

**ORAL FOCAL MUCINOSIS OF GINGIVA: A RARE CASE REPORT**

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**ABSTRACT**

A cutaneous focal mucinosis (CFM) is a common benign neoplasm affecting any part of the body, but OFM is also being reported to occur intraorally. One such case of OFM on the lower gingiva in a 67 year old male is being reported. Histologically, it is characterized by focal myxoid area of connective tissue. The final diagnosis was completely depending upon the histopathological diagnosis. Surgical excision of the tumor was done and regular follows up of the patient for 11 months has not shown any evidence of recurrence or complication. The case which is presented in this article bring OFM to the clinician to consider as one of the differential diagnosis of myxoid lesions of the oral cavity.

**KEYWORDS:** OFM; Oral Focal Mucinosis; Myxoid Tumor; Gingiva; Soft Tissue Lesions.

**INTRODUCTION**

Oral Focal Mucinosis (OFM) is an oral counterpart of dermal lesion known as cutaneous focal mucinosis (CFM) or cutaneous myxoid cyst which is misdiagnosed as intraoral myxoma.(1,2) OFM is a uncommon clinicopathologic disease of unknown etiology, possibly resulting from overproduction of hyaluronic acid by fibroblasts (3). OFM is presented as an asymptomatic swelling that may be pedunculated or sessile. The gingiva was confirmed as the most common site for OFM, with predominance in females. Histologically characterized by a localized area of myxomatous connective tissue containing mucinous material surrounded by relatively dense collagenous connective tissues.(1) Here, we are reporting a case of OFM, since it is difficult to diagnose based on clinical feature because clinician tend to diagnosed as fibroma or pyogenic granuloma or epulis or granuloma. Thus, the histopathological diagnosis becomes important in these conditions and biopsy became the one of the important investigative procedure.

## **CASE REPORT**

A 67-year-old male presented with a painless swelling over lower gums since 8 months. On clinical examination, a single, soft to firm nodular mass about 1cm in dimension extending up to mucogingival junction. The growth appeared reddish in color, without any secondary changes. It was not tender, bleeding on probing. There was no radiographic changes were noted. Provisional diagnosis was made as peripheral giant cell granuloma.(Fig.1) Excisional biopsy was performed under local anaesthesia and specimen sent for histopathological analysis.

Fixation of the specimen was performed in 10% buffered formalin and embedded in the paraffin wax. 4- $\mu$  thickness of tissue sections were obtained from the paraffin-embedded block and stained with haematoxylin-eosin. Microscopically, the surface of the lesion was covered by stratified squamous keratinised epithelium and underlying connective tissue stroma shows localized area of myxoid material consists of stellate/spindle shaped fibroblasts interspersed between thin delicate collagen fiber and numerous small blood capillaries.(Fig.2,3) These histopathological features were suggestive of Oral focal mucinosis. Post-operatively the patient showed good healing and a regular follow up for 11 months since the diagnosis and treatment has been uneventful without any evidence of recurrence or complication.

## **DISCUSSION**

Solitary asymptomatic dome shaped skin nodules seen usually on face, trunk and extremities of CFM were described in 1966 by Jhonson and Helwig (3) Later OFM was first described by Tomich in 1974 who reported 8 cases as oral counterpart of CFM or cutaneous myxoid cyst and stated that most of the lesions were diagnosed as oral soft tissue myxomas instead of this entity.(1) (3).

Oral lesions of myxomatous nature are relatively rare which include nerve sheath myxoma, soft tissue myxoma, oral focal mucinosis and odontogenic myxomas. (4,5) Histopathologically, fibroblast-like cells and foamy cells diffusely increased in a well localized area of myxoid matrix, surrounded by collagenous fibrous connective tissue. Histochemically, fibroblast-like cells, foamy cells and myxoid matrix were revealed on metachromasia with toluidine blue at pH4.1 and pH 7.0. Johnson et al. attributed the pathogenesis of the cutaneous lesions to an overproduction of hyaluronic acid by fibroblast at expense of collagen production, replacing most of the collagen.(4,6) As the histochemical

