

SYNCHRONOUSLY OCCURRING PLEOMORPHIC ADENOMA OF THE HARD PALATE AND LABIAL MUCOSA - A RARE CASE

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ABSTRACT

Pleomorphic adenoma of minor salivary glands is a rare benign tumour. It is the most common salivary gland tumour of the minor salivary glands. It may be most commonly seen in the hard palate and unusually areas like the buccal mucosa, labial mucosa, lips among others. Presents as a submucous mass, it is a slow growing neoplasm that is benign in nature. In this case report we are presenting a rare case of simultaneous occurrence of Pleomorphic adenoma at the hard palate and labial mucosa.

KEY WORDS

Minor salivary glands, Palate, Labial mucosa, Excision, Synchronous

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INTRODUCTION

Pleomorphic adenoma (PA) is defined as a benign salivary gland tumour composed of a mixed stroma of epithelial and myoepithelial cells, in various patterns, generally lined by a fibrous capsule. PA affects both major and minor salivary glands and accounts for 40–70% of all salivary gland tumours, making it the most common salivary gland tumour^[1]. In the major salivary glands, the Parotid gland is the most commonly affected one, the palatal glands being the most common site affected among the minor salivary glands. Other sites of this tumour are the lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, and retromolar area^[2].

PA of hard palate usually presents as a firm, smooth, slow growing mass without much signs of inflammations or ulceration around it. Usually easily demarcated from the normal architecture of the surrounding tissue, generally doesn't cause much pain. Similarly, in labial or buccal mucosa, the lesion is usually submucosal, well demarcated and rubbery in consistency. PA of the minor salivary glands, in two distinct intra oral sites, occurring simultaneously is a rarity.

CASE REPORT

A 60 year old male patient reported to the Department of Oral and Maxillofacial Surgery of Burdwan Dental College and Hospital, Burdwan, with a chief complaint of large swelling in his left side palate and a small hard mass in his upper left lip (Image 1). Patient had noticed a small swelling on his palate and lip about 1 year back, which gradually increased to the current size. He had difficulty in swallowing and articulation. He did not have any associated symptoms of pain or nasal regurgitation, etc. Past medical history was unremarkable and had history of few teeth extractions 7-8 years back which was uneventful. On systemic examination, the patient was healthy and there was mild ipsilateral regional lymphadenopathy.

Extra orally no gross facial asymmetry or swelling was noted. Intra oral examination revealed a large, (5 cm by 3 cm by 2 cm) fleshy, dome shaped, bosselated mass involving the upper left hard palate,



Image 1 – Lesion on the palate



Image 2- Lesion on the left labial mucosa

SITE : Hard palate.
 CLINICAL EXAMINATION : Single, soft, dome shaped smooth swelling over hard palate noticed for a year.
 MATERIAL ASPIRATED : Blood tinged particulate.
 MICROSCOPIC EXAMINATION : Smears examined show very high cellular yield of myoepithelial cells and ductal epithelial cells admixed with extracellular stroma. Plasmacytoid myoepithelial cells are predominant cellular component. Lesser quantity of bland ductal epithelial cells were identified. Moderate amount of fibrillary extracellular matrix with bright magenta color on Romanowsky stains are appreciated.
 No unequivocal evidence of malignancy is seen in the smears examined.
 CYTODIAGNOSIS : Hard palate swelling : PLEOMORPHIC SALIVARY ADENOMA (CELLULAR TYPE).

Image 3- Fine needle aspiration biopsy report



Image 4- Excision of the palatal lesion

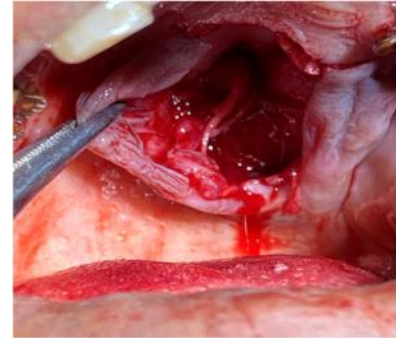


Image 5- Exposed Greater palatine artery after excision



Image 6 – Palatal lesion excised



Image 7- Excision of Left Labial lesion

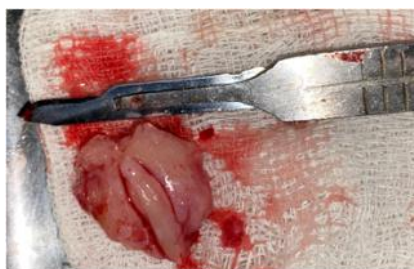


Image 8- Excision of Left Labial mucosa lesion



Image 9 – Healing of palatal surgical site with BIP paste pack



Image 10 - Acrylic palatal plate for protection of surgical site.

extending from the alveolus till the left tuberosity region. The surface appeared non ulcerated, smooth, with marked telangiectasia and a bluish hue.

