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LONG-STANDING PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLANDS: A CASE STUDY WITH UNUSUAL HISTOLOGICAL FEATURES

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ABSTRACT

Pleomorphic adenoma, the most common benign salivary gland neoplasm, presents a unique case when located in the hard palate, a rare site for this tumor. We report a case of a 34-year-old male with a 13-year history of a slowly growing, asymptomatic swelling in the right hard palate. Clinical examination revealed a 5x4 cm, non-fluctuant, non-pulsatile, firm mass extending from the rugae to the soft palate, crossing the midline. Radiographic imaging showed no bony erosion. Fine-needle aspiration cytology suggested pleomorphic adenoma, which was confirmed by histopathological analysis after total excision. The tumor was well-encapsulated, composed of epithelial and myoepithelial elements in a myxoid stromal background. Numerous duct-like structures with eosinophilic secretions were identified. Notably, the tumor exhibited prominent squamous metaplasia with keratin pearl formation and extensive keratin cyst formation, which are rare histological features that can mimic malignancy. The absence of cytological atypia, abnormal mitoses, and invasive patterns confirmed the benign nature of the lesion. This case highlights the importance of considering pleomorphic adenoma in the differential diagnosis of palatal swellings and emphasizes the role of comprehensive clinical, radiographic, cytological, and histopathological evaluation in accurately diagnosing and managing these tumors, particularly when unusual histological features are present.

KEYWORDS: Pleomorphic Adenoma, Salivary Gland tumour, Mixed tumour, Squamous Metaplasia, Hard Palate.

INTRODUCTION

Pleomorphic adenoma is the most common benign salivary gland neoplasm, accounting for 45–75% of all salivary gland tumours.[1,2] The parotid gland is the primary site for pleomorphic adenomas; but can also occur in submandibular and minor salivary glands.[3,4] Pleomorphic adenomas have a unique histology, containing both epithelial and myoepithelial cells, as well as differentiated stroma that can be mucoid, myxoid, or chondroid.[5] These tumors typically present as hard, slowly expanding masses with clear borders. Proper management of pleomorphic adenomas is crucial due to their potential for malignant transformation if left untreated.[6]

Pleomorphic adenomas are more common in females, particularly between 30 and 60 years of age.[3] In most cases, it manifests as a solitary, painless mass that moves around and grows over time, with little to no impact on facial nerve function.[7] Histological analysis often reveals a fibrous capsule enclosing a chondromyxoid matrix including both epithelium and myoepithelial cells.[6,7] Pleomorphic adenomas have the potential to grow in size and recur if not removed completely, or may potentially transform into carcinoma ex pleomorphic adenoma in extremely unusual instances.[8]

While pleomorphic adenomas are typically found in the parotid gland, their presence in minor salivary glands is a less common occurrence. Among these rarer cases, the palate stands out as the most frequently affected intraoral site. Consequently, the presented case of a pleomorphic adenoma located in the hard palate of a 34-year-old male carries significant clinical importance.

CASE REPORT

A 34-year-old male patient reported to the dental out-patient department with a chief complaint of swelling in the hard palate that had persisted for the past 13 years. The swelling had progressively increased in size, asymptomatic, with neither aggravating nor relieving factors. On intra-oral examination the mouth opening was normal, a single solitary swelling of 5x4 cm in size was noted on the right side of the hard palate extending anteriorly upto the rugae and posteriorly upto soft palate, medially crossing the midline and laterally 5 mm from attached gingiva [Figure 1].

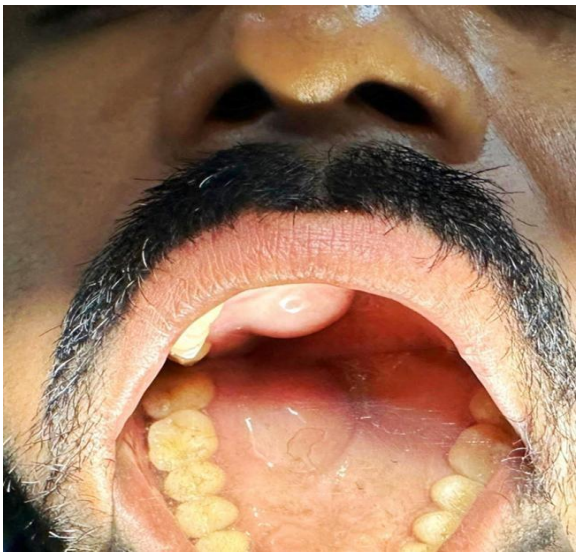

Figure 1: Reveals a single solitary swelling of 5x4 cm in size was noted on the right side of the hard palate.

