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Intraoral variant of yellow epulis – Case report and review of literature

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Abstract

Lipoma also known as yellow epulis is a common benign, slow growing, usually asymptomatic mesenchymal tumour composed of mature fat cells predominantly affecting people in the fourth or fifth decade of life. It is rarely seen in the oral cavity, but is commonly observed in the head and neck region. A less common histological variant of the typical lipoma is known as fibrolipoma, accounting for 1.6% of all facial lipomas. Fibrolipomas, mainly affecting the buccal mucosa and buccal vestibule, can lead to functional and cosmetic problems. Owing to its non-invasive behaviour and low rate of recurrence, surgical excision is usually considered as the best therapeutic option. In this article, we present the case of a 53-year-old female patient with fibrolipoma of the left buccal mucosa.

Keywords: yellow epulis, lipoma, fibrolipoma.

Introduction

Epulis is any tumour like enlargement in the gingiva or alveolar mucosa. It's worth noting that Roux, in a review of alveolar masses dating back to 1848, referred to this Type of lesion as a “yellow epulis”, This marked the first recorded description of an oral lesion of this nature.

Lipomas are the most prevalent mesenchymal neoplasms, typically occurring in the trunk and proximal extremities. Their occurrence in the oral cavity is relatively rare, with an overall reported incidence of 4.4%. (2) Lipomas tend to occur more frequently in areas where adipose tissue accumulates, such as the cheeks, tongue, floor of the mouth, vestibule and buccal sulcus, lip, palate, and gingiva. (3)

The World Health Organization [WHO] (4) categorizes fibrolipoma as an infrequent histological variation of the typical lipoma, characterized by the presence of dense collagen and mature fat cells. Patients aged 40 and older constitute the majority of cases. Fibrolipoma can manifest in various anatomical sites, including the lips, tongue, palate, floor of the mouth, and buccal mucosa. Although it is benign, the tumor's gradual enlargement may impede speech and chewing due to its size. (4,5)

Case-report

A 53-year-old female patient visited the dental outpatient department with the chief complaint of a painless swelling in the left buccal mucosa which had been present for a period of three months (figure 1). She has been a known diabetic for the past five years and is currently receiving medication for the same. Intraoral examination revealed a well-defined swelling in the left buccal mucosal area, measuring approximately 1x1 cm with the surface mucosa having a yellowish tinge. On palpation, the swelling seemed to be localized, lobulated, pedunculated, compressible, fluctuant and non-tender. Subsequent to the surgical excision of the lesion under local anaesthesia, a provisional diagnosis of fibroma was made. The grossing specimen measured 1x1cm, appeared to be white in colour and was firm in consistency (figure 2). Microscopic examination revealed a well-encapsulated lesion consisting of mature adipocytes with clear cytoplasm and an eccentric nucleus. These tumour cells were organized into lobules composed of "chicken wire" appearing benign adipocytes with intervening wide and dense collagenous bands (figure3, 4 & 5). There was no evidence of cellular atypia or metaplasia. The final diagnosis of fibrolipoma was made histopathologic ally. There was no evidence of recurrence after a follow-up period of 1 year

Figure 1: Intra-Oral image revealing the localisation and site of swelling.

Figure 2 : specimen received for grossing

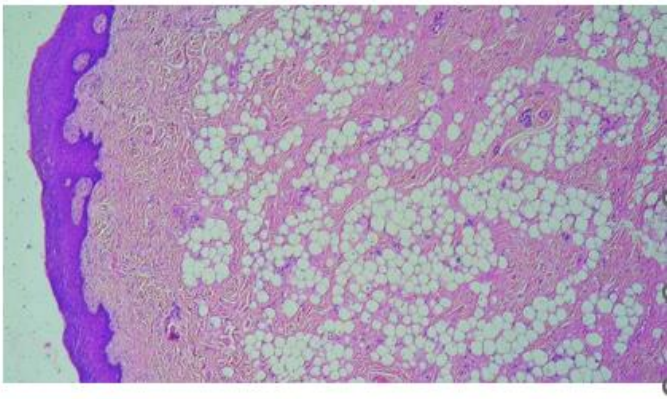

Figure 3 : Histopathological image (4x) showing a non-keratinized stratified squamous epithelium and densely collagenous connective tissue.

