

A Buccal Mucosal Swelling – Diagnostic Dilemma

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ABSTRACT

Buccal mucosal swellings are commonly seen in day-to-day practice and are frequently misdiagnosed. Mostly, they are associated with buccal pad fat, buccal space, and major/minor salivary glands. These lumps are clinically diagnosed as either fibroma, lipoma, mucocele, or buccal abscess. Although a wide range of the differential diagnosis is made by the clinicians toward a single buccomucosal swelling without any other specific changes on inspection and palpation, a prompt “histopathological” diagnosis plays a pivotal role in diagnosing the exact lesion. The present case report aims to enlighten about a buccal mucosal swelling which was clinically diagnosed as lipoma but on microscopic examination identified as a different entity.

Key words: Buccal mucosa, diagnosis, histopathology, minor salivary gland

INTRODUCTION

Buccal mucosal swellings are clinically diagnosed as either fibroma, lipoma, mucocele, or pyogenic granuloma and are commonly seen associated with buccal pad fat, buccal space, and major/minor salivary glands.^[1] These lumps occurring in minor salivary glands are very rare accounting for <1% of all head-and-neck tumors.^[2] Most of them are malignant with only 18% being benign in nature.^[3]


Minor salivary gland tissue can be seen anywhere along the aerodigestive tract, but mostly concentrated along the buccal mucosa, labial mucosa, lingual mucosa, soft/hard palate, and floor of mouth. It is not generally seen on conventional imaging, but can become evident when replaced by tumor or benign processes.^[4] Minor salivary gland tumors usually present as a smooth, submucosal mass, or nodule and the rate of growth is usually

indolent. Rapid enlargement is rare in benign tumors, although might be seen in high-grade malignant salivary tumors.^[5]

Salivary gland neoplasms may present in a diversified manner of their distinct histological characteristics and clinical behavior. This marked variation warrants a thorough knowledge of anatomy and physiology, as well as expertise in pathology.^[6] The present case report aims to throw a light on a benign salivary gland neoplasm of uncommon location which was clinically diagnosed as lipoma.

CASE REPORT

A 19-year-old male patient reported to Kamineni Institute of Dental Sciences, Narketpally, Telangana, India, with a chief complaint of swelling in the right cheek area for 1 year. History revealed that the swelling was painless and gradually grew over 1 year to its present size of 1.5 × 1.1 cm². Medical history and dental history revealed the patient was healthy and had no systemic diseases nor deleterious habits. On general physical examination, the patient was moderately built and conscious, with a normal gait. His vital signs were within normal limits. The extraoral examination showed no facial asymmetry or lymphadenopathy. On palpation, the swelling was non-tender, non-pulsatile, firm, and immovable with well-defined

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margins. The mucosa over the lesion was stretched and non-pinchable [Figure 1]. Based on these features, a clinical diagnosis of lipoma was made.

Surgical excision was done under local anesthesia [Figure 2]. A single bit of tissue was received for histopathological examination which was measuring about 1.2 × 0.8 cm², brownish-white in color and firm in consistency [Figure 3]. On microscopic examination of the hematoxylin and eosin stained, tissue sections showed the presence of connective tissue stroma with numerous neoplastic salivary glandular elements showing features such as multiple duct like structures filled with homogenous eosinophilic coagulum [Figure 4]. The duct like structures in few areas are lined by single layer of cuboidal cells and in other areas

by double layer of cells, where inner cells are cuboidal and outer layer of cells are epithelioid myoepithelial cells [Figure 5]. Epithelioid cells are arranged in the form of sheets and islands within the connective tissue stroma. There is also presence of few myxoid areas and squamous metaplasia of ductal cells along with few keratin pearls formation [Figure 6]. Evidence of fibrous capsule surrounding the lesional tissue and few hemorrhagic areas was seen. Considering the ductal structures, myoepithelial cell differentiation into cellular and ectomesenchymal areas, a diagnosis of pleomorphic adenoma was made.



