Oral Focal Mucinosis: A Painful Lesion of the Palate

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Abstract: Oral focal mucinosis (OFM) is a rare disease of unknown etiology, in which the connective tissue undergoes a focal myxoid degeneration. Its pathogenesis is related to overproduction of hyaluronic acid by fibroblasts during the collagen production, resulting in focal myxoid degeneration. It has no distinctive features and diagnosis depends on histological analysis. This paper reports a case of a 30 years old female with a lesion growth and progressive pain in the palate, after performing dental treatment and discusses the clinical characteristics and differential diagnosis of myxomatous lesions of the oral cavity.

Since oral focal mucinosis has no distinguishing clinical features and the diagnostic is based on a histopathological examination, we ratify the importance of this procedure to confirm the diagnosis.

This article is based on literature review and reports a case of oral focal mucinosis, its clinical and surgical outcome and demonstrates the importance of biopsy and pathological examinations in differential diagnosis of nodular masses in the oral cavity.

Keywords: Focal mucinosis, polyp, oral tumor.

CLINICAL RELEVANCE

This article is based on literature review and reports a case of oral focal mucinosis, its clinical and surgical outcome and demonstrates the importance of biopsy and pathological examinations in differential diagnosis of nodular masses in the oral cavity.

INTRODUCTION

The oral focal mucinosis (OFM) is a rare disease of unknown etiology in which the connective tissue is found in the form of focal myxoid degeneration [1]. It was first described in 1974 by Tomich [1]. Myxoid degeneration involves the presence of a jelly intratumoral focus containing mucopolysaccharide cointaning hyaluronic acid. It is suggested that oral lesions were similar to focal skin mucinosis and / or skin myxoid cyst [2-4].

Clinically oral focal mucinosis can be found in the form of small nodules elevated up to 2 centimeters in diameter, rounded, asymptomatic, pinkish in color, similar to adjacent normal mucosa and seldom presents secondary ulceration [4, 5].

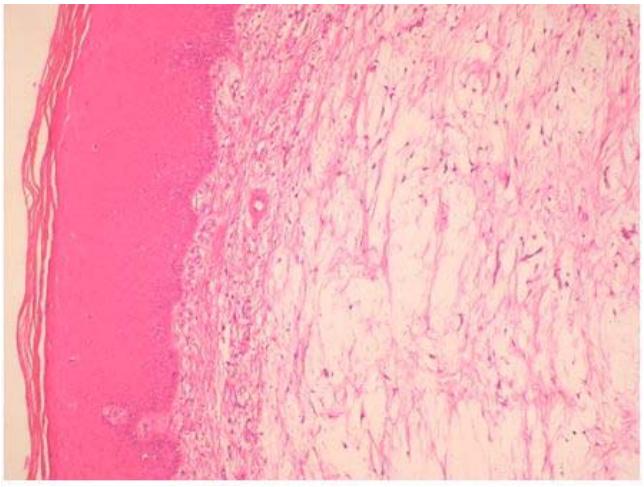

Figure 1: Lamina propria of the oral mucosa with well-defined myxomatous area.

Its pathogenesis is unclear, but suggests an increase in the production of hyaluronic acid by fibroblasts in collagen production expense [1]. Most cases of OFM are described as lesions on keratinized mucosa as the gum or palate, with 80% of lesions

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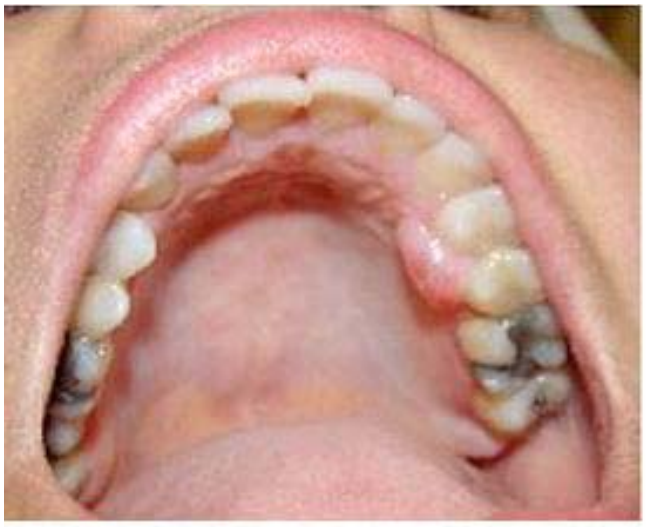

Figure 2: Nodules resembling normal mucosa.

developing at the gum and the remaining at the palate [1]. However, some cases of OFM were reported also at the tongue [5]. Due to their clinical characteristics are very similar to the other most common lesions of the oral cavity, its clinical diagnosis becomes difficult. Histologically, the lesions consist of OFM well located areas of myxomatous connective tissue surrounded by dense fibrous connective tissue. The myxomatous zone consists of collagen fibrils widely separated intercalated with starry fibroblasts [6]. Radiologically the OFM can be seen as a radiolucent homogeneous and well defined lesion [5].

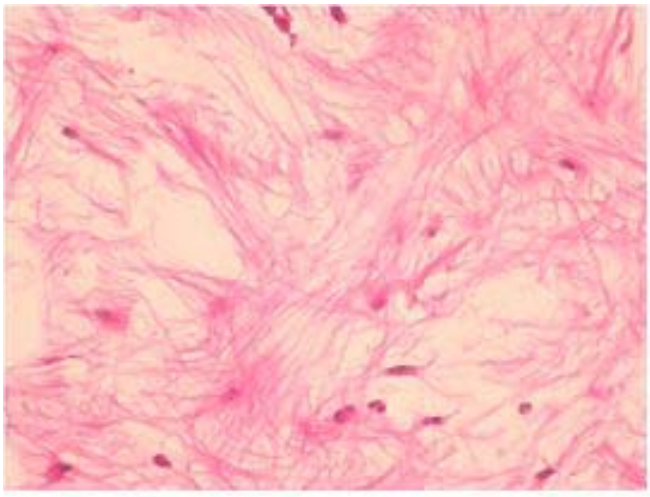

Figure 3: Fusiform and stellate fibroblasts with small amount of thin collagen fibers intermingled.

CASE REPORT

A thirty-year-old female patient was treated at the dental clinic and Medical Laser Multidisciplinary Unit of the Clinical Hospital - CH / UNICAMP due to lesion growth and progressive pain at the palate, after performing dental treatment years ago (Figure **2**).

According to the patient, the lesion waxed and waned in size, causing nuisance to brush and floss teeth, presenting sporadic bleeding in small quantities. She also reported that sporadically injury when chewing more consistent food. There was no history of medication use, personal or family pathologies or previous smoking and drinking. Physical examination of the oral cavity showed lesion between 24 and 25 teeth (left premolars) at the left palatine region, nodular, with a fibroelastic consistency, pale pink, pedicled with size of 1 cm in diameter, without ulcerated areas. The teeth and gums were in a good state of hygiene and conservation.

A week after the consultation, an excisional biopsy of the lesion with a scalpel blade and subsequent application of CO_2 laser vaporization of its base were held at Unity Multidisciplinary Medical Laser. The material was sent for pathological examination, as shown in Figure **4**, and the result was compatible with oral focal mucinosis.

The patient attended for her first return to get the biopsy results and subsequently did follow up. She attended for reevaluation every six months, without recurrence of the lesion since its excision a year ago.

DISCUSSION

The oral focal mucinosis was first described in 1974 in a series of eight cases, but similar lesions were noted previously by Johnson and Helwig in 1966 that described isolated dome shaped skin lesions, asymptomatic, usually seen on the trunk, extremities and face [3]. It usually can be found as a pinkish small nodules and can also present as a skin lesion dome shaped, appearing on the trunk, face and extremities. Histologically, it is characterized by a mucinous accumulation interspersed with starry fibroblasts [3].

Myxomatous oral lesions are relatively rare and include: myxoma of the nerve sheath myxoma of soft tissue, oral focal mucinosis and odontogenic myxomas. The latter is a bone neoplasm, but can occur in soft tissue when it pierces the cortex [3]. Oral focal mucinosis and soft tissue myxoma of the oral cavity are distinguished by the presence of reticulin fibers and its relation to the surrounding connective tissue [4].

The differential diagnoses include: inflammatory lesions (gingivitis, fibrous hyperplasia, peripheral giant

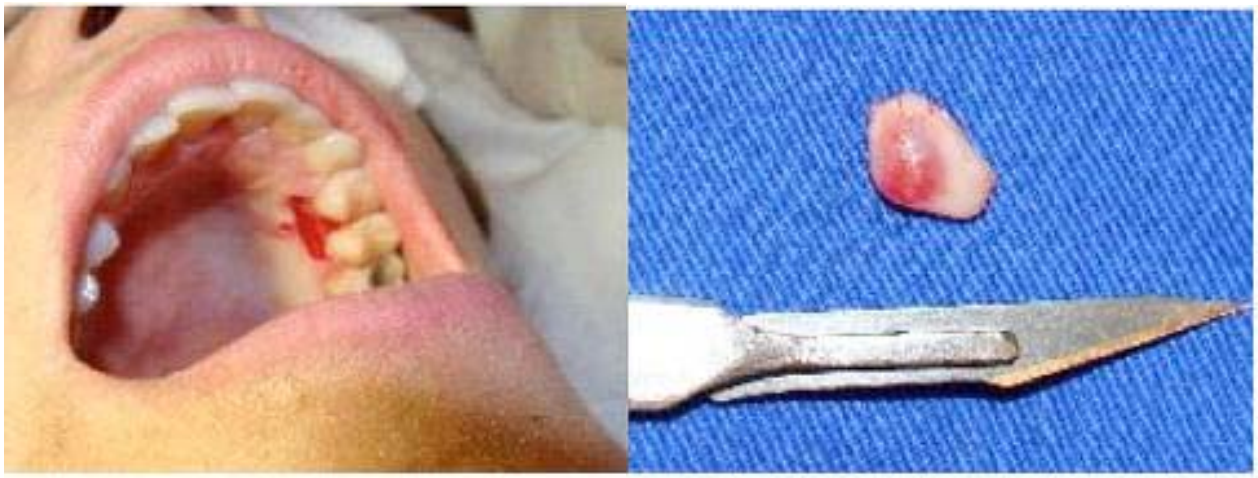

Figure 4: Appearance after excisional biopsy and the material sent for analysis