Case Report

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Dentistry Section

A Case of Oral Focal Mucinosis on Hard Palate: A Lesion Derived from the Periodontal Ligament

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ABSTRACT

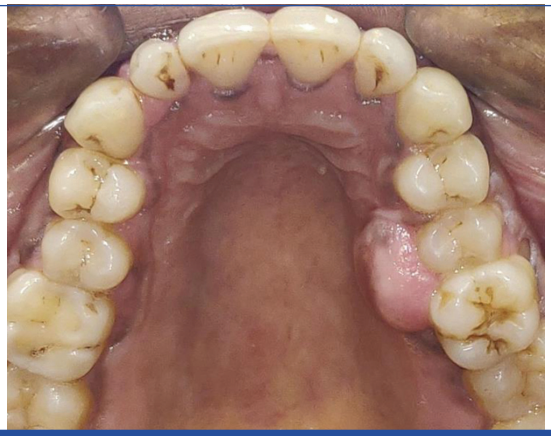
Oral Focal Mucinosis (OFM) manifests on the gingiva as a painless mass, asymptomatic except for the presence of a fibrous mass, and is of the same colour as the surrounding mucosa. It is commonly reported on the gingiva. Histologically, it is characterised by focal myxoid degeneration of connective tissue. Its diagnosis seldom can be made clinically and predominantly relies on histopathological analysis. This case report is about a 40-year-old female who had a gradually increasing swelling on the palate in relation to upper right posterior teeth, for 4-5 months. The lesion was excised and sent for histopathological and immuno-histochemical analysis. There was presence of foci of loose mucinous connective tissue stroma interspersed within dense connective tissue stroma. The mucinous stroma showed numerous stellate and spindle-shaped fibroblasts evident between thin collagen fibres with evidence of separation of collagen fibres.

Keywords: Diagnosis, Gingiva, Oral lesion, Palate, Pathology

CASE REPORT

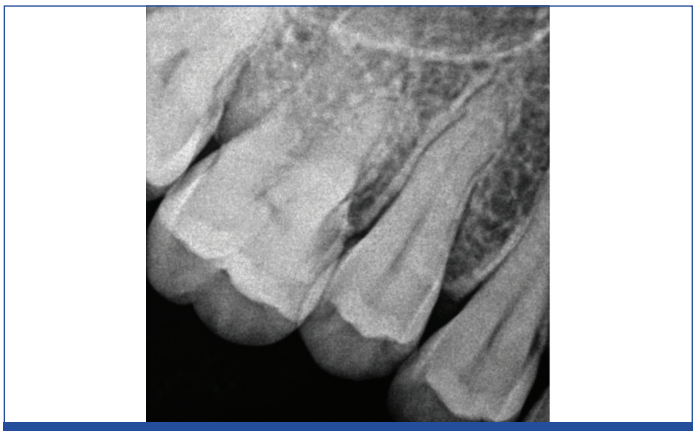
A 40-year-old female patient reported to the Department of Periodontics and Oral Implantology with a chief complaint of a swelling on the palate in relation to upper right posterior teeth for 4-5 months, which was gradually increasing in size. The patient had a history of controlled diabetes and hypertension for the past five years and was under medication. The patient had no adverse habits such as smoking etc.

On clinical examination, there was a pedunculated mass on the right posterior aspect of the hard palate in relation to the distal aspect of 14 extending to mesial aspect of 16 [Table/Fig-1]. On examination, the lesion was seen to be approximately 1.5×1.2 cm in size surrounding the palatal aspects of 15 and 16, with a duration of two months, normochromic and smooth surface. On palpation, the lesion was firm in consistency, non tender without any history of bleeding. Based on the history and clinical examination, a provisional diagnosis of fibroma was made.



[Table/Fig-1]: Clinical presentation of the lesion on the gingiva of the hard palate.

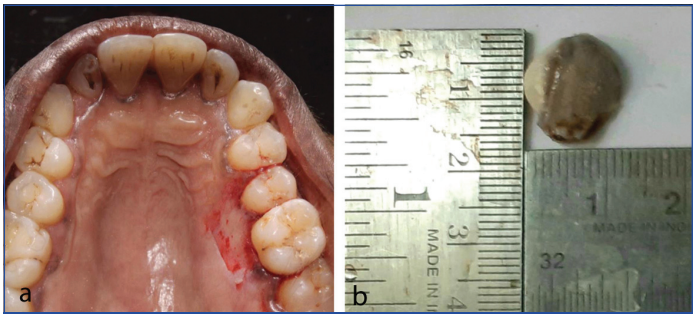
Laboratory investigations were within normal limits [Hb% (13.5 g/dL), bleeding time (1 min 36 sec) and clotting time (6 min 20 sec)]. A slightly increased level of random blood sugar of 180 mg/dL was observed. The intraoral periapical radiograph revealed no bone loss in relation to 14 and 16, lamina dura was intact and a slight widening of the Periodontal Ligament (PDL) space was seen [Table/Fig-2].



