*Case Report*
**Journal of Pierre Fauchard Academy (India Section),** *Vol 37(1), DOI: 10.18311/jpfa/2023/33826, January-June 2023 p. 37-424*
*ISSN (Print) : 0970-2199*
*ISSN (Online) : 2405-772X*

# Case Report and Literature Review of a Rare Oral Lesion: Oral Focal Mucinosis

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## Abstract

Focal gingival lesions are quite commonly encountered by general dental practitioners. These gingival growths may range from being reactive lesions like inflammatory hyperplasia to malignant growths. Hence, histopathological examination and diagnosis of all these lesions on excision are mandatory. One such uncommon tumor-like growth of the gingiva is Oral Focal Mucinosis (OFM). It is a rare lesion originally reported by Tomich in 1974. Clinically, it mimics the common benign oral tumors of the gingiva. Only while grossing of the specimen, the first indication of it being an OFM with a differential diagnosis of myxoma is evident. Histopathological examination and special stains help in the final diagnosis. Currently, considered to be a degenerative change in a reactive lesion, simple excision has been the line of treatment with a good prognosis. Through these case reports we attempt to make this lesion familiar to the practicing dentists.

**Keywords:** Gingival Lesions, Oral Focal Mucinosis, Oral Lesions, Oral Pathology, Periodontal Pathology

(**Article chronicle: Received:** 19.05.2023; **Revised:** 04.06.2023; **Accepted:** 14.07.2023)

## 1. Introduction

Oral Focal Mucinosis (OFM), an uncommon tumor-like growth of unidentified etiology is considered to be the oral counterpart of a cutaneous lesion. The corresponding dermal lesion is Cutaneous Focal Mucinosis (CFM) or cutaneous myxoid cyst[1]. The disease is more common in females[2] and in the oral cavity, gingiva is the most common site of occurrence. The lesion mostly presents with the same colour as adjacent mucosa, as a painless mass that may be pedunculated or sessile.

The etiology of the disease is not specified but it is suggested to be due to the hyaluronic acid overproduction by fibroblasts. Histologic features show focal myxoid degeneration of the connective tissue[3]. The lesion is a soft tissue rounded elevation which is asymptomatic. Diagnosis is made by the histopathological examination and complete surgical removal is the suggested treatment modality[4].

Hereby, reporting two cases of OFM sent to our department with the provisional diagnosis of fibroma.

## 2. Case Presentation

### Case I

A 38-year-old female reported soft tissue growth between her anterior teeth. On oral examination, a soft tissue mass was present in the interproximal area between 11

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and 21 that had gradually increased to the current size over 3-4 months. The lesion was pedunculated 1×1cm in dimensions and reddish pink in appearance. No pain or bleeding was associated with the lesion. (Figure 1) Scaling, root planning, and curettage were done for the same and on the next recall visit, excision of the lesion was done under local anaesthesia and sent for histopathological examination. On gross examination, the tissue specimen was soft to firm in consistency, creamish white in color, with irregular shape, surface, and margins. The lesion was excised and sent to the Department of Oral Pathology for the final diagnosis.

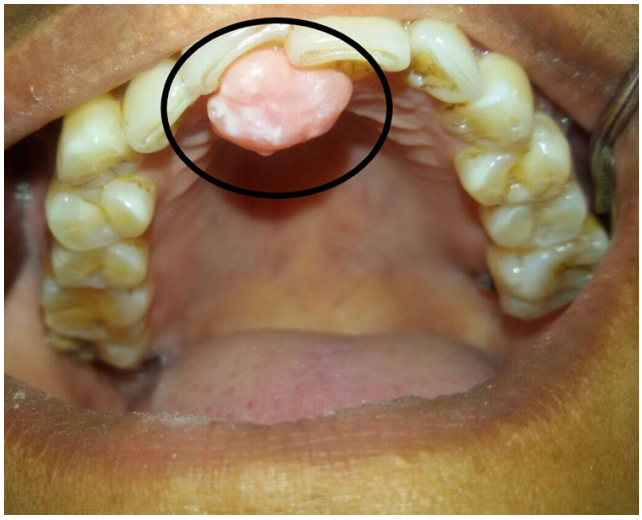


**Figure 1.** Clinical presentation of lesion in the interproximal area between 11 and 21.

## Case II

A 35-year-old female patient came with the complaint of a painless sessile soft tissue growth in the right maxillary posterior quadrant on the palate for 3 months before she reported to the department and the lesion had increased gradually in size (Figure 2). Oral prophylaxis of the patient was done, and the lesion was excised after a week and sent to our department for histopathological examination. On gross examination, the tissue was soft to firm (jelly-like) on palpation, creamish to brownish white in colour, and ovoid in shape but with an irregular surface measuring 1.3×0.5×0.5 cm in dimensions.