characteristics of oral and cutaneous lesions are same, hence the same pathogenesis has been attributed to the oral lesion too (3). But the cause of this overproduction is unknown. (7) According to some authors trauma the etiology is unknown. (2) However, Reed et al. have proposed trauma as an etiologic factor but Tomich said trauma does not play any role in this pathogenesis. (3) Another possible etiologic factor of cervical external root resorption is mechanical pressure against the outer wall of the root, which is caused by tissue mass. The mechanism of root resorption is a sterile inflammatory process, initiated by the application of external force. The constant mechanical pressure can promote an external root resorption that can occasionally be seen in orthodontic therapy, cysts and benign tumors. Indentations were also noted in regions of reported lesions. (8) Alexandre SG et al suggested that in our orthodontic patient, the development of OFM was, probably, induced by bacterial plaque accumulation, after the placement of the orthodontic tube, associated with the orthodontic movement causing a local inflammatory process in the gingival area. (9) But in our case, the etiology was unknown.

OFM occurs predominantly in adults during the fourth and fifth decade of life, although it has been reported infrequently in children and adolescents. (1) The literature shows a female predilection has more predilection than male with a ratio of 2:1. (3) Gingiva is the most common site followed by hard palate. Out of 54 cases reported till date, 35 cases have been noted on the gingiva, 10 cases on the hard palate and 3 cases each on alveolar mucosa, buccal mucosa, and tongue, 2 cases on lips and 4 cases were not reported the site of lesion. (1) Thus, our case showed lesion was present in 67yr old male patient over mandibular gingiva.

Clinically these lesions present as sessile or pedunculated, painless nodular mass and are of same color as surrounding normal mucosa. Surface is typically smooth and non-ulcerated, although occasional cases exhibit a lobulated appearance. (5)(10) Size varies from few mms to 2 cm in diameter. The patient often has been aware of mass for many months or years before diagnosis is made (3). In our case there was a pink, sessile, well defined, smooth non ulcerated mass of about 1cm in dimension on lower gingiva in relation to 43. OFM has no distinctive clinical features and most often thought to be clinically as fibroma, pyogenic granuloma. Gingival epulis, mucocele, giant cell granuloma, minor salivary gland tumor were also considered in the differential diagnosis. A review of all reported cases show that it was never diagnosed clinically as 'oral focal mucinosis' (3,5, 7). It was always diagnosed by histopathological feature. Hence the oral biopsy is considered as most important diagnostic

aid for the diagnosis.(8) Radiographic examination of the lesion in our cases did not reveal any abnormality.

Microscopic examination of OFM showed a well localized loose, myxomatous connective tissue stroma composed of pale eosinophilic myxoid stroma representing an overproduction of hyaluronic acid with stellate shaped fibroblasts were noted. Deeper stroma showed spindle fibroblasts, thin bundles of collagen fibers and numerous small blood capillaries along with diffuse infiltration of mixed inflammatory cells consisting of mainly lymphocytes and plasma cells. (3) Surface epithelium showed normal with flattening of rete ridges. All these features were showed in our case of OFM.

The histopathological differential diagnosis of oral focal mucinosis includes, inflammatory fibroepithelial hyperplasia, nerve sheath myxoma, and myxoma.(8) Similar myxoid areas may be found in inflammatory fibroepithelial hyperplasia, but unlike oral focal mucinosis, these are accompanied by inflammation and fibrosis elsewhere. Nerve sheath myxoma is characterized by a whorled arrangement of tumor cells in an organoid, multinodular, or lobular structure. 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. Myxoma may present as an infiltrative growth pattern, while focal mucinosis usually manifests as a localized area of myxomatous connective tissue. Small pools of mucinous material are a feature in many cases of focal mucinosis, but are not present in myxomas. (5,8) The literature stated that conservative surgical excision of the OFM has been suggested as the treatment of choice, without any recurrence or complication. (6,8)

Even though the occurrence of lesion is infrequent, it is recommended that OFM should be considered as a differential diagnosis of oral soft tissue lesions

## **CONCLUSION**

Oral focal mucinosis is a benign tumor of a mesenchymal derived lesion composed predominantly of fibroblasts. Based on clinical feature, it is highly impossible to diagnosis OFM. The histopathological feature has a vital role to arrive at definitive diagnosis. Hence, the lesion demonstrates the importance of biopsy and histopathologic examination to arrive at final diagnosis.