The swelling was oval in shape with no discharge and the overlying mucosa appeared normal. On palpation, the swelling was non-fluctuant, non-pulsatile and firm in consistency. The patient has no other systemic illness. A panoramic radiography showed no bony erosion. Fine-needle aspiration cytology (FNAC) was carried out, which showed a blood mixed material. The smears were cellular with clusters of ductal cells in a background of myxoid ground substance. Correlating with the clinical, radiographical and FNAC findings, a provisional diagnosis of pleomorphic adenoma was made. The lesion was totally excised and sent for histopathological evaluation. Histopathological evaluation revealed a well-encapsulated tumour, composed of epithelial and myoepithelial elements in a variable stromal background which was myxoid [Figure 2]. Numerous duct-like structures were identified and some of them showed eosinophilic secretion within the lumen [Figure 3]. The present case of ours showed prominent squamous metaplasia with keratin pearl formation in many areas [Figure 4]. Extensive keratin cyst formation was seen with concentrated lamellated keratin debris [Figure 5]. The post-operative for the patient was uneventful.

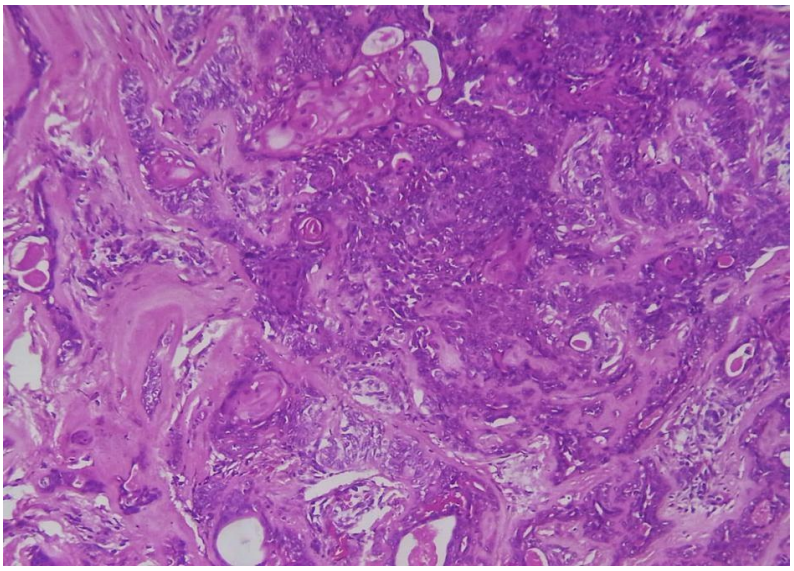

Figure 2: Shows presence of Ductal and Myoepithelial cells in a myxoid stroma with eosinophilic secretions.

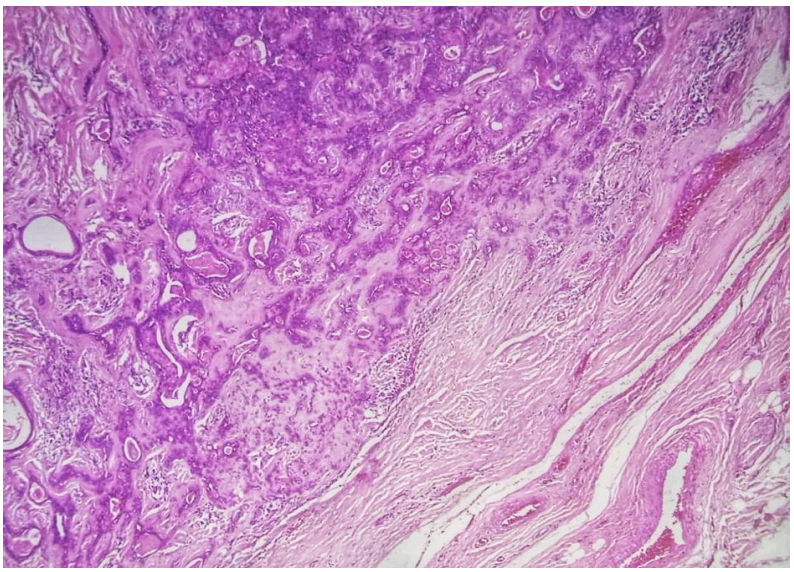

Figure 3: Illustrates a well-encapsulated tumour characterised by proliferation of ductal cells.

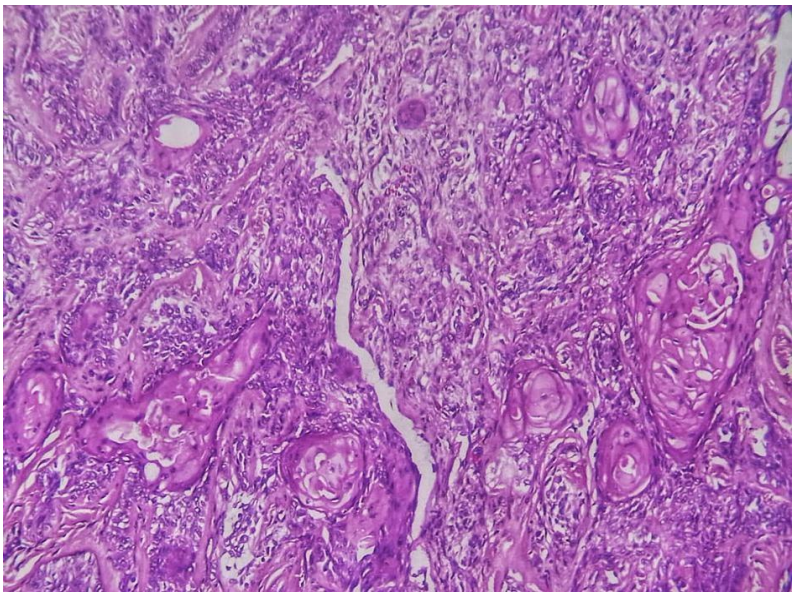

Figure 4: Shows ductal cells demonstrating the evidence of squamous metaplasia.