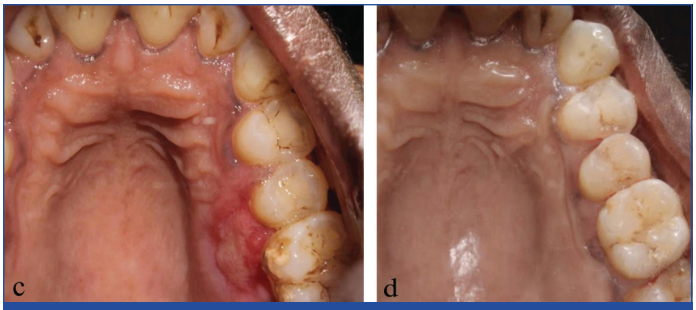
[Table/Fig-2]: Intraoral periapical radiograph depicts slight PDL widening in relation to 15 region with no evident bone loss in the 15, 16 regions.
PDL: Periodontal ligament

An informed written and video consent was taken prior to any intervention, including non surgical therapy i.e., scaling, root planing, surgical excision of the lesion and consent to perform pathological analysis on the excised tissue. To eliminate local irritants, thorough scaling and root planing was performed.

The lesion was excised in its entirety under local anaesthesia using a no. 15 BP blade [Table/Fig-3]. The excised lesion was preserved in 10% formalin before being sent for histological analysis [Table/Fig-3a]. After a week, the patient was recalled and uneventful healing by secondary intention was observed [Table/Fig-3b].

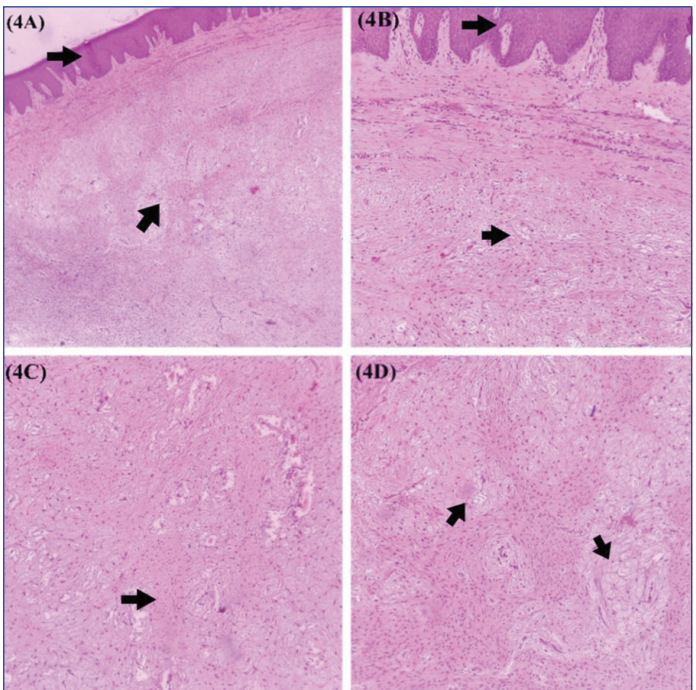


[Table/Fig-3a]: Excision of the lesion at the palatal site.
[Table/Fig-3b]: Macroscopic appearance of the lesion; the excised lesion measured about 1.5x1.4 cm in length and diameter.



[Table/Fig-3c]: Postoperative one week healing.
[Table/Fig-3d]: Three month postoperative healing.

