Oral focal mucinosis of palatal mucosa: A rare case report

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Abstract

Oral focal mucinosis (OFM), an oral counterpart of cutaneous focal mucinosis, is a rare disease of unknown etiology. Its pathogenesis may be due to the overproduction of hyaluronic acid by a fibroblast, at the expense of collagen production, resulting in focal myxoid degeneration of the connective tissue, primarily affecting the mucosa overlying the bone. It has no distinctive clinical features, as the diagnosis is solely based on the histopathological features. This article reports of a 32-year-old female having the rare disease of oral focal mucinosis, involving the posterior palatal mucosa, and discusses its clinicopathological features and differential diagnosis of myxomatous lesions of the oral cavity.

Keywords: Cutaneous focal mucinosis, focal myxoid degeneration, hyaluronic acid, myxomatous lesion, oral focal mucinosis

Introduction

Oral Focal Mucinosis (OFM) is an uncommon clinicopathological condition that is considered to be the oral counterpart of cutaneous focal mucinosis (CFM). It is a disease of unknown etiology, possibly resulting from the overproduction of hyaluronic acid by fibroblasts.[1] Clinically the lesion appears as asymptomatic round elevations, which are histologically characterized by a localized area of myxomatous connective tissue, containing mucinous material, surrounded by relatively dense collagenous connective tissue.[2]

It was first described and named by Tomich in 1974. He reported eight cases as oral counterparts of cutaneous focal mucinosis. However, similar lesions were also previously described, in 1966, by Jhonson and Helwig, as solitary, asymptomatic, dome-shaped skin nodules seen usually on the face, trunk, and extremities.[2,3] A review of the literature revealed that since the original article by Tomich, 57 cases of oral focal mucinosis were reported in English literature,[2,4-13] while only three cases has been reported in Indian literature[14] including the present case. [Table 1].

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The mucosa directly overlying the bone appears to be particularly vulnerable, with the gingiva being the most common site, followed by the hard palate. Despite being the second most common site, the lesions of the hard palate are rare, with only seven cases being reported to date. The lesions are generally the same color as the surrounding normal mucosa, and skin lesions do not seem to accompany the oral lesions. The lesions are difficult to diagnose clinically, as there are no clinical distinctive features. A review of the available literature shows that it is most commonly diagnosed as gingival hyperplasia; fibroma ,or epulis.[3] Thus, the histopathological diagnosis becomes important in these conditions.

Case Report

A female patient aged 32 years, reported to the Department of Periodontology, Government Dental College and Hospital, Patiala, with a chief complaint of a swelling on her right posterior palatal mucosa, since four to five months [Figure 1]. The growth had increased in size since three months, but it showed no significant increase in size since one month. The medical history was non-contributory. An intraoral examination revealed a pinkish red, painless, sessile, firm, tender mass, measuring 1.5 cm at its greatest dimension. The lesion had extended from the distal aspect of 16 to the mesial aspect of 18. No surface ulceration or secondary infection was noted and the oral hygiene of the patient was reasonably good. The first clinical impression at examination was that of fibrous hyperplasia.

Investigations

The laboratory investigations for blood were within normal limits. The intraoral periapical radiographic examination revealed no evidence of crestal bone loss, and the lamina dura was intact around the roots of both the maxillary second and third molar regions.

Management

After one week of thorough scaling and root planing, the growth was surgically excised involving the second and