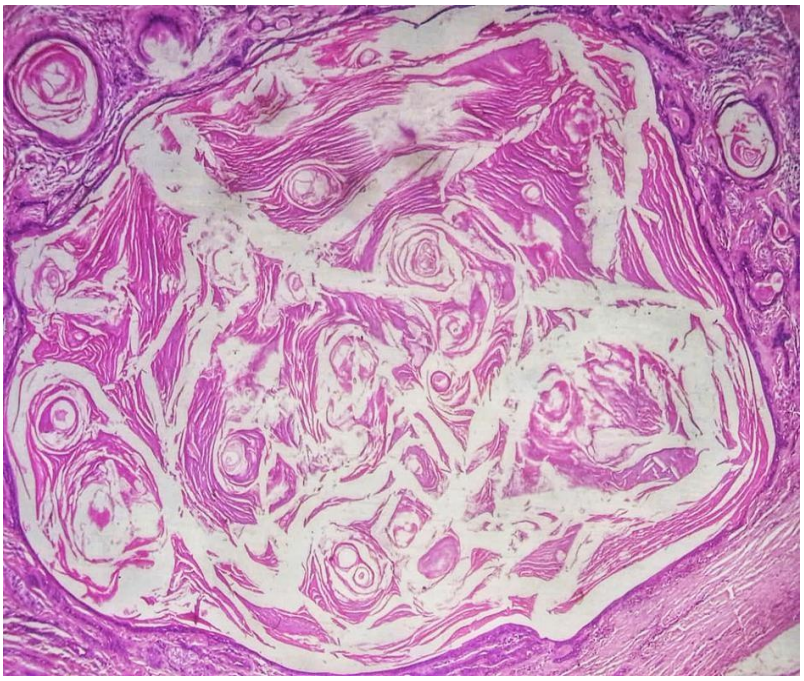

Figure 5: Reveals keratin cyst formation was seen with concentrated lamellated keratin debris.

DISCUSSION

Pleomorphic adenoma, often dubbed the 'chameleon' of salivary gland tumors due to its diverse histological patterns, continues to present diagnostic challenges despite its benign nature. The hard palate is one of the most common sites for pleomorphic adenoma arising from minor salivary glands.[9,10] In the present case, a 34-year-old male presented with a long-standing palatal swelling, which is consistent with the slow-growing nature of this tumour. However, the differential diagnosis of other minor salivary gland tumors—including mucoepidermoid carcinoma, basal cell adenoma, and polymorphous adenocarcinoma—must always be considered due to overlapping clinical and radiographic features.[11,12]

Clinically, pleomorphic adenoma typically manifests as firm, painless, non-ulcerated swelling.[13] The 13-year gradual enlargement of the lesion in our patient strongly suggested a benign nature, as malignant tumors usually demonstrate rapid, destructive growth and may cause ulceration or nerve involvement.

Histopathologically, pleomorphic adenomas are characterized by a combination of epithelial and myoepithelial elements within a stromal background that may be myxoid, chondroid, or fibrous.[5,6] A notable feature in the present case was the presence of extensive squamous metaplasia, keratin pearl formation, and lamellated keratin cysts. While rare, these features have been documented and can mimic oral squamous cell carcinoma (OSCC) or carcinoma ex pleomorphic adenoma, particularly in small biopsy samples.[14-16] However, absence of cytological atypia, abnormal mitoses, and invasive patterns helps distinguish this benign metaplastic variant from malignancy.[14]

Squamous metaplasia in Pleomorphic adenoma is thought to occur as a result of ischemic or degenerative changes, and although it may resemble malignancy histologically, it lacks the cytological features of true carcinoma.[17] In our case, FNAC showed ductal cells in a myxoid background, consistent with Pleomorphic adenoma, but definitive diagnosis was confirmed only after excision and histopathological analysis.

Though considered benign, pleomorphic adenomas have the potential to undergo malignant transformation into carcinoma ex pleomorphic adenoma, especially in long-standing or recurrent lesions.[18,19] Incomplete excision, capsular rupture, or extracapsular invasion are known risk factors for recurrence and malignant transformation.[20] In our case, no evidence of extracapsular invasion was found, indicating a lower likelihood of recurrence. However, complete excision and long-term follow-up remain critical due to the possibility of late recurrence or transformation.

In summary, this case represents a classic presentation of a palatal pleomorphic adenoma with an unusual histological profile. The combination of clinical presentation, imaging, cytology, and histopathology is essential for accurate diagnosis and effective management, especially in lesions showing metaplastic changes that could mimic malignancy.

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