Additionally, a papular, well defined, exophytic, non-ulcerated, firm mass of around 2 cm by 2 cm by 1 cm in size, involving upper left labial vestibule was noted (Image 2). It extended till the commissure of the upper lip and was painless. On bimanual

palpation, the mass could be felt between buccal mucosa and skin and was not fixed to the deeper structures.

Fine needle aspiration cytology, done elsewhere, suggested PA of the minor salivary glands (Image 3). After routine preoperative investigations, the case was planned for surgical excision. Under local anaesthesia, incision was given on the overlying

mucosa of the palatal swelling. The healthy appearing mucosa was dissected of the mass by combination of blunt, sharp and hydro-dissection. The lesion was adherent to the underlying bone of the hard palate in some areas but there was no evidence of any bony erosion (Image 4). There was some adherence of the mass to the greater palatine vessels, so the vessel was skeletonized from the mass and was ligated (Image 5). Then the mass was delivered as a whole. The mucosa, sutured back after trimming any redundant tissue (Image 6).

Similarly, the mass in the labial mucosa was separated from the mucosa and deeper muscle layers and was removed in toto, and area was closed (Image 7 and 8). Post operatively, the area was healing with minimal complications but developed some ulcerations at around 15th post-operative day. Hence the area was debrided and a BIPP (Bismuth Iodine Paraffin pack) dressing was given for secondary healing and an acrylic palatal plate was placed. Dressing was changed regularly and the surgical site went on to heal uneventfully. Histopathology examination revealed both the lesions to be pleomorphic adenoma of the minor salivary glands.

DISCUSSION

PAs are derived from a mixture of ductal and myoepithelial elements.³ It is the most common benign salivary gland tumour. PA has a slight female predilection. It may arise at any age although fourth and fifth decades of life, being the most common. The most common site of this minor salivary gland PA is the palatal area (approximately 73%),^{1,3} followed by upper lip (17%)⁴ buccal mucosa, floor of the mouth, tongue tonsil, pharynx, and retromolar area.

It arises in the oral cavity as a slowly growing, painless, firm swelling, commonly seen on the any one side of the hard palate. Generally presenting as a smooth, dome-shaped mass⁴. It gets tightly bound to the underlying bone due to the adherent nature of the palatal mucosa covering. While in cases of labial and buccal mucosa, it is freely movable. PA of palate is seldom allowed to attain a size greater than 1–2 cm in diameter because it causes difficulty in mastication, speech, and swallowing, unless it is neglected by the patient.

If the overlying mucosa is ulcerated and ulceration is not due to any trauma or biopsy, malignancy should be suspected. Computed tomography (CT) scan is an important diagnostic tool for these tumours;⁶ it helps to determine the extension of the lesion. It cannot invade bone but may lead to a cupped out resorption of bone due to pressure effect.

The PA of the labial mucosa usually presents as a painless, rubbery, lobulated submucosal swelling that does not cause ulceration of the overlying mucosa.

These benign neoplasms are usually well circumscribed and round or oval in shape. They vary

in consistency from soft and fluctuant to firm and rubbery, depending on the presence of cystic or mucoid degeneration or the formation of chondroid or osteoid tissues^{6,7}. The size of the tumours range from 1 to 7 cm in diameter with some PA of cheek may attain larger sizes

It is an epithelial tumour of complex morphology, possessing epithelial and myoepithelial elements arranged in varieties of patterns and embedded in mucopolysaccharide stroma. Formation of the capsule is a result of fibrosis of the surrounding salivary parenchyma which is composed of the tumour and is referred to as false capsule.²

Tumours of hard palate are excised below periosteum, may or may not include the overlying mucosa, with 1 cm clinical margins at the periphery.³ Excision of palatal bone is not required as periosteum is an effective anatomical barrier. In the tumour of the labial mucosa, the excision includes the fascia over muscles.

PA generally does not recur after adequate surgical excision. Reasons for recurrence include incomplete excision, seeding, cutting through the microscopic extracapsular projections thereby leaving some tumour behind, or rupture of the capsule and accidental seeding of tumour cells, as is more likely to occur when dissecting close to the capsule⁶.

CONCLUSION

Pleomorphic Adenoma of the minor salivary glands itself is a rare entity in the general population. PA appearing in two separated intraoral sites synchronously is virtually unheard of. We, as maxillofacial surgeons, should be on the lookout for other sites too once we find a PA. Hence we presented this unique case. We have kept the patient on long term follow up. Palatal surgical site ulceration and recurrence are the common complications which can be managed easily as in our case.

DECLARATION

No known conflict of interest in the among the authors.

This case report is not being published or being considered for publication anywhere else as far as our knowledge.

No financial help obtained from anywhere.

Consent was obtained by all participants in this report.

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