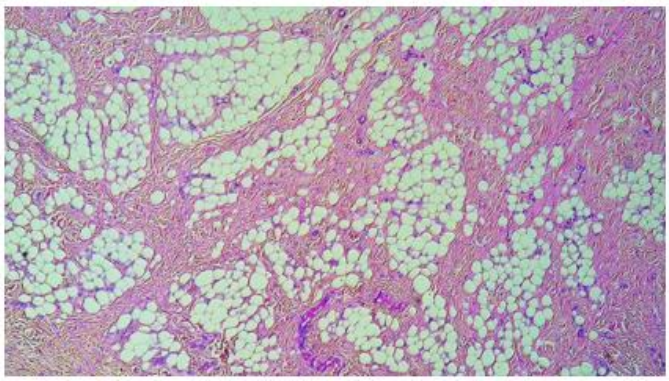

Figure 4: Histopathological image (10x) showing lobules composed of "chicken wire" appearing benign adipocytes with intervening wide and dense collagenous bands.

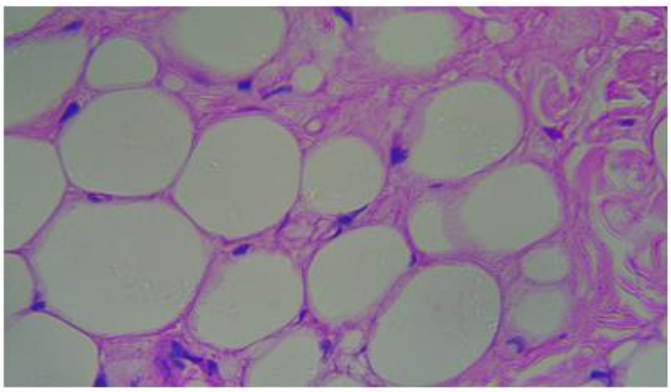

Figure 5: Histopathological image (40x) showing mature adipocytes with eccentrically placed nucleus and a clear cytoplasm.

Discussion

Lipomas are the most prevalent mesenchymal neoplasms, but they account for less than 4.4% of all benign mesenchymal tumors found in the oral cavity. Their occurrence in the oral region is relatively rare, with a prevalence rate of only 1 in 5000 adults. In contrast, lipomas more commonly develop in other locations, particularly within subcutaneous tissues of the neck, trunk, and proximal extremities.

Lipoma affects both genders equally and is more prevalent in individuals over the age of thirty. It accounts for approximately 5% of all soft tissue tumors. At the time of diagnosis, the size of a lipoma typically does not exceed 3 cm; however, over the years, it may gradually grow to a maximum size of 5–6 cm. Intraoral locations where lipomas can be found include the tongue, floor of the mouth, gingiva, mucobuccal or labial folds, palate, and major salivary glands. While most oral lipomas do not usually produce symptoms, larger lesions have the potential to impair oral function.

Both fibrolipoma and lipoma have an unknown etiopathogenesis. Although they typically suggest a developmental anomaly, they can also result from factors such as trauma, hormonal fluctuations and rearrangements involving 12q, 13q, and 6p chromosomes. (1,4) According to Epivatianos et al, fibrolipoma may be a congenital lesion triggered by an endocrine imbalance, originate from the maturation of lipoblastomatosis, or result from the degeneration of a fibromatous tumor. (4) As per the "metaplasia theory" suggested by Evans R. Winston, the formation of lipomatous tissue results from the aberrant differentiation of local mesenchymal cells into lipoblasts. It has been suggested that the disturbances in glucose and lipid metabolism and hormonal therapy are

probable contributory factors for the development and proliferation of lipoma. (6)

While lipoma cells share histological similarities with normal fat cells, it appears that they are not metabolically identical. As a result, fat from a lipoma will not be lost by someone undergoing a starvation diet; rather, the body's regular fat stores will be utilized. Additionally, lipoprotein lipase activity is reduced, and fatty acid precursors are absorbed into lipoma fat more rapidly compared to normal fat. (1)

In accordance with their morphological characteristics, Rajendran and Sivapathasundharam have classified intraoral lipomas into three categories: (1,6)

- The diffuse form which affects deeper tissues,
- The superficial form and
- The encapsulated form.

Fibrolipoma are characterized by lobules composed of "chicken wire" appearing benign adipocytes with intervening wide and dense collagenous bands. These lesions often demonstrate a thin capsule and shows a well-defined appearance. (4,6) The histopathological features observed in this particular case are in accordance with these characteristics.

Additionally, Table 1 provides a list of histologic variations of lipomas.

Table 1: Histologic variants of lipomas

Variants	Features
Fibrolipoma	Adipocytes intermixed with densely packed collagen bundles. Significant fibrous component is seen.
Intra and intermuscular lipoma	Deeply situated and extends between skeletal muscle bundles.
Angiolipoma	Exhibits a combination of mature adipocytes and numerous small blood vessels
Myxoid/ Myxolipoma	Numerous adipocytes and scattered spindle cells in a myxoid background
Spindle cell lipoma	Presence of abundant spindle-shaped cells with lipomatous component
Pleomorphic lipoma	Spindle-shaped cells and pleomorphic multinucleated giant cells showing hyperchromatic nuclei often showing a floret-like appearance.
Myolipoma	Presence of abundant spindle-shaped cells arising from smooth muscle intermixed with Adipocytes.
Angiomyolipoma	Lesion arising from arterioles of smooth muscle and shows spindle-shaped cells
Osteolipoma/Ossifying lipoma	Metaplastic osteoid scattered in a background of lipomatous proliferation
Chondrolipoma	Adipocyte proliferation associated with mature cartilaginous/chondroid tissue
Myelolipoma	Adipocytes intermingled with myeloid tissues.
Adenolipoma	Presence of scattered duct or tube-like Adnexal structure in a lipomatous background
Perineural lipoma	Presence of nerve tissue entrapment within fat cells.
Sialolipoma	Lipomatous proliferation with salivary gland structures with varying degree of acinar atrophy, often seen in parotid and rarely seen in minor salivary glands

Multiple head and neck lipomas have been associated with various medical conditions, including Neurofibromatosis, Gardner syndrome, Proteus

syndrome, Multiple family lipomatosis and Encephalocraniocutaneous lipomatosis. (1,4)

To the best of our knowledge, the English literature review has documented only 43 previous cases of oral cavity fibrolipoma. The following table 2 illustrates the distribution specific to each site, with the instance described in this article being a fibrolipoma located in the buccal mucosal region.

Table 2: Site distribution of oral fibrolipoma [4]

Sn.	Site	Number of cases reported
1.	Buccal mucosa	19
2.	Lateral border of tongue	5
3.	Palate	4
4.	Floor of mouth	4
5.	Lower lip and upper lip	3
6.	Alveolus (including retromolar area)	3
7.	Gingiva	2
8.	Vestibular region	2
9.	Tonsil	1
10.	Intra osseous lesion	1
	Total case (including Current case in Buccal mucosa)	44 cases

Lipomas are known to exhibit immunohistochemical staining positivity for vimentin, proliferating cell nuclear antigen (PCNA) & Ki-67. In contrast to conventional lipomas, fibrolipoma shows increased expression of the Ki-67 antibody on immunohistochemical labelling, indicating a higher potential for proliferation (7,9).

The range of potential diagnoses includes a variety of conditions, such as cysts like mucocele, lymphoepithelial cyst, benign tumors like nodular fasciitis, lipoblastoma, benign salivary gland tumors, myxoid neurofibroma, neuroma, leiomyoma, as well as malignant tumors like malignant fibrous histiocytoma and liposarcoma (7).

The gene responsible for fatty acid-binding protein 4 (FABP4), also known as adipocyte P2 or aP2, is actively expressed in lipoblasts and preadipocytes. All cases of adipose differentiation tumors exhibited aP2/FABP4 expression, while this expression was absent in other types of lesions. A lack of High Motility Group AT Hook 2(HMGA2) expression serves as a diagnostic indicator for hibernomas, unlike lipomas, which do not exhibit these markers. In contrast, liposarcomas can display varying degrees of differentiation, atypical cells, increased mitotic activity. Lipoblastomas can be distinguished by variably sized fat vacuoles with a light, wispy cytoplasm and presence of some cells showing signet-ring appearance (1,8). Surgical excision is the typical approach for treating lipomas, including fibrolipoma. The potential for malignant transformation and recurrence is rare.

Conclusion

The diagnosis of fibrolipoma should be done with caution, since this tumor has a higher potential for proliferation and recurrence among the lipomas. This is very essential for a corrective treatment plan and a complete follow-up.

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