Figure 1: Clinical picture showing a solitary, spherical, smooth surface swelling on right buccal mucosa



Figure 2: Clinical picture showing surgical excision of the lesion



Figure 3: Grossing specimen measuring about 1.2 × 0.8 cm², brownish white in color and firm in consistency was received for processing

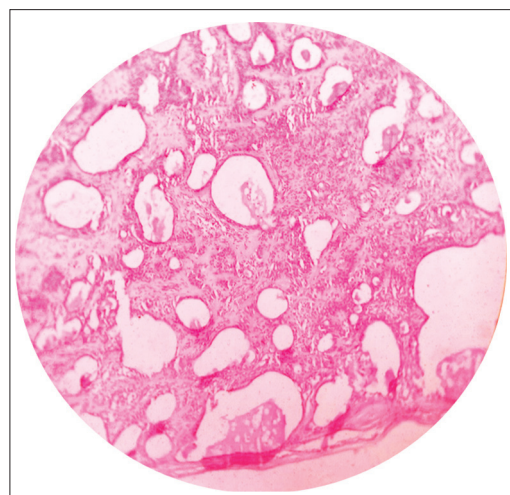


Figure 4: H&E stained section shows connective tissue stroma with numerous duct like structures filled with homogenous eosinophilic coagulum (10×)

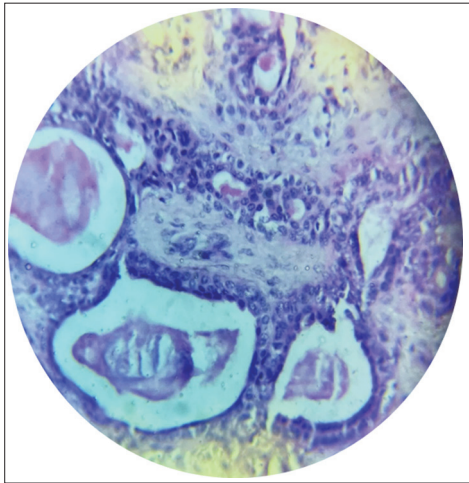


Figure 5: H&E stained section shows duct like structures lined by double layer of cells (40×)

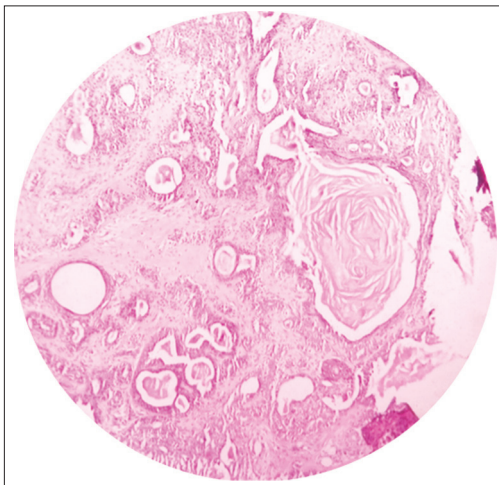


Figure 6: H&E stained section shows few myxoid areas and squamous metaplasia of ductal cells along with keratin pearl formation (10×)

DISCUSSION

Approximately 8% of pleomorphic adenomas (PA) involve minor salivary glands with palate being the most common site, followed by the upper lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, retromolar area, and nasal cavity.^[7,8] Occurrence of PA in minor salivary glands of the cheek has been variably reported in several studies.^[9] Middle age group are commonly involved; however, its occurrence in younger individuals was relatively frequent. Women are more commonly involved than men.^[7,10]

The PA of intraoral minor salivary glands seldom attains a size greater than 1–2 cm and present as a

smooth, submucosal mass, or nodule. It was usually detected earlier than tumors of major salivary glands as it causes difficulty in chewing, talking, and breathing. Apart from the size, the intraoral minor salivary gland tumor does not differ from its counterpart occurring in major salivary glands.^[7] In contrast to the conventional demographics, the present case of PA was seen in younger age grouped male patient on the buccal mucosal site.

