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Perplexity in Diagnosing Pleomorphic Adenoma of Minor Salivary Gland with Plasmacytoid Cell

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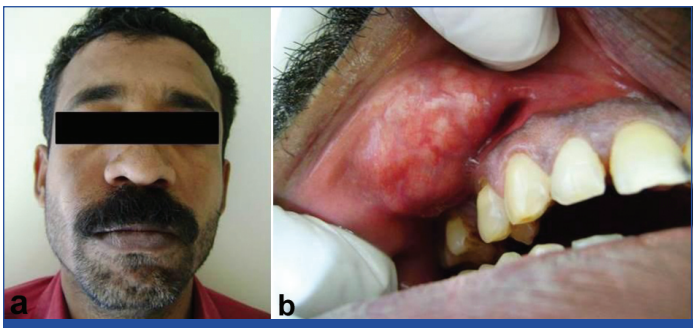
A 46-year-old male reported to the Outpatient Department with the chief complaint of slow growing swelling in relation to upper front tooth region since one year. The swelling was gradual in onset with no history of pain, paresthesia or discharge. His personal history revealed that he was a cigarette smoker (4-5 times per day for 8 years), left his habit since a month. Medical history and family history were non contributory. The patient was moderately built with all vitals within normal limits.

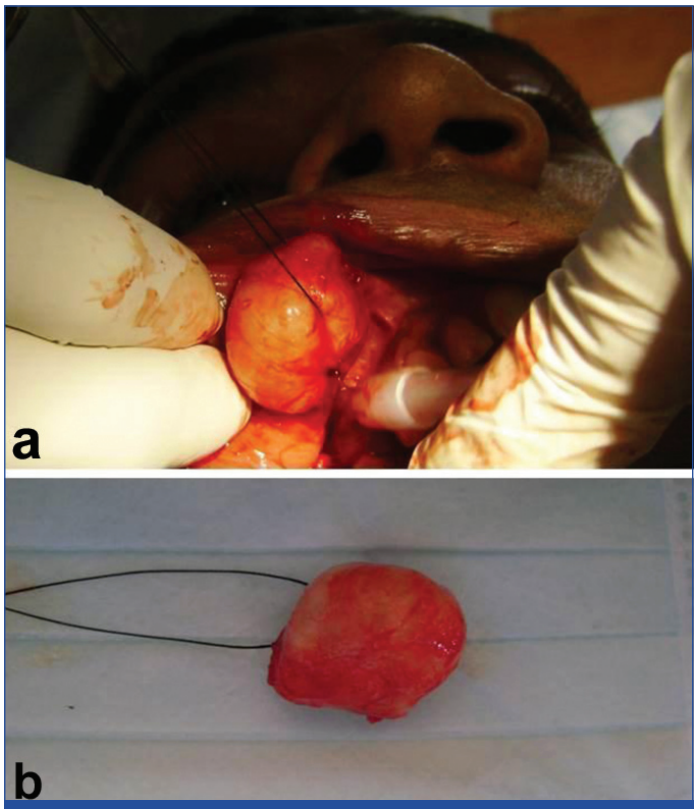
Extraoral examination showed the presence of diffuse, mobile, non tender swelling surrounded within the boundary of right nasolabial fold superiorly, vermillion border of upper lip inferiorly, right commissural area laterally and philtrum mesially. There was no regional lymphadenopathy noted and overlying skin was free with no evident change in colour and texture. Intraorally, well defined, single, firm, non tendered, non ulcerated swelling was noted in relation to 12, 13 and 14 resulting in the obliteration of the right maxillary labial vestibule. The swelling was dome shaped measuring 1.5×2 cm showing pale pink superficial mucosa with prominent vascular markings [Table/Fig-1].



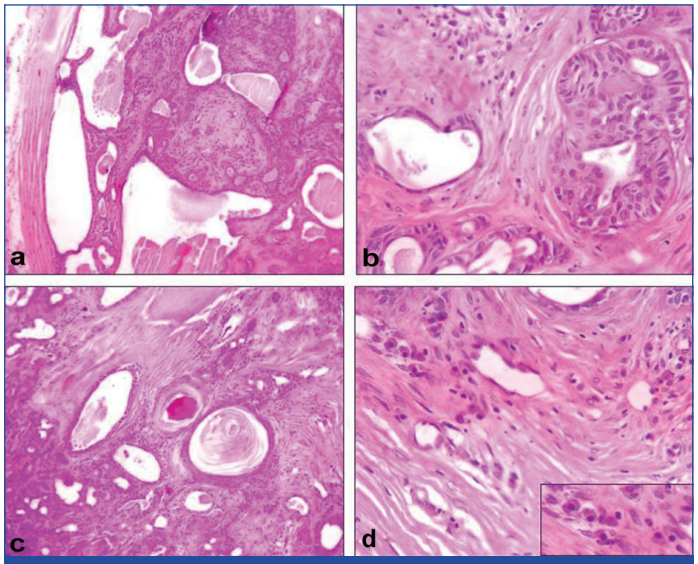
[Table/Fig-1]: (a) Extraoral photograph showing diffuse swelling lateral to the nose above the right upper lip region; (b) Intraoral photograph showing large non ulcerated lesion with obliteration of labial vestibule.

Keeping the clinical features in view, the provisional diagnosis of benign salivary gland neoplasm was made. Differential diagnosis including fibroma, lipoma, canalicular adenoma, Pleomorphic Adenoma (PA) and abscess were considered. The excisional biopsy was performed under local anaesthesia after obtaining informed consent from the patient. The gross specimen comprised of an encapsulated soft tissue mass measuring 1.5×2×1.5 cm with rubbery consistency [Table/Fig-2].

The tissue sections were stained with Haematoxylin and Eosin (H&E) for microscopic evaluation showed the presence of thick capsule. Neoplastic glandular epithelial cells were arranged in the form of sheets with numerous ductal structures containing eosinophilic coagulum. The lumen of the ducts was lined by cuboidal cells surrounded by abluminal myoepithelial cells. Wide spread squamous metaplasia was observed with multiple cystic spaces filled with whorls of keratin. Few areas showed presence of plasmacytoid cells with eccentric nuclei and eosinophilic hyalinised cytoplasm [Table/Fig-3]. These microscopic features were suggestive of PA



[Table/Fig-2]: (a) Intraoral photograph showing complete surgical excision of the nodular mass, (b) Encapsulated gross specimen measuring 1.5×2.0×1.5 cm.



[Table/Fig-3]: (a) Histopathology showing glandular epithelial cells surrounded by thick fibrous capsule with numerous duct like spaces filled with eosinophilic coagulum (40X); (b) Ducts lined by luminal cuboidal cells and abluminal myoepithelial cells with scattered plasmacytoid cells (100X); (c) Squamous metaplasia with whorls of keratin in the cystic spaces. (10X); (d) Plasmacytoid cell in hyalinised eosinophilic stroma (40X), Inset: Round cell with eccentric nucleus (H&E stain).

with extensive squamous metaplasia. The patient is under follow-up since two years and no recurrence is observed till date.

Pleomorphic Adenoma (PA) is the most common benign salivary gland tumour that rarely involves minor salivary glands. The term PA was first described by Willis in 1953 to address diverse clinical and histological appearance of the tumour. The onset is usually reported in third to fourth decade with twice common female preponderance compared to the males. Total 50-60% cases are usually seen in the palate followed by upper lip (15-20%), buccal mucosa (8-10%) and tongue with few sporadic cases reported in maxillary sinus and pterygopalatine space. Benign tumours of salivary glands are six times more common in upper lip while malignant tumours have more frequency of occurrence on lower lip. Clinically, small lesions of the upper lip are asymptomatic, sessile, mobile rubbery mass with smooth surface. However, larger tumours are firm with bosselated appearance that may cause noticeable swelling with asymmetry of face [1-3]. In the present case, PA affecting upper lip was found in male patient in 4th decade with single, ovoid firm swelling with superficial vascular marking.

Pleomorphic adenoma of minor salivary gland may present as well circumscribed, encapsulated mass but incomplete pseudocapsule or pseudopodia and extracapsular extensions are not rare in tumours with expansive growth. The acinar, ductal and myoepithelial cells are often arranged in ductal and cystic pattern undergoing aberrant metaplastic changes. The most classical feature is the presence of admixture of polygonal epithelial and spindle-shaped myoepithelial elements in a mucoid, myxoid, cartilaginous, or hyaline background stroma. Few plasmacytoid cells with eosinophilic cytoplasm and eccentrically placed nuclei may be present. Though, these cells are ectodermal in origin, yet smooth muscle expression is observed in the desmosomes, intermediate size filaments, endocytic vesicles, and microfilaments of myoepithelial cells [4].

The histopathology of present case showed the metaplastic squamous epithelium-lined cystic spaces containing numerous keratotic lamellae and some solid squamous cell islands with keratin whorls. Literature suggests that only 25% of PA may manifest squamous metaplasia posing considerable challenge in diagnosing such lesions. Ischaemia may induce epithelial morphogenesis in myoepithelial cells, thereby producing increased amounts of tonofilaments and desmosomes with loss of myofilaments at acinar-intercalated duct cell complex [4,5]. Other neoplasms showing squamous differentiation include mucoepidermoid carcinoma, adenosquamous cell carcinoma, keratocystoma and conventional squamous cell carcinoma. However, absence of cellular and nuclear atypia, lack of invasion, minimal cellular activity with presence of intact fibrous capsule eliminates the diagnosis of malignancy [6].

The exact pathogenesis behind PA still remains controversial and has created a way for the researchers to intrude into the molecular level of genes. The complex process of morphogenesis of salivary gland is tightly regulated by growth molecules and transcription factors. The presence of chromosomal abnormalities due to translocation t(9;13)(p13;q12),) or reciprocal translocation t(9:12)(p13-21;q13-15) related to PLAG1 are regarded as major responsible event that may cause homeostatic errors. Therefore, loss or alteration of these factors increases the probability of progression to malignant PA [2].

The treatment modality of PA arising from minor salivary glands was complete surgical excision with clear margins followed by uneventful healing.

CONCLUSION(S)

The salivary glands may show a diverse range of lesions involving spectrum of cells affecting its overall biological presentation. It also mandates the inclusion of PA in differential diagnosis of swelling involving upper lip region. The heterogeneity of tissue in PA and potential neoplastic transformation demands meticulous approach in differentiating it from malignant lesions. The present case revealed cystic spaces resembling cribriform pattern with areas showing metaplastic squamous cells and plasmacytoid cells. The conspicuous nature of myoepithelial cells to mimic plasmacytoid cells or squamous cells accentuates the risk of misdiagnosis. This type of lesions demands early diagnosis with complete excision for optimal management along with regular follow-up.

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