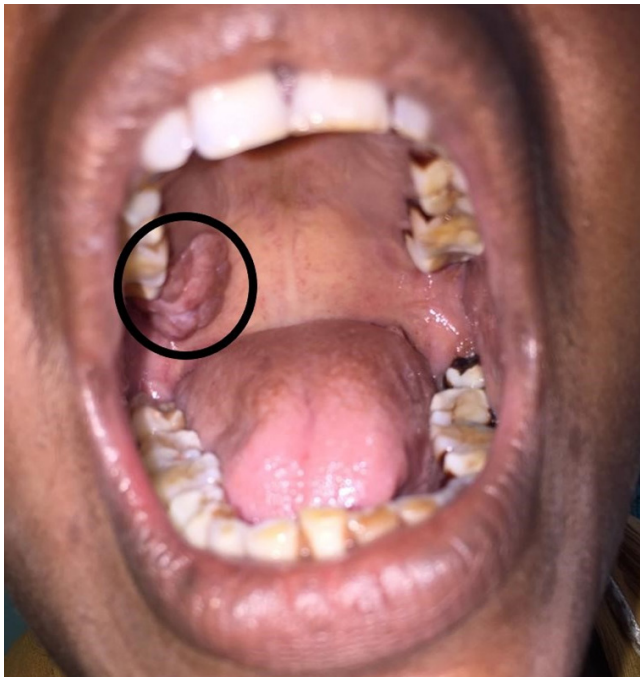


**Figure 2.** Clinical presentation of lesion on the right maxillary posterior palate.

### 2.1 Histopathological Examination

Grossing of both specimens showed a similar appearance of the cut surface. It was a greyish-white gelatinous mass with a smooth glistening surface (Figure 3). The haematoxylin and eosin-stained slides of the two cases revealed common para keratinized stratified squamous epithelium with flattened rete ridges. The myxomatous lesion extended up to the surface epithelium. The lesion showed loose, fibromyxoid stroma with fusiform to stellate-shaped fibroblasts interspersed by thin collagen fibers. Fewer blood capillaries in comparison to the deeper normal connective tissue stroma were seen.




**Figure 3.** Gross examination showed a smooth glistening white surface.

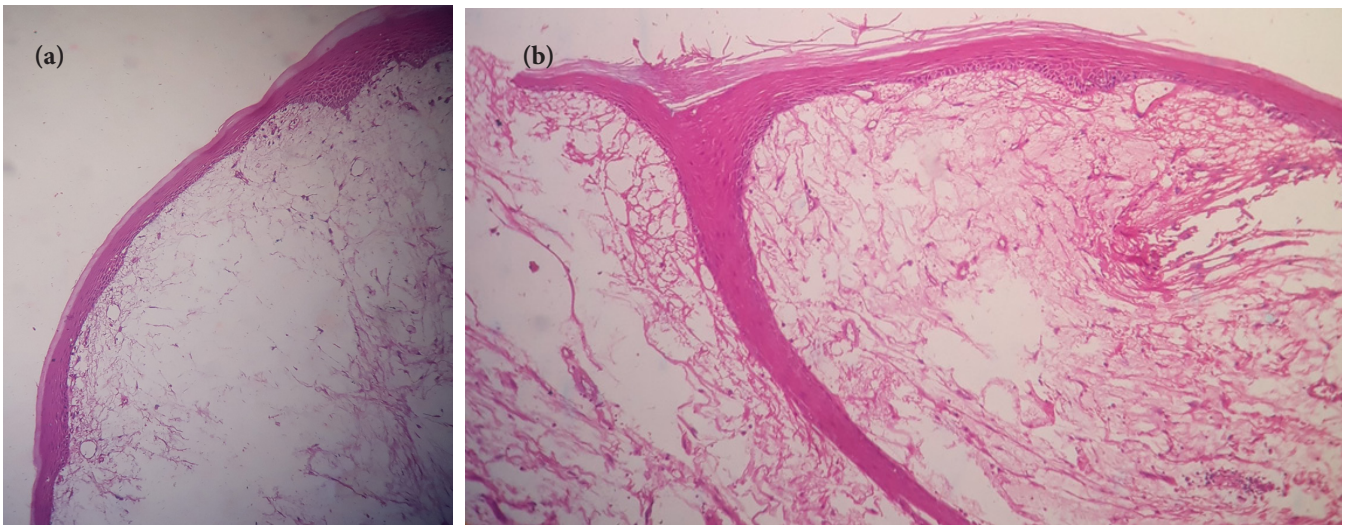



**Figure 4.** **(a)** H and E-stained sections showing loose myxoid connective tissue with atrophic stratified squamous epithelium at 10X.; **(b)** H E-stained sections showing loose myxoid connective tissue with atrophic stratified squamous epithelium at 10X.