There are various theories proposed in describing the histogenesis of this benign tumor. Hubner and his associates have hypothesized that the myoepithelial cell is responsible for the morphological diversity of the PA, including the production of the fibrous, mucinous, chondroid, and osseous areas. Regezi and Batsakis postulated that the intercalated duct reserve cell can differentiate into ductal and myoepithelial cells, and these myoepithelial cells may undergo mesenchymal metaplasia, since they inherently possess smooth muscle-like properties. Intercalated duct reserve cell may also act as a histogenetic precursor of the pleomorphic adenoma according to Batsakis *et al.*, and he also proposed that there is uncertainty in the role of myoepithelial cell, that it may be either an active or a passive participant histogenetically. Finally, Dardick and his associates said that a neoplastically altered epithelial cell with the potential for multidirectional differentiation might be histogenetically responsible for pleomorphic adenoma.^[7,11]

Genetically, putative pleomorphic adenoma gene (PLAG1) appears to be the target gene in pleomorphic adenoma with abnormalities in chromosome 8q12. Translocations involving 8q12 result in promoter swapping/substitution between PLAG1 and translocation partner genes such as CTNNB1-PLAG1 and LIFRPLAG1. This, in turn, leads to activation of PLAG1 expression. It has been postulated that deregulation of PLAG1 target genes, including IGF2, may have a significant role in the genesis of pleomorphic adenoma.^[12]

Histologically, PA shows morphological diversity with both epithelial and mesenchymal components. According to Foote and Frazell (1954), PAs are categorized into (i) principally myxoid, (ii) myxoid and cellular, (iii) predominantly cellular, and (iv) extremely cellular. Epithelial cells are arranged in cord-like and duct-like cell patterns, along with areas of epidermoid metaplasia. The connective tissue shows fibrous, hyaline, myxoid,

Table 1: Differential diagnosis of buccal mucosal swellings

Differential diagnosis	Features
Benign mesenchymal tumors	
Lipoma	The positive slip test clinically and presence of lipomatous component histologically
Schwannoma	Schwannoma shows a well-defined and Encapsulated tumor mass comprised of schwann cells
Neurofibroma	An ill-defined mass and composed of schwann cells, perineurial cells and endoneurial fibroblast-like cells
Fibroma	Dense collagenous stroma
Leiomyoma	Benign smooth muscle tumor, composed by spindle shaped cells with eosinophilic cytoplasm and cigar-like shaped nuclei admixed with variable vascular stroma
Myofibromas	Spindle cell proliferation exhibiting a biphasic pattern around irregular blood vessels
Reactive lesions	
Mucocele	Mucocele exhibits mucus extravasation surrounded by a wall of granulation tissue
Fibrous hyperplasia	Characterized by hyperplastic epithelium and large collagen fibers deposition
Malignant minor salivary gland tumors	
Mucoepidermoid carcinoma	Presents mucous cells together with epidermoid and intermediate cells
Adenoid cystic carcinoma	Exhibits distinct architectural patterns described as tubular, cribriform, and solid
Benign/reactive and malignant lymphoid proliferations	
Follicular lymphoid hyperplasia	The lesion is composed of multiple Follicles with well-defined germinal centers
Non-hodgkin lymphomas	Presence of Reed Sternberg cells and lymphoid tissue
Buccal space abscess	Shows signs of inflammation
Mature dermoid cyst.	Lack of tissue showing the three germ layers
Sebaceous cyst	Shows punctum and fixed mass
Polymorphic low-grade adenocarcinoma.	Absence of perineural invasion and mitotic figures

cartilaginous, and osseous areas. In the minor glands, lesions are often more solid or cellular

than those seen in the major glands, and the myoepithelial cells are often polygonal with a pale eosinophilic cytoplasm giving an epithelioid or plasmacytoid phenotype.^[2,7,9,10] Crystalloids, including collagenous material and tyrosine-rich crystals, which form flowerlike (“daisy head”) structures and oxalate such as crystals, may be seen.^[12] Most of the tumors of minor salivary glands are unencapsulated in nature.^[7] In contrast to the conventional features, the present case microscopically showed encapsulation and contains both myxoid and cellular components.

Various differential diagnosis of buccal mucosal swellings is given in the Table 1.^[13]

Fine needle aspiration cytology is recommended in obtaining biopsy sample for diagnosis as, incisional biopsy is not advised for salivary gland tumors due to recurrence. Surgery is the treatment of choice for PA. Resection of the tumor with an adequate margin is necessary to prevent local recurrence as these tumors are known to have microscopic extension into the surrounding tissue.^[5,8,14]

CONCLUSION

A buccal mucosal swelling may present with diagnostic challenges in clinical practices. Hence, careful evaluation of patient history is needed and histopathology plays a key role in confirming the diagnosis. In the present case, a clinical diagnosis of lipoma was ruled out by histopathological diagnosis of pleomorphic adenoma.

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