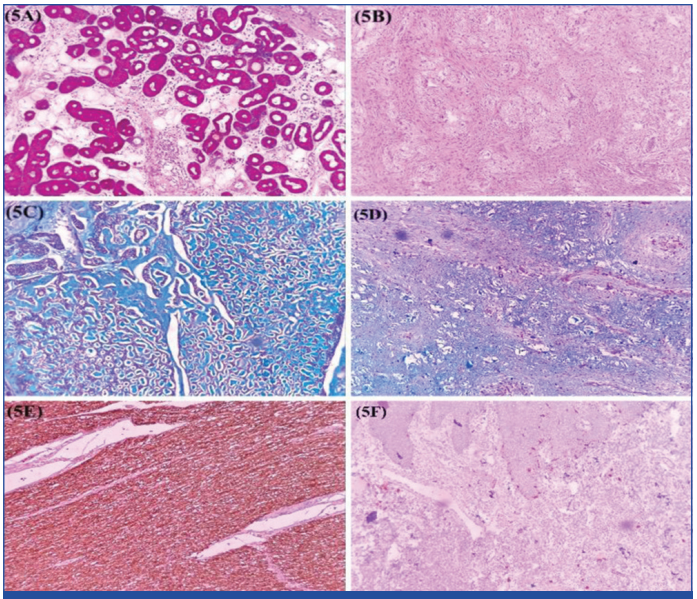
Histopathological examination identified foci of loose mucinous connective tissue stroma interspersed within dense connective tissue stroma [Table/Fig-4a]. The mucinous stroma showed numerous stellate and spindle-shaped fibroblasts evident between thin collagen fibres with evidence of separation of collagen fibres [Table/Fig-4b]. Minimal chronic inflammatory cell infiltrate, predominantly plasma cells and lymphocytes with moderate vascularity was evident. The overlying epithelium was parakeratinised, stratified squamous epithelium with confluent rete ridges with few areas showing evidence of pseudoepitheliomatous hyperplasia [Table/Fig-4c,d]. Alcian blue and Periodic acid-Schiff (PAS) staining of the mucinous substrate of the tissue demonstrated a positive reaction with Alcian blue [Table/Fig-5a,b] and a PAS-negative reaction [Table/Fig-5c,d]. S-100 (performed to exclude myxoid peripheral nerve sheath tumours) positive cells were not identified in immunohistochemistry [Table/Fig-5e,f]. Correlating clinically, a histopathological diagnosis of OFM was made. Patient was recalled after one week to assess the postoperative healing [Table/Fig-3c] and follow-up was done till three months.



[Table/Fig-4]: (A,B) Showing foci of loose mucinous connective tissue stroma interspersed within dense connective tissue stroma (10x magnification). The overlying epithelium was parakeratinised stratified squamous epithelium with confluent rete ridges with few areas showing evidence of pseudoepitheliomatous hyperplasia. (C,D) Showing the mucinous stroma with numerous stellate and spindle shaped fibroblasts evident between thin collagen fibres with evidence of separation of collagen fibres. Minimal inflammatory cell infiltrate, predominantly plasma cells and lymphocytes also present (40x magnification).

DISCUSSION

The OFM is a tumour-like growth, although rarely reported, occurs predominantly on the gingiva, manifesting as localised gingival enlargement arising due to either inflammatory or non inflammatory aetiology [1]. It was first described in 1974 by Tomich CE who



[Table/Fig-5]: Photomicrograph showing various special stain reactions and immunohistochemical reactions (40x magnification). (A,B) Showing PAS stain. A) Demonstrating positive reaction with PAS in a control slide; B) Demonstrating negative reaction with PAS in OFM. (C,D) demonstrating Alcian blue stain reaction; c) Showing positive reaction in control; D) Showing positive reaction in OFM; E) Showing positivity for S-100 marker; and F) Showing a non specific reaction with S-100 marker in OFM; OFM: Oral Focal Mucinosis.

reported eight cases with no established aetiology [2]. Clinically, it is described as a sessile or pedunculated mass, the same colour as the surrounding mucosa on the gingiva, histologically a localised myxoid degeneration of connective tissue is seen. OFM primarily affects adults in their fourth and fifth decades of life [3]. The diagnosis cannot be made clinically and is mostly based on histological investigation.

Oral soft tissue myxomas develop as a result of local hyaluronic acid overproduction by fibroblasts [4]. The aetiology of OFM is unknown and could possibly form due to overproduction of hyaluronic acid by fibroblasts [5]. The gingiva is the most common site for OFM, with predominance in females, although some reports also exist on males at sites such as the lips, buccal mucosa and even the tongue [2]. Clinically, the lesion appears as asymptomatic, ovoid raised lesions, presented as a pedunculated or sessile swelling [6,7].