Inflammatory cells were minimal and those present were around the capillaries. Few cleft-like spaces/ cavities were present within the lesion. The margins of the lesion were partially delineated with areas of gradual transition from the myxomatous to collagenous tissue (Figure 4(a) and (b)). Special stains were used and both cases showed positive results with alcian blue stain (Figure 5(a) and (b)) and negative results with reticulin stain. A final diagnosis of Oral Focal Mucinosis was given.

### Clinical Course

The cases were treated with complete excision of the lesion. Instructions for oral hygiene were given to the patients as in these two cases the oral hygiene was also compromised. During post-operative follow-up, the lesions healed satisfactorily without any consequences. After a follow-up of 3 years and 1 year respectively neither of the cases presented any recurrence.

## 3. Discussion

Oral Focal Mucinosis lesions do not have any distinctive clinical features of identification. The first clinical impression is similar to a fibroma, pyogenic granuloma, mucocele, or peripheral ossifying fibroma. A review of all the cases that have been reported shows that the lesion was never clinically diagnosed as Oral Focal Mucinosis[5,6].

The majority of the lesions occurred on the gingiva and palate while the remaining few occurred on the buccal mucosa, tongue, retromolar area, and lip. Of all the cases that occurred on the gingiva and hard palate, three fourth of them presented with normal surface coloration. The rest were with a pale appearance. On visual examination, it presents as a soft, painless, and sessile nodule[7]. The differential diagnosis for such benign oral lesions could be fibrous hyperplasia, traumatic fibroma, peripheral giant cell granuloma, pyogenic granuloma; peripheral ossifying fibroma, peripheral ameloblastoma, malignant or metastatic tumors in gingiva. However, the basis of final diagnosis always is the histopathological evaluation.

As per the best of our knowledge and literature reviewed by the author, a total of 86 cases of OFM have been reported till now since the original case was reported by Tomich[8]. Out of these cases only 10 cases have been reported by Indian authors. A review of OFM cases reveals that the lesion has been observed more in the mandibular arch as compared to the maxillary arch[9] and 68% of case reports show gingival predilection (Table 1). Most of the reported cases (55%) were not clinically diagnosed as a particular clinical lesion, only a differential diagnosis was given. Out of the total clinical diagnosis, the majority were fibroma. On clinical examination, OFM presented as a painless, solitary, round, non-ulcerated, elevated lesion[10]. Our cases corroborate all these observations except that both cases were observed in the maxilla.

**Table 1.** Data of literature reviewed 87 cases of oral focal mucinosis[2 -11,13,15-38]

| Classification | Frequency |
|---|---|
| **Age (years)** | **Range 2-68 years** |
| **Male: female ratio** | **29:58** |
| Region of origin | |
| Gingiva<br>Palate<br>Buccal mucosa<br>tongue<br>Retromolar<br>Lip<br>Others | 59(68%)<br>12<br>4<br>5<br>2<br>2<br>3 |
| Clinical diagnosis | |
| Fibroma<br>Epulis<br>Papilloma<br>Mucocele<br>Periodontal abscess<br>Giant cell granuloma<br>Pleomorphic adenoma<br>Unknown | 23(27%)<br>9<br>2<br>2<br>1<br>1<br>1<br>48(55%) |
| Treatment method | |
| Surgical resection | 87(100%) |
| Recurrence | 1 |

The etiology of this lesion is unknown. Overproduction of hyaluronic acid has been suggested as the probable mechanism by Tomich, but no reason was suggested to support this. He suggested that hyaluronic acid is overproduced by fibroblasts at the expense of collagen production[7].

Gnepp *et al.,* supported trauma as a contributing factor by documenting its role as a contributing factor[11] while Tomich did not support trauma as the etiological factor[8]. In our case reports, neither patient reported a history of trauma. Neither was there any surface ulceration or lesional inflammatory reaction to support the same.

