Keratinizing Pleomorphic Adenoma in Minor Salivary Gland: A Case Report

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INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign mixed tumor of the salivary glands characterized by broad variation in the stromal and parenchymal differentiation by tumor cells.[1] This neoplasm makes up to 73% of all salivary gland tumors. Parotid gland is the most commonly involved site. Globally, 14.0% to 52.0% of salivary gland tumors affect minor salivary glands and out of them up to 67.0% are benign.[2] PA affects females (60%) more frequently than men and (40%) present in the fourth, fifth, and sixth decades of life.[3] Clinically, it presents as an asymptomatic, slow growing swelling.[4] The optimal treatment for pleomorphic adenoma is comprehensive excision with safety margins and regular follow up for at least 3–4 years because of the high risk of recurrence.[5]

CASE REPORT

A 45 years old male reported with slow growing, painless swelling on lip that increased in size gradually over a period of 3 years. Clinically it was seen as well-defined, lobulted, swelling measuring 2.0 cm × 1.5 cm × 1.0 cm in size while the overlying surface was slightly ulcerated (Fig. 1). Patient underwent cheiloplasty 1.5 years back. There was no regional lymphadenopathy.

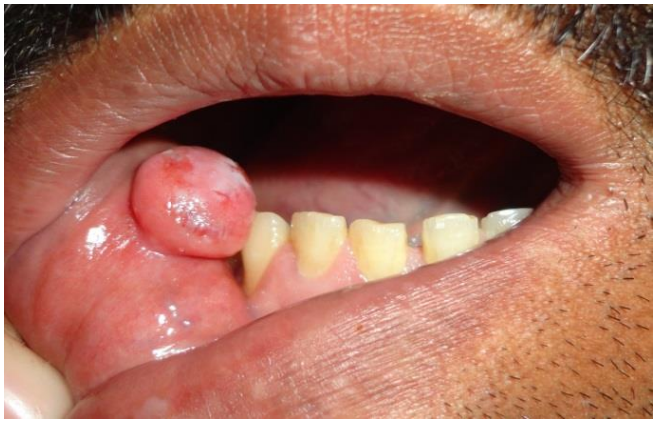

Fig. 1: *Well-defined, lobulated swelling on labial mucosa right side.*

Depending upon the clinical features, a provisional diagnosis of irritational fibroma was made while PA and lipoma were considered as differential diagnosis. The mass was excised under general anesthesia and sent for histopathological examination.

Grossly it was a nodular tissue piece measuring 1.6 cm × 1.0cm × 0.8 in size. Serial slicing revealed whitish cut surface with hemorrhagic areas (Fig. 2). Multiple representative sections were taken and submitted for processing in two cassettes.

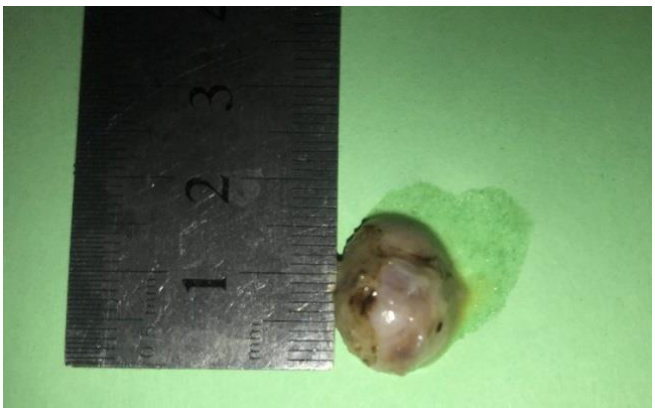

Fig. 2: *A nodular tissue with hemorrhagic areas.*

The histopathological examination of the Hematoxylin and Eosin stained sections revealed benign mixed neoplasm of salivary glands. There was a lack of capsule. Ducts and tubules were scattered in the chondromyxoid background. Multiple keratinous cysts lined by stratified squamous epithelium and containing lamellated keratin were seen. Foci of squamous metaplasia were also evident. These cysts were of varying sizes (Fig. 3). A diagnosis of keratinizing pleomorphic adenoma with squamous metaplasia was made.

DISCUSSION

Pleomorphic adenoma with squamous metaplasia is not common thus it creates a difficulty in making the histopathological diagnosis.[6] Microscopically, squamous metaplasia observed in benign and

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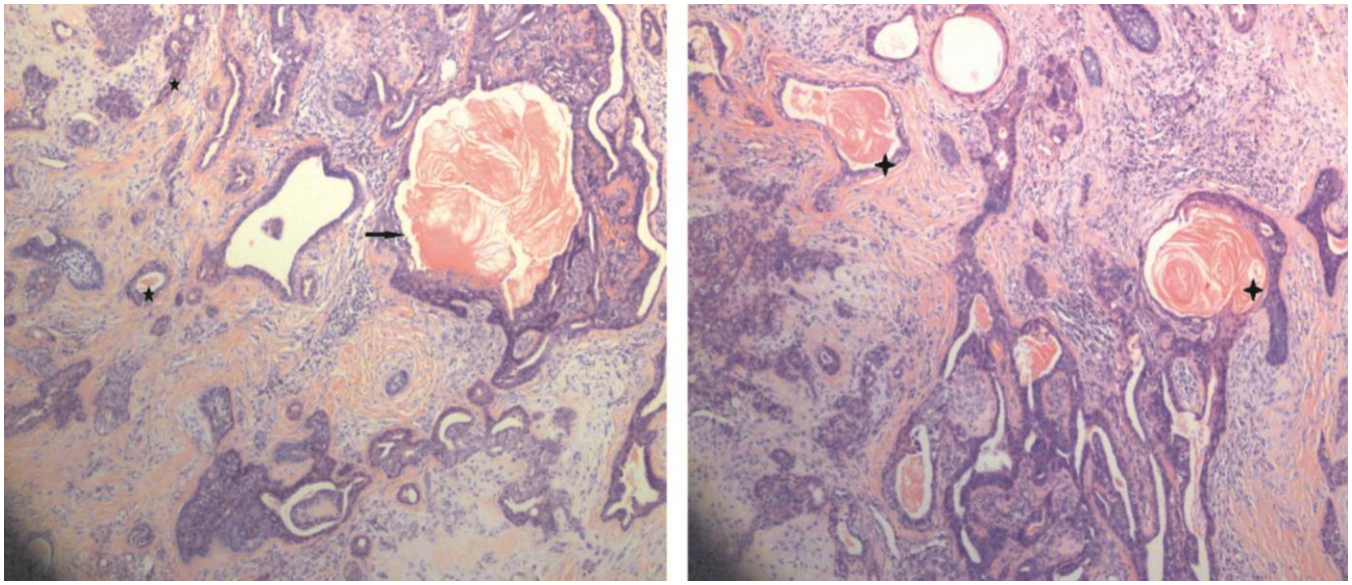

Fig. 3: *Photomicrograph of H& E stained tissue showing well circumscribed lesion with multiple cystic spaces containing lamellated keratin (✦) and lined by squamous epithelium (→).*

malignant tumors is supposed to be related with altered tumor environment caused by trauma, ischemia and repair following infarction. The most likely reason for squamous metaplasia in the salivary glands seems to be ischemia as it may cause the rapid and easy shift of cytokeratin filaments genetic programming.[7] Also,loss of differentiation, successive hyperplasia in cells of acini, duct lumina, myoepithelium and ligation of salivary gland parenchymal artery may lead to metaplastic changes in pleomorphic adenoma.[8] It has also been proposed that minor salivary gland ductal epithelium can be affected by exposure to different irritants and may play a significant role in formation of keratin pearls.[8]In present case, there was a history of trauma as patient underwent cheiloplasty, 1.5 years back.

Excessive squamous metaplasia particularly in the absence of chondromyxoid stroma can lead to misdiagnosis such as choristoma or keratocystoma, mucoepidermoid carcinoma and squamous cell carcinoma, especially on fine-needle aspiration and incisional biopsy, because of limited sampling. So it is important to discuss the differentials to make a definite diagnosis because of its uncommon presentation.[9]Mucoepidermoid carcinoma is usually multicystic with infiltrating growth pattern and also the keratinization is not common.[10] PA can be differentiated from squamous cell carcinoma based on the absence of cellular atypical changes, metastasis, necrosis, invasion and low proliferation. PA is also difficult sometimes to differentiate from choristoma however the presence of glandular structures in pleomorphic adenoma can be helpful to reach conclusive diagnosis.[11] Different studies report that immunohistochemical analysis can help in making definite diagnosis. As squamous epithelium lining the keratin cyst shows strong positivity for high molecular weight cytokeratin while comparing to low molecular weight cytokeratin and p63. These histological similar squamoid cells are actually metaplastic squamous and ductal epithelial cells thus explaining the pattern of cytokeratin expression from strong to weak.[12]Also the positive immune expression of high molecular weight cytokeratin, P63, S-100, vimentin, smooth muscle actin, muscle specific actin and glial fibrillary acidic protein in myoepithelial and albuminal cells has been reported that can aid in diagnosis.[8] The benign nature of tumor is characterized by squamous epithelium without atypia, absence of tumor necrosis, metastasis, low cellular proliferation and considerably slow growth of the tumor. The treatment of choice for these lesions is usually wide local excision.[8]Similarly surgical excision was done in the present case and patient was asked for follow-up after 6 months.

CONCLUSION

It is hereby concluded that pleomorphic adenoma with metaplastic changes is challenging to diagnose and can lead to misdiagnosis in case of extensive metaplasia. Keratinizing PA in lip area is seldom reported. Hence a meticulous approach should be adopted by histopathologists to reach an accurate and conclusive diagnosis for optimal management of such cases.

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AUTHOR'S CONTRIBUTION

RA: Conception of work, drafting the manuscript
MRS: Acquisition and interpretation of data
TMT: Acquisition of data
MJ: Drafting, contribution to intellectual content
KF: Drafting the manuscript, revising critically
MM: Drafting the manuscript, revising critically
NN: Conception of work, interpretation of data, final approval of the manuscript

CONFLICT OF INTEREST

None to declare.

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None to disclose.

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