head and neck neoplasms.[1] It is the most common salivary gland tumor occurring mainly in parotid and sub mandibular salivary gland. Pleomorphic adenomas are more likely to be malignant when associated with minor salivary glands (50%). As far as intra oral sites are concerned, palate (42.63%) is the most commonly affected site, followed by lip (10%), buccal mucosa (5.5%), retromolar area (0.7%) and lastly affecting the floor of the mouth.[2] Pleomorphic adenoma usually presents as a mobile, slow growing, painless, firm swelling that does not cause ulceration of the overlying mucosa but may cause erosion of the underlying bone. Wide surgical excision is the treatment of choice.[3]

CASE REPORT

A 26 year old male patient reported to the OP of the Faculty of Dental Sciences, Sri Ramachandra University with a complaint of a swelling in the palate for the past 40 days. History revealed that the patient noticed a swelling on the roof of the mouth which was small in size initially and gradually increased to reach its present size. Patient had visited a dentist in his hometown who carried out a fine needle aspiration procedure and subsequently advised surgery. However, the patient did not have any concrete medical report for the procedure undergone. Personal history revealed that the patient was a non-smoker, non-alcoholic and did not have any habits of chewing tobacco or betel nut. On intraoral examination, mouth opening was found to be normal. A single, well defined, localized swelling was evident on the right side of the palate measuring about 3cms both antero-posteriorly and medio-laterally. The movements of the soft palate were not restricted and the uvula and fauces appeared normal. Anteriorly the swelling extended 3cms away from the rugae upto 2cms posteriorly away from the hard palate. Laterally, it extended 2cms away from the marginal gingiva, medially up to the midline of the palate. The mucosa over the palate appeared ulcerated and erythematous in few areas. The swelling was firm in consistency, non-tender, non-compressible, non-reducible and non-fluctuant. Orthopantomogram (OPG) revealed the presence of a well-defined radiolucency in relation to the palate. Based on these findings, a provisional diagnosis of palatal cyst was provided. An incisional biopsy was performed and a diagnosis of pleomorphic adenoma was confirmed. The tumour was excised completely. Excisional biopsy revealed the presence of a parakeratinised stratified squamous surface epithelium with an underlying connective tissue (Fig.1) showing cuboidal cells arranged in a ductular pattern containing eosinophilic coagulum (Fig.2) and spindle shaped cells

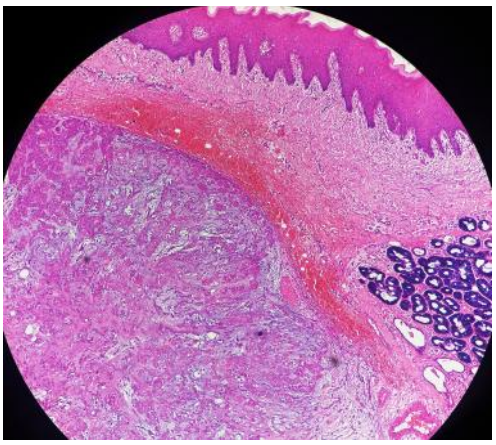

Fig 1 : Partially encapsulated tumor with associated salivary glands and overlying peripheral epithelium (H &E x 4X)

arranged in nests and sheets. Plasmacytoid cells with eccentrically placed nuclei (Fig.3) were also identified. Squamous cell islands, keratin pearls (Fig.5), areas of chondro-myxoid and hyalinised stroma (Fig.6), adipose tissue (Fig.4) and peripheral areas of mucous salivary glands were also seen. A few areas also showed the presence of giant cells (Fig.7). The tumor was partially encapsulated with a fibrous connective tissue (Fig.1). All

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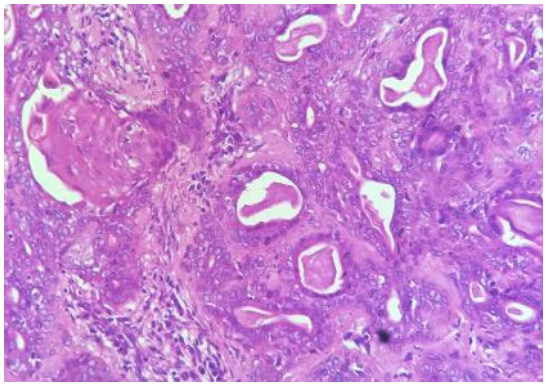

Fig 2 : Cuboidal cells arranged in ductular pattern with eosinophilic coagulum (H &E x 10X)

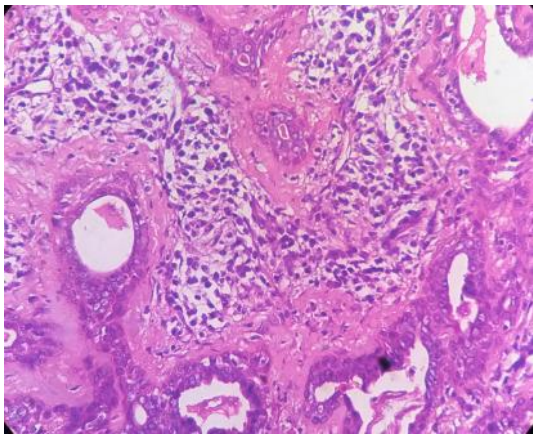

Fig 3 : Characteristic plasmacytoid cells in nests and sheets (H &E x 10X)