The cut section shows a glistening surface of a gelatinous mass, unlike that of fibromas or other solid cellular tumors. This appearance mimics that of myxomas. Based on the appearance of the cut surface at grossing, the differential diagnosis of OFM is odontogenic myxoma. The histopathology of odontogenic myxomas shows the loosely arranged stellate- cells with an intermingled moderate number of delicate reticulin fibers in a homogenous mucoid ground substance probably hyaluronic acid. OFM lacks this reticulin network. Also, an odontogenic myxoma is non-capsulated and may invade the surrounding tissue, unlike OFM which does not have the invading character. OFM presents as a peripheral soft tissue lesion[10]. Histopathology of Peripheral odontogenic myxoma shows mast cells,

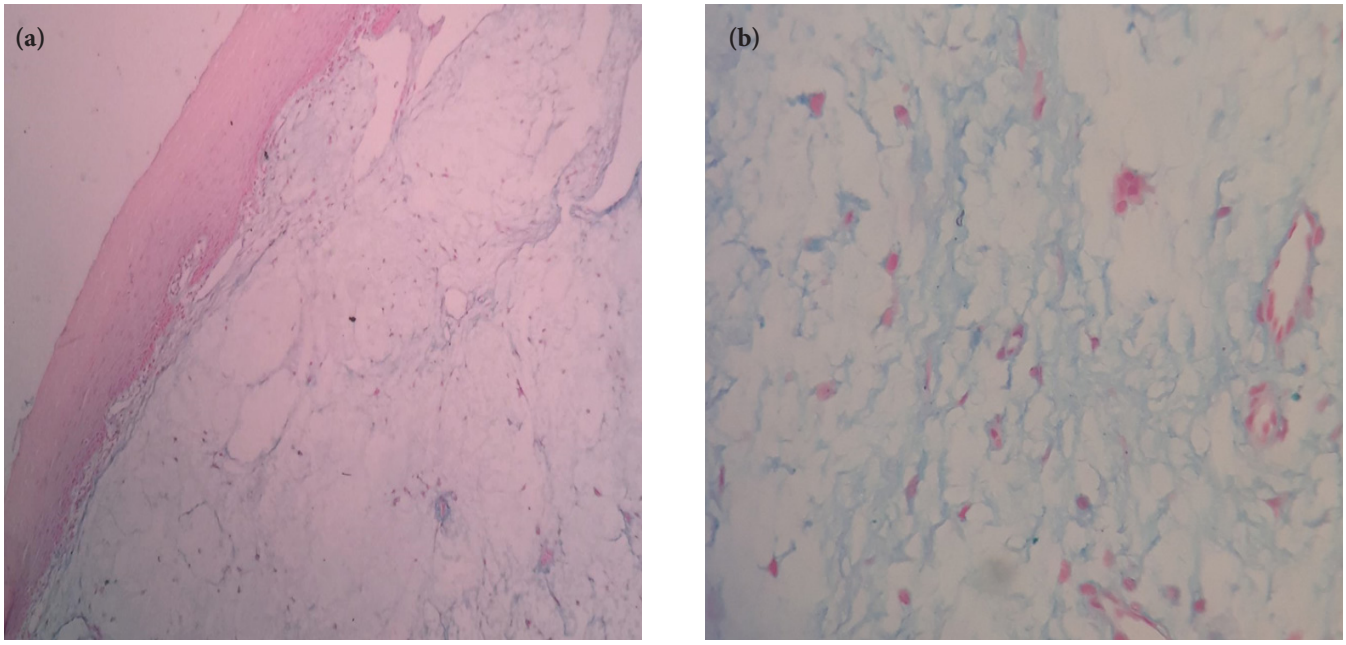



**Figure 5.** **(a)** Alcian blue stain positive myxoid stroma at 10X and 40X; **(b)** Alcian blue stain positive myxoid stroma at 10X and 40X.

odontogenic epithelial cell islands, and reticulin fibres[10,13] whereas central odontogenic myxoma presents clinically as a symptomless bony swelling with expanding cortices. The above-said histopathology was not seen in our study cases. The tissue showed positive results with an alcian blue stain and negative for reticulin stain. Hence, the diagnosis of OFM was confirmed. Oral Focal Mucinosis has not been reported to be associated with any systemic conditions so far. It is considered the oral counterpart of cutaneous focal mucinosis which too has not been reported to be associated with any systemic disease so additional laboratory investigations are not needed[14].

## 4. Conclusion

Based on the literature review it is reported that the clinical diagnosis of the lesion was not OFM at first clinical examination. This is due to the rarity of this lesion. Histological analysis has always been the basis for final diagnosis[15,16]. The treatment approach is the surgical excision of the lesion without any recurrence reports.

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