**FIGURE LEGENDS**

Fig 1: Photomicrograph of the oral focal mucinosis showing nodular mass over mandibular gingiva.

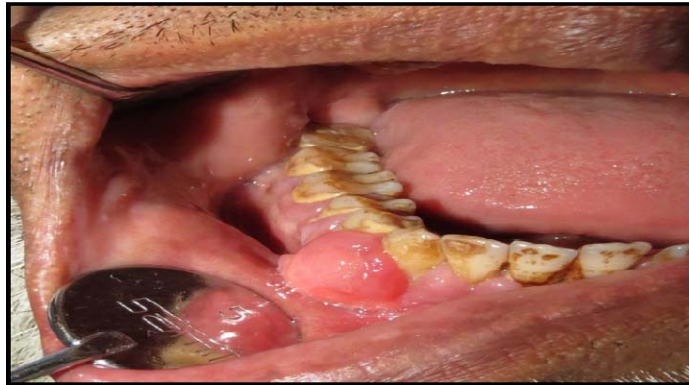


Fig 2: Photomicrograph of the oral focal mucinosis showing surface epithelium and well defined myxomatous area in connective tissue (hematoxylin and eosin, magnification- 100x).

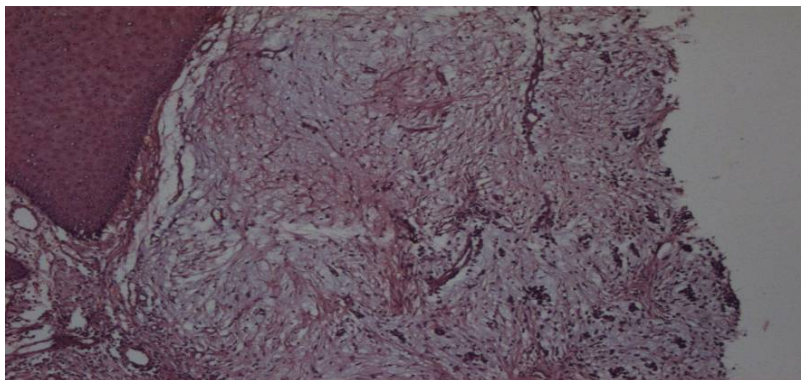
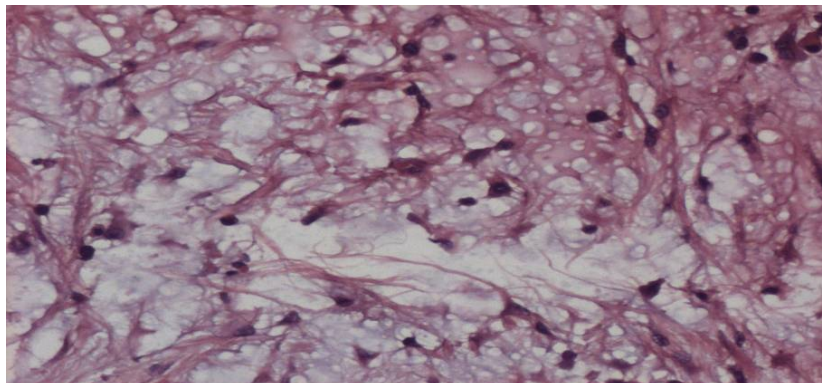


Fig 3: Photomicrograph of oral focal mucinosis showing stellate/spindle shaped fibroblasts interspersed between thin delicate collagen fiber (hematoxylin and eosin, magnification- 400x).



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