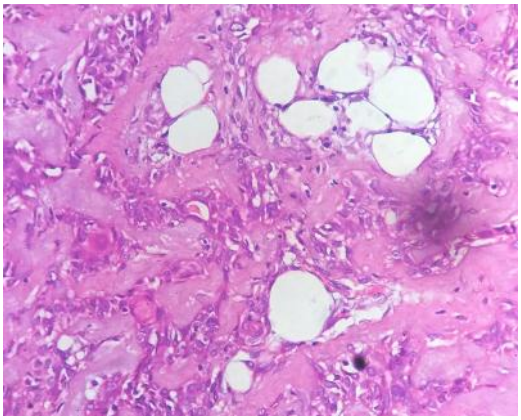

Fig 4 : Presence of adipose tissue (H &E x 10X)

these histopathological features were compatible with the diagnosis of Pleomorphic adenoma of the palate.

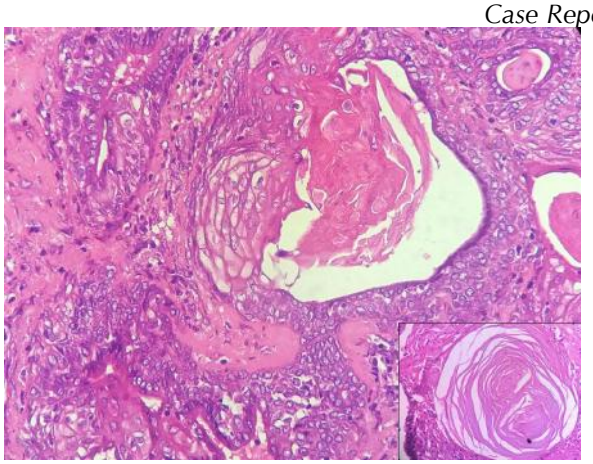

Fig 5 : Areas of squamous metaplasia along with keratin pearls(H &E x 10X)

Inset showing high power view of a keratin pearl

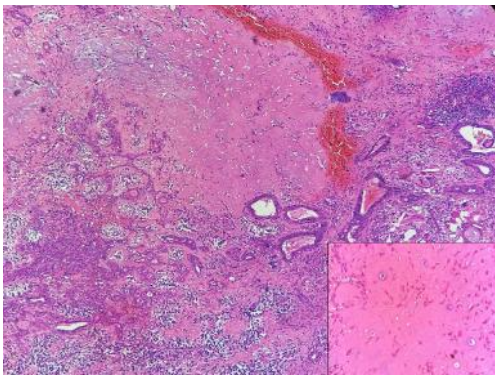

Fig 6: Hyalinised chondro-myxoid areas (H &E x 10X)

Inset showing high power view of chondroid area.

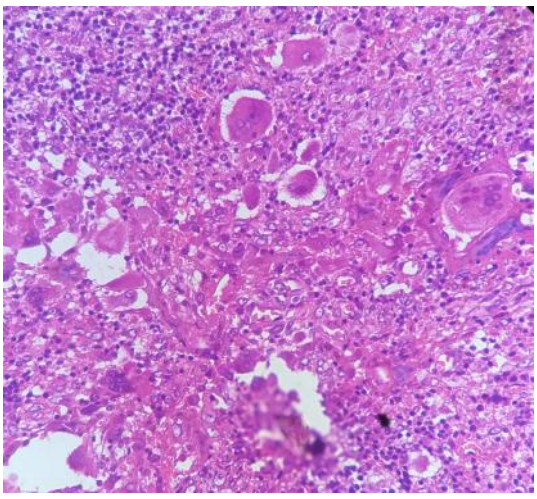

Fig 7: Few areas of giant cells (H &E x 10X)

DISCUSSION

The term Pleomorphic adenoma was suggested by Willis as it closely characterizes the histopathological appearance of this benign salivary gland tumour. Although it is occasionally referred to as 'Mixed Tumour', it is a misnomer as the tissues are not truly derived from more than one type of primary tissue.[4] The tumour cells undergo metaplasia and exhibit a complex histopathological picture consisting of fibrous, myxoid, chondroid, osseous and hyalinized areas. This can be explained by the reserve cell theory put forth by Batsakis and his associates who postulated that the intercalated duct reserve cell is the histogenetic precursor of Pleomorphic adenoma. Recent concepts related to the pathogenesis of this unique tumour revolve around both myoepithelial cells and the reserve cells of intercalated ducts. Studies suggest chromosomal aberrations in the

long arm of 8 and 12 and chromosomal translocations between chromosomes 3 and 8 thereby juxtaposing the PLAG 1 gene with the ß-catenin gene. This arrangement activates the ß-catenin pathway resulting in inappropriate division of cells.[5]