Table 1: Published reports of oral focal mucinosis

Authors	Age sex	Location	Clinical diagnosis	Treatment	Recurrence
Tomich[4]	40 / F	Palate	Fibroma	Excision	No
	31 / F	Gingiva	NA	Excision	No
	16 / M	Gingiva	Fibroma	Excision	No
	NA / F	Buccal mucosa	Papilloma	Excision	No
	45 / M	Tongue	Mucocele	Excision	No
	28 / M	Alveolar mucosa	Fibroma	Excision	No
	22 / F	Hard palate	Mucocele	Excision]	No
	19 / F	Hard palate	Fibroma	Excision	No
Saito *et al.*[6]	35 / M	Gingiva	Fibroma	Excision	No
	50 / F	Gingiva	Fibroma	Excision	No
Gnepp *et al.*[5]	4 / F	Hard Palate	NA	Excision	No
Buchner *et al.*[2]	18 / F	Gingiva	NA	Excision	No
	30 / M	Gingiva	NA	Excision	No
	32 / F	Gingiva	NA	Excision	No
	22 / F	Gingiva	NA	Excision	No
	53 / F	Gingiva	NA	Excision	No
	16 / F	Gingiva	NA	Excision	No
	43 / M	Gingiva	NA	Excision	No
	61 / F	Alveolar mucosa	NA	Excision	No
	37 / F	Alveolar mucosa	NA	Excision	No
	41 / F	Gingiva	NA	Excision	No
	37 / F	Gingiva	NA	Excision	No
	46 / M	Gingiva	NA	Excision	No
	38 / F	Hard palate	NA	Excision	No
	46 / M	Gingiva	NA	Excision	No
	50 / M	Tongue	NA	Excision	No
Soda *et al.*[7]	68 / M	Tongue	NA	Excision	NA
Etges *et al.*[8]	40 / F	Gingiva	Fibroma	Excision	NA
	43 / M	Gingiva	Fibroma	Excision	NA
Iezzi *et al.*[9]	48 / M	Gingiva	Periodontal abscess	Excision	No
Aldred *et al.*[10]	38 / F	Lip	NA	Excision	No
	30 / F	Gingiva	Fibrous epulis	Excision	No
	16 / F	Gingiva	Fibrous hyperplasia	Excision	No
	56 / F	Buccal mucosa	NA	Excision	NA
	60 / F	Mouth (?)	Papilloma	Excision	NA
	49 / M	Gingiva	Polyp	Excision	No
	31 / F	Gingiva	Giant-cell granuloma	Excision	No
	52 / M	Gingiva	Fibrous epulis	Excision	No
	74 / M	Lip	NA	Excision	No
	40 / F	Gingiva	Fibroma	Excision	No
	55 / M	Tongue	Fibroepithelial polyp	Excision	No
	37 / F	Gingiva	NA	Excision	NA
	35 / F	Gingiva	Fibrous epulis	Excision	NA
	33 / F	Gingiva	Fibrous epulis	Excision	
	68 / M	Gingiva	Fibroepithelial polyp	Excision	

Table 1: Contd...

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Authors	Age sex	Location	Clinical diagnosis	Treatment	Recurrence
Talacko *et al.*[11]	63 / F	Buccal mucosa	Swelling with fistula	Excision	NA
	24 / M	Gingiva	NA	Excision	NA
Lima *et al.*[3]	36 / F	Gingiva	Fibroma	Excision	No
Madhusudhan AS *et al.*[14]	50 / F	Palate	Gingival epulis	Excision	No
	26 / F	Gingiva	Fibrous epulis	Excision	No
Narayana N *et al.*[12]	37 / F	Gingiva	Fibroma	Excision	No
	54 / F	Gingiva	Fibroma	Excision	No
	49 / M	Palate	Pleomorphicadenoma	Excision	No
	25 / F	Gingiva	Fibroma	Excision	Yes
	26 / M	Gingiva	Fibroma	Excision	No
	32 / F	Gingiva	Gingival cyst	Excision	No
	48 / F	Gingiva	Gingival cyst	Excision	No
Gabay E *et al.*[13]	44 / F	Gingiva	NA	Excision	NA
Lee JG *et al.*[19]	17/F	Gingiva	NA	Excision	No
Bharti V *et al.* (present study)	32/F	Palate	Fibrous hyperplasia	Excision	No