Clinically, the differential diagnosis of OFM could be inflammatory, which includes fibrous hyperplasia, peripheral giant cell granuloma, Epulis Fissuratum (EF), pyogenic granuloma or originating from tumours such as peripheral fibroma, peripheral odontogenic myxoma, peripheral ameloblastoma, malignant and metastatic tumours in gingiva or non plaque induced lesions such as an irritation fibroma [8,9]. This case report highlights a presentation of OFM on the hard palate and the associated possible aetiology of this lesion in this discussion.

Histopathologically, non encapsulated areas of fibrous connective tissue with widely dispersed collagen fibres interspersed with stellate or spindle-shaped fibroblasts are seen [7]. The interface between the myxoid stroma and the connective tissue is usually well demarcated and the fibroblasts contain varying amounts of cytoplasm. Special stains such as Alcian blue and PAS help in the diagnosis. OFM shows a positive reaction with Alcian blue and a negative reaction with PAS, revealing the presence of abundant amounts of acid mucin. The mucinous material observed can be suggestive of the excessive production of hyaluronic acid by the fibroblasts in the connective tissue. OFM shows a non specific reaction with S-100 marker in immunohistochemistry, suggesting the absence of neural involvement [8]. A clear understanding of these clinicopathological entities is needed to provide the appropriate treatment as these lesions require only conservative

excision. Recurrence has not been reported in the literature after the complete excision of the lesion [10-12].

"Overproduction of hyaluronic acid by the fibroblasts at the expense of collagen production" was described as the possible aetiology of the lesion by Tomich CE. However, the reason behind this overproduction is not well understood [2]. Matrix Metalloproteinases (MMPs) play an important role in resorbing both the bone and matrix [13]. All active MMPs are inhibited by Tissue Inhibitors of Metalloproteinases (TIMPs) [13,14]. TIMPs are secreted as endogenous inhibitors and TIMP bind tightly to active MMP in a 1:1 ratio. Therefore, connective tissue turnover is prevented if TIMP levels exceed those of the active enzyme [15]. The post-translational regulation of MMP activity depends on the counteracting interactions between MMP and inhibitory TIMP. Thus, the regulation of MMP or TIMP at the PDL- bone interface seems crucial for the remodelling process [16]. A study found that Hyaluronan (HA), a large glycosaminoglycan composed of repeating disaccharides of D-glucuronic acid and N-acetylglucosamine, is one of the major components of joint fluid and connective tissues. This is a structural component that helps maintain the extracellular matrix architecture by joint lubrication and can be a biological inhibitor of joint degradation [17]. HA could inhibit Interleukin-6 (IL-6)-induced MMP production in human chondrocytes. The mechanism of overproduction of hyaluronic acid at that particular site could be a protective one, according to studies that suggested HA inhibits the production of MMP and the expression of Receptor Activator of Nuclear Factor Kappa-beta Ligand (RANKL). The mechanism of action of HA on these proteins remains to be elucidated. HA is known to associate with several cell-surface molecules, such as CD44 and Intercellular Adhesion Molecule-1 (ICAM-1) [18]. There is controversy as to whether local trauma plays a role in the aetiology of OFM. Tomich CE said it does not play a role while Gnepp DR et al., documented trauma as a contributing factor [19]. It is suggested that most of these lesions actually represent myxomatous or "mucinous" change in a preexisting fibrous lesion [8,20].

From a histological aspect, OFM needs to be differentiated from nerve sheath myxoma and peripheral odontogenic myxoma. The nerve sheath myxoma is more circumscribed, has fibrous septa between multiple myxoid nodules and has more plump stromal cells. Peripheral odontogenic myxoma shows presence of mast cells, increased reticular fibres and islands of odontogenic epithelium [7,21]. Central odontogenic myxoma is a symptomless bony swelling expanding cortices from within, whereas OFM is a peripheral soft tissue lesion [21,22]. None of these histologic features were found in our case. Hence, diagnosis of OFM was confirmed. However, none of the articles reported a clinical diagnosis of OFM at its first presentation. Histological analysis is always the basis for diagnosis [23]. Thus, due to the rarity of its occurrence, a preoperative diagnosis is almost impossible [10,11]. Nevertheless, surgical excision of the lesion seems to be a safe approach without any reports of recurrence.

CONCLUSION(S)

The OFM is a rare soft tissue lesion of the oral cavity, seen most often on the gingiva. In the above case report the lesion was reported, diagnosed and managed by a multidisciplinary approach between the pathologist and the clinician. Misdiagnosis of such lesions should be avoided and managed by detailed histopathological information, thorough conservative surgical excision and postoperative management. Recurrence, although rare, must be monitored by diligent follow-up appointments.

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