Pleomorphic Adenoma of minor salivary glands is most commonly seen in palate (42.63%), followed by lip (10%). The unusual sites are larynx epiglottis, sinuses and trachea. PAs have also been reported in tongue, soft palate, uvula and external auditory canal. It can sometimes be clinically challenging when presented with extensive involvement of oropharynx leading to airway obstruction.[3] Surface ulceration as was observed in our case is not very characteristic of Pleomorphic adenomas. Pleomorphic adenomas exhibit a female predominance with a female to male ratio of 1.9:1. In a study of 74 cases of palatal pleomorphic adenomas by Wu et al, a ratio of 2:1 with a marked female predilection was reported. In the same study it was correlated that the mean age of occurrence was 47 years and the peak age of incidence was 5th decade of life.[6] We have herewith reported a case in a 26 year old male patient. Histopathologically, presence of ducts or duct-like structures begets the tumour the name 'adenoma'. Foote and Frazell in their histological classification of Pleomorphic adenoma in the year 1954 have identified four major types: Principally myxoid, myxoid and cellular components in equal proportion, predominantly cellular and extremely cellular.[4] Principally myxoid type of Pleomorphic adenoma was found to be extremely rare in the palate according to a study by Wu et al.[6] Our case was representative of equal proportion of cellular and myxoid components. According to Krolls & Boyers, tumors with a prominent myxoid component tend to recur more.[7] Myoepithelial cells were seen as plasmacytoid cells and a considerable amount of squamous metaplasia in the form of keratin pearls was also observed. Keratin pearls, characteristic of well differentiated squamous cell carcinoma, occurs in pleomorphic adenoma due to metaplasia of the epithelial cells. This feature when extensively present and insufficiently sampled can be over-diagnosed as a malignancy. Presence of ulcerated palatal mucosa in our case was also misleading. This was avoided by adequate sampling of the specimen. In pleomorphic adenomas, fibrosis of the salivary gland tissue which gets compressed by the tumor elements results in formation of a pseudo/ false capsule.[8] Minor salivary gland pleomorphic adenomas are usually partially encapsulated as was seen in our case.

In recent years fine needle aspiration cytology guided by ultrasound is used for rapid diagnosis of lesions including Pleomorphic adenoma. Ultrasonography can help in distinguishing a benign from a malignant lesion in over 80% of cases. Fine needle aspiration cytology will aid in differentiating an inflammatory process from a neoplastic process. It can also help in distinguishing a primary tumour from a metastatic disease. Core needle biopsy of the lesion can also be performed which can help in yielding more tissue aiding histological typing of the lesion.[9] The treatment of choice for Pleomorphic adenoma is wide surgical resection of the affected gland along with a margin of normal tissue. Recurrence is rare although 6% recurrence rate is observed in cases of benign minor salivary gland adenomas.[10] This could be attributed to inadequate excision of the surgical margins of the tumour.

CONCLUSION

This case report brings out the need for the clinicians and histopathologists to be aware of the diverse morphological presentation of pleomorphic adenoma in a minor salivary gland thereby aiding in definitive diagnosis and treatment of the condition. This article also emphasizes the fact that a thorough histopathological sampling of a specimen can help in arriving at an accurate diagnosis.

REFERENCES

1. Dhir P, David CM, Dhaduti KG. Pleomorphic adenoma of the parotid gland with cystic degeneration: A rare case report. J Indian Acad Oral Med Radiol 2014;26:450-3.
2. S Moghe, AK Pillai, S Prabhu, S Nahar, UK Kartika. Pleomorphic adenoma of the palate: Report of a case. International Journal of Scientific Study 2014;2:54-56.
3. Sahoo NK, Rangan MN, Gadad RD. Pleomorphic adenoma palate: Major tumor in a minor gland. Annals of Maxillofacial Surgery 2013;3:195-7.
4. Neville BW, Damm DD, Allen CM, Chi AC, . Oral and maxillofacial pathology. 4th ed. Philadelphis: Elsevier 2015.
5. Mansur Rahnama, UrszulaOrzedala-Koszel, Lukasz Czupkallo, MichalLobacz. Pleomorphic adenoma of the palate:a case report and review of the literature. Contemp Oncol (Pozn) 2013;17:103-6.
6. Wu YC, Wang YP, Cheng SJ, Chen HM, Sun A, Chang JYF. Clinicopathological study of 74 palatal pleomorphic adenomas. Journal of the Formosan Medical Association 2016;115:25-30.
7. Krolls SO, Boyers RC. Mixed tumor of salivary glands. Long term follow-up.Cancer 1971;30:276-81.
8. Heeneman H. Parapharyngeal Space tumours. In: Kerr AG, editor. Scott Brown's Otolaryngology. 5. Butterworth & Co Ltd 1987;380-91.
9. Malathi N, Ravindran C, Thamizhchelvan H, Sharada T Rajan. Ultrasound guided fine needle aspiration cytology: A new diagnostic approach to pleomorphic adenoma. MIDAS Gazette 6:20-22.
10. Spiro RH: Salivary neoplasms: overview ofa 35-year experience with 2807 patients. Head Neck Surg 1986;8:177-84.