*NA = not available

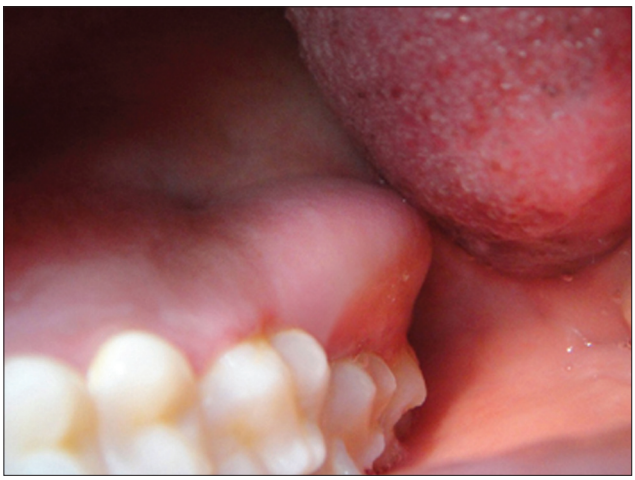

Figure 1: Clinical aspect of oral focal mucinosis involving the right maxillary second and third molar area

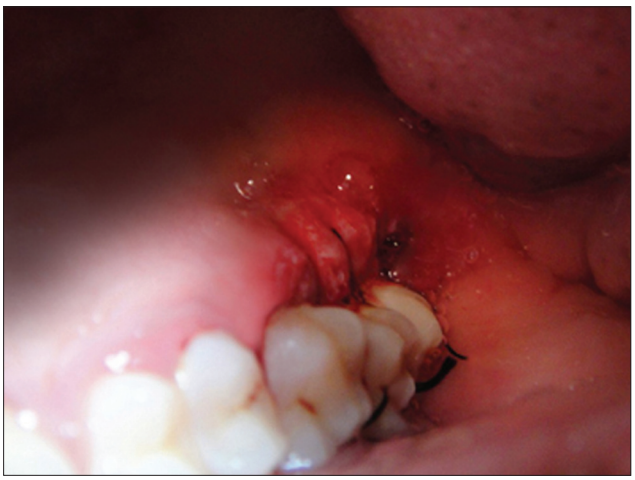

Figure 2: Immediate operative view following periodontal surgery

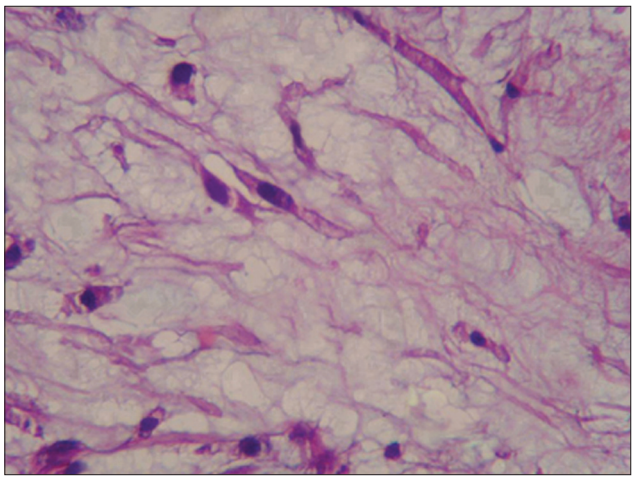

Figure 3: Shows loose myxomatous connective tissue stroma with fusiform or stellate-shaped fibroblasts. ×400 (H and E)

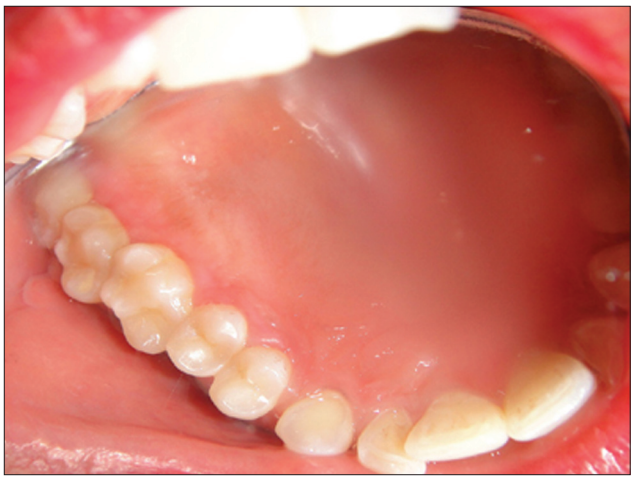

Figure 4: Shows no sign of recurrence at the one-year follow-up

third molar area and a thorough curettage of the area was performed under local anesthesia. The excised lesion was transported in 10% formalin to the laboratory for histopathological examination. Sutures and periodontal dressing were placed on the operated area [Figure 2]. After one week the sutures and dressing were removed.

