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Myoepitheliomatous Lesion Arising From a Pleomorphic Adenoma of Palate: A Rare and Unique Case Report

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ABSTRACT: **Background:**Pleomorphic adenoma is a well documented benign salivary gland beoplasm, which has been known by a variety of names where the cells' ability to differentiate into ductal and non-ductal epithelial cells as well as mesenchymal chondroid, osseous and myxoid cells.

Case presentation: A 39 year old female patient reported with a chief complaint of a painless swelling on the right palate region.Intra-oral examination revealed a painless hard swelling in relation to teeth 23 to 27. firm, exophytic, sessile lesion with fixation to the underlying tissues was seen, measuring approximately 3.5 X 5 cm, ovoid in shape with a smooth surface and no indurated margins. No associated symptoms were elicitated. Incisional biopsy was performed and the tissue was sent for histopathological assessment. neoplastic cells arranged in ductal pattern of nests, sheets and cords with a delicate collagenous stroma.

Conclusion: The presence of both an appreciable stroma, chondromatous and myxomatous features alongside spindleoid and plasmacytoid variants of myoepithelial cells thus indicates the uniquity and rarity of the case. The documentation of this case will in future aid pathologists to avoid diagnostic pitfalls as well as help in better understanding of the tumor histiogenesis

KEYWORDS: Salivary gland tumors, pleomorphic adenoma, mixed tumor, benign mixed tumor.

I. INTRODUCTION

Pleomorphic adenoma is a well documented benign salivary gland neoplasm, which has been known by a variety of names; mixed tumor, enclavoma, branchioma, endothelioma, enchondroma, mixed tumor and benign mixed tumor. The term pleomorphic adenoma was coined by Willis characterizing the histologic nature of the lesion. The cells' ability to differentiate into ductal and non-ductal epithelial cells as well as mesenchymal chondroid, osseous and myxoid cells. Although initially named mixed tumor, in light of a truly teratomatous lesion, as being derived from multiple primary tissues; the pleomorphic differentiation of cells is accredited to the metaplastic properties exhibited by the tumor cells per se.[1]

Pleomorphic adenoma is the commonest salivary gland tumor accounting for almost 57.7% of all reported salivary neoplasms. Although several theories have been put forward in light of tumor histiogenesis, the exact mechanism is still not clearly understood. Regezi and Batsakis postulated the property of intercalated ductal reserve cells to differentiate into myoepithelial cells and ductal cells, the former in turn undergoing mesenchymal metaplasia, showing inherent smooth muscle like properties. On the other hand, Hubner et al. proposed that the myoepithelial cell claims responsibility for the morphologic diversity of the cells including the production of fibrous, mucinous, chondroid, myxoid and osseous areas. Both myoepithelial and ductal reserve cells are seen in the lesion, as confirmed by ultramicroscopic studies.[2][3]

II. CASE HISTORY

A 39 year old female patient reported with a chief complaint of a painless swelling on the left palate region. The lesion developed approximately 2 years ago, with a small nodular swelling on the palate which gradually increased in size. The lesion although initially mobile later got fixated, with no associated symptoms. The medical, dental and personal histories along with familial history did not elicitate any such relevant information.

III. CLINICAL FEATURES

General physical examination, along with a detailed local intra-oral examinations were performed. Local examination was done both extra-oral and intra oral. The extra-oral examination revealed no significant findings and no other abnormalities were detected in light of lips and temporomandibular joint. No palpable lymph nodes were elicitated. The mouth opening was also normal, recorded at 38 mm.

Intra-oral examination was done for both hard and soft tissues, which revealed a painless hard swelling in relation to teeth 23 to 26. The intra-oral hard tissue

showed no deviations from normalcy, with no missing teeth, stains, calculi, mobility or non-carious destruction. On palpation a firm, exophytic, sessile lesion with fixation to the underlying tissues was seen, measuring approximately 3.5 X 5 cm, ovoid in shape with a smooth surface and no indurated margins. No associated symptoms were elicitated. A provisional diagnosis of pleomorphic adenoma of the palate was given. Incisional biopsy was performed and the tissue was sent for histopathological assessment.

IV. RADIOGRAPHIC FEATURES

Pleomorphic adenoma is a purely soft tissue lesion. No radiographic features have ever been documented, despite the often presence of osseous and chondroid components.[1][2]
No radiographic findings were seen in this case.

V. HISTOPATHOLOGY

Histopathology revealed typical features of pleomorphic adenomas in minor salivary glands[1]; neoplastic cells arranged in ductal pattern of nests, sheets and cords with a delicate collagenous stroma, showing myxoid features. Areas of cystic degeneration consisting of an eosinophilic coagulum within them are seen. A notable finding is the presence of spindleoid and plasmacytoid myoepithelial cells, round to ovoid in shape, present in clusters. Vacuolar degeneration is seen in a few cells. Myoepithelial cell-containing areas are appreciable in both high and low magnifications.

VI. CASE SUMMARY

A 39 year old female patient reported with a chief complaint of a painless swelling on the right palate region.Intra-oral examination revealed a painless hard swelling in relation to teeth 23 to 26. firm, exophytic, sessile lesion with fixation to the underlying tissues was seen, measuring approximately 3.5 X 5 cm, ovoid in shape with a smooth surface and no indurated margins. No associated symptoms were elicitated. Incisional biopsy was performed and the tissue was sent for histopathological assessment. neoplastic cells arranged in ductal pattern of nests, sheets and cords with a delicate collagenous stroma, showing myxoid features, cystic degenration containing eosinophilic coagulum, vacuolar degeneration, myoepithelial cells and plasmacytoid cells.

VII. DISCUSSION

Although ductal and non-ductal neoplastic cells, along with spindleoid myoepithelial cells are seen, a striking feature of the lesion is the presence of plasmacytoid type of cells, round to ovoid in shape, with eccentrically placed nuclei, present in clusters. These cells are believed to be modified myoepithelial cells present within a chondromyxoid stroma, with delicate collagenous arrangement.[1][2][5]

VIII. TREATMENT AND PROGNOSIS

Conservative surgical excision is the best line of treatment.[1][2] Radiotherapy and Chemotherapy may prove effective in reduction of tumor size before excision. Regular follow ups are advisable.[2]

The overall prognosis as concluded by several authors is found to be excellent in minor salivary glands, and recurrences are rare. Malignant transformation to carcinoma ex pleomorphic adenoma, although a rarity, may result in a poorer prognosis, however, no CXPA features were seen in any sections provided by incisional biopsy.[3][5][6][7][8][9][14]

IX. CONCLUSION

Pleomorphic adenoma is a well documented lesion, being the commonest salivary gland, as well as parotid gland tumor, accounting for nearly 58% of all benign salivary gland lesions. The presence of both spindleoid and plasmacytoid type of myoepithelial cells in a delicate stroma is also notable, as multiple variants of myoepithelial cells are usually seen in a more cellular variant of this tumor, as well as in myoepitheliomas, with scant stroma. The presence of both an appreciable stroma, chondromatous and myxomatous features alongside spindleoid and plasmacytoid variants of myoepithelial cells thus indicates the uniquity and rarity of the case, where pleomorphic adenoma gives rise to a myoepithelioma like lesion. The documentation of this case will in future aid pathologists to avoid diagnostic pitfalls as well as help in better understanding of the tumor histiogenesis in light of genetic, epigenetic and post-translational modification of proteins translated from the micro-RNAs exhibited at various loci.[4][10][11][12][13][15]

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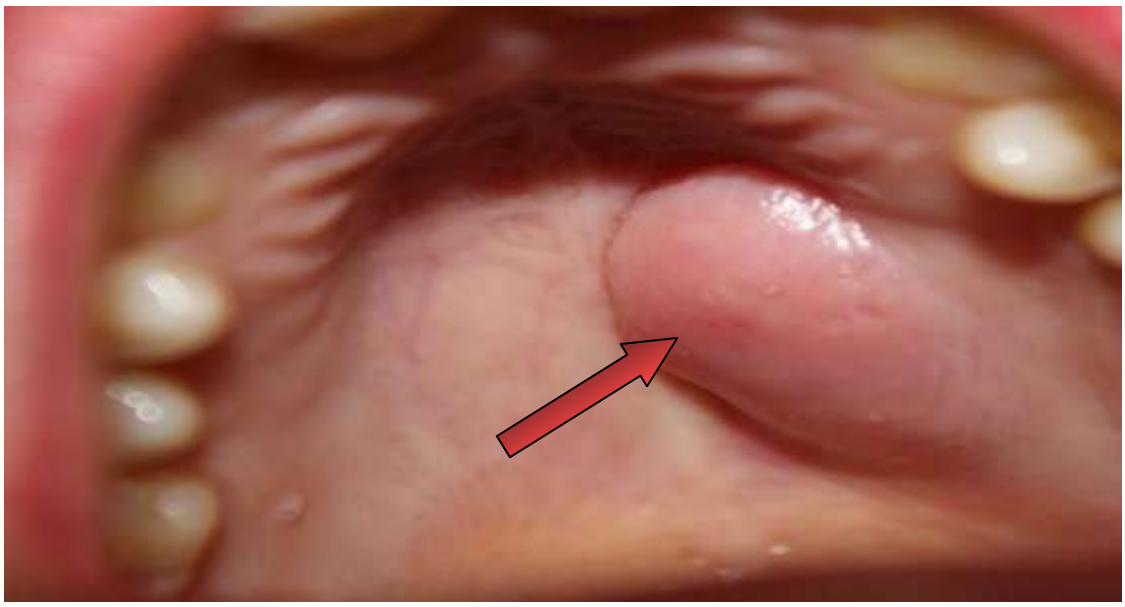

IMG-1: INTRA ORAL PICTURE SHOWING SITE, SIZE AND EXTENT OF THE LESION

IMG-2: PANORAMIC RADIOGRAPH REVEALING NO SIGNIFICANT FINDINGS

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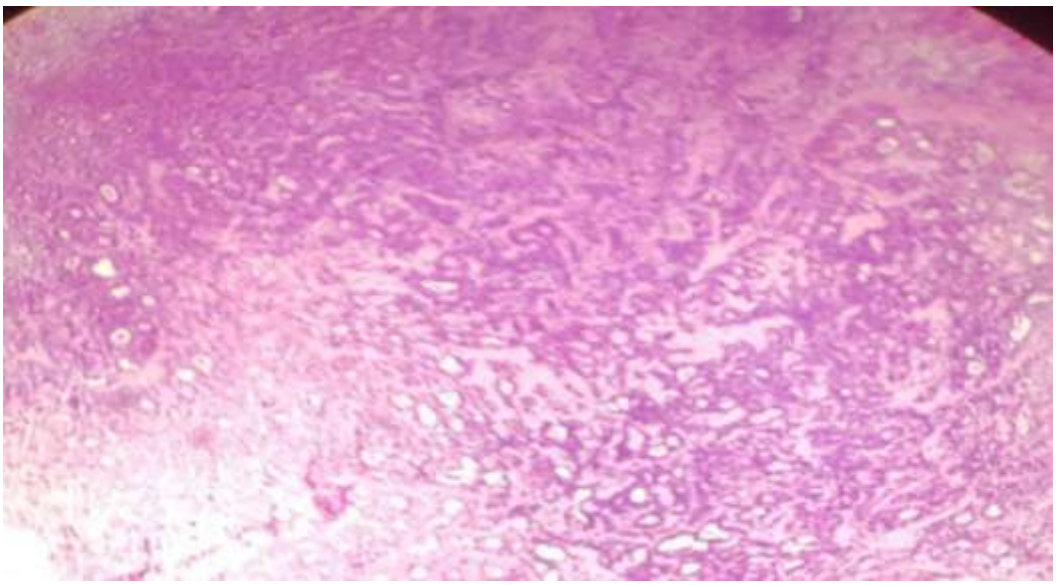
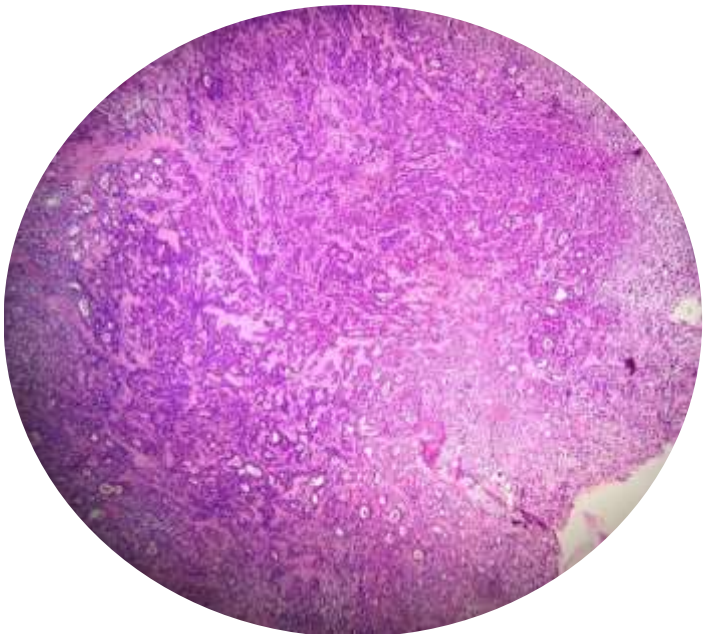


IMG-3: H & E SECTION SHOWING SCANNER VIEW (4X)



(A)

IMG-4: (A) H & E SECTION SHOWING A DELICATE COLLAGENOUS STROMA, DUCTAL CELLS IN NESTS, MYOEPITHELIAL CELLS (SPINDLE AND PLASMACYTOID) (10X)

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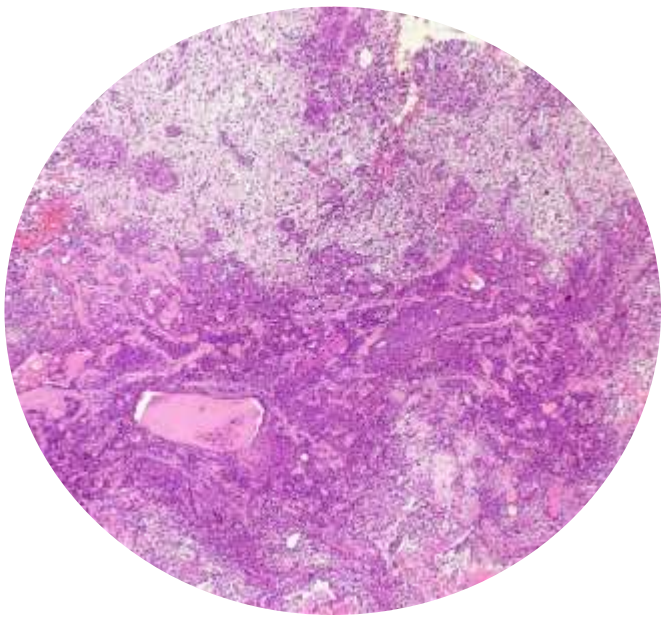
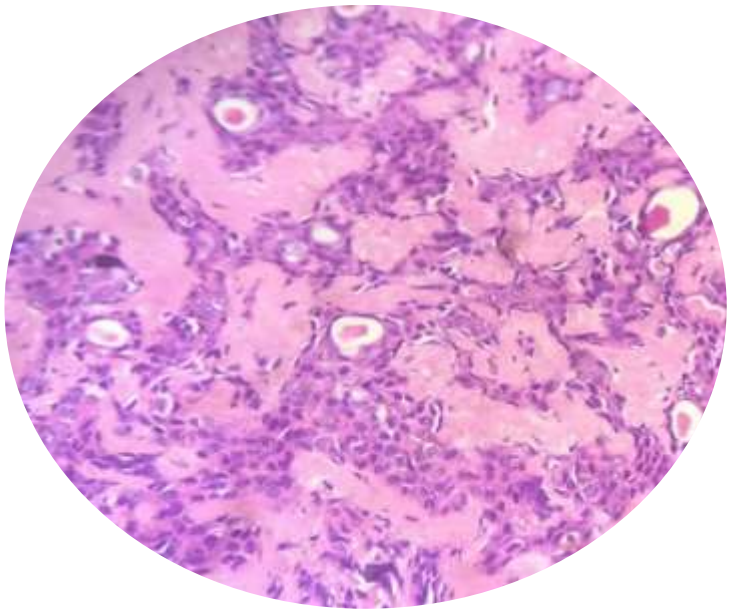


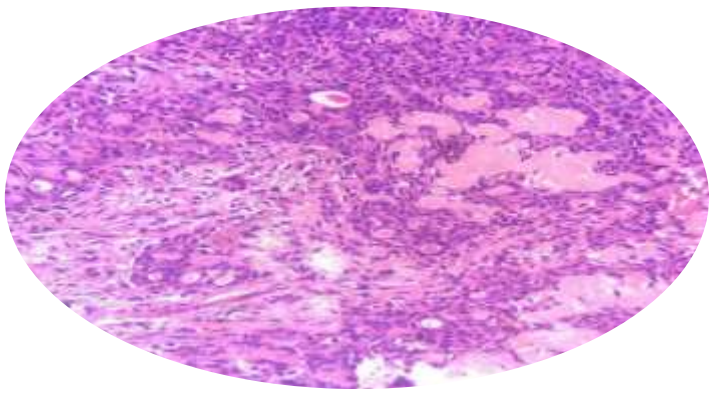
(B)

IMG-4: (B) CYSTIC DEGENERATION WITH EOSINOPHILIC COAGULUM AND FEW VACUOLAR DEGENERATION WITH CHONDROID AND MYXOID APPEARANCES. (10X)



(A)

IMG-5: (A) SPINDLEOID AND PLASMACYTOID MYOEPITHELIAL CELLS (40X)



(B)

IMG-4: (B) EOSINOPHILIC COAGULUM IN AREAS OF CYSTIC DEGENERATION(40X)

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