Histopathological findings

The hematoxylin and eosin–stained microscopic slide revealed a loose, myxomatous connective tissue stroma, with ovoid, fusiform or stellate-shaped fibroblasts. Fibroblasts represent delicate fibrillar processes extending from the cytoplasm into the surrounding matrix. A scarcity of inflammatory cells and reticulin fibers was observed. These histopathological features were suggestive of Oral Focal Mucinosis (OFM) [Figure 3].

Clinical course

The patient has been observed on a regular basis; no recurrence was observed at the one-year follow-up [Figure 4].

Discussion

Oral focal mucinosis is a rare disease of unknown etiology where the connective tissue undergoes focal myxoid degeneration, first described by Tomich, in 1974.[4] It is the oral counterpart of cutaneous focal mucinosis and the cutaneous myxoid cyst. Cutaneous focal mucinosis seems to be a mesenchymally derived lesion, composed predominantly of fibroblasts.[15] Oral focal mucinosis may be the result of the overproduction of hyaluronic acid by fibroblasts at the expense of collagen production.[2,4,5-7] There is a predilection for the mucosa of the overlying bone, and keratinized mucosa is involved almost exclusively, with 80% of the lesions developing on the gingiva and the remainder on the palate;[4] other locations are the alveolar mucosa and tongue.[6]

From a clinical point of view, the differential diagnosis must be made with inflammatory lesions (i.e., fibrous hyperplasia, peripheral giant cell granuloma, epulis fissuratum, pyogenic granuloma) and tumors (i.e., peripheral fibroma and peripheral ossifying fibroma).[16] Also, other lesions such as primary benign (i.e., peripheral ameloblastoma and peripheral odontogenic fibroma), malignant, or metastatic lesions may appear as a gingival mass. Finally, a number of non-plaque–related lesions (irritation fibroma, fibrous hyperplasia) must be included in the differential diagnosis.[16] A review of all reported cases show that they were never diagnosed clinically as oral focal mucinosis.[16] The histological features are always the basis for diagnosis.

The histological differential diagnosis of oral focal mucinosis includes, inflammatory fibroepithelial hyperplasia, mucocele, nerve sheath myxoma, and myxoma.[5,6,8] Similar myxoid areas may be found in inflammatory fibroepithelial hyperplasia, but unlike oral focal mucinosis, these are accompanied by inflammation and fibrosis elsewhere. Unlike oral focal mucinosis, extravasation mucoceles (that result from minor trauma of the lips) have an associated inflammatory cell infiltrate and a granulation tissue wall. Nerve sheath myxoma is characterized by a whorled arrangement of tumor cells in an organoid, multinodular, or lobular structure.[17] The myxoma is a true neoplasm resembling embryonal mesenchyme, consisting of widely separated stellate and sometimes spindle-shaped cells in a loose mucoid stroma, with a network of delicate reticular fibers.[17] Myxoma may present as an infiltrative growth pattern, while focal mucinosis usually manifests as a localized area of myxomatous connective tissue.[4] Small pools of mucinous material are a feature in many cases of focal mucinosis, but are not present in myxomas.[18]

Conclusion

It must be emphasized that in many of the focal gingival lesions, a preoperative diagnosis is almost impossible, due to their rarity. The histological evaluation is always the basis for diagnosis. Oral focal mucinosis is an uncommon condition that occurs predominantly in females and in adults. The gingiva is the most common site of its occurrence, followed by the hard palate. Despite being the second most common site, the lesions of the palatal mucosa are rare, with only seven cases being reported to date. Oral focal mucinosis rarely occurs upon surgical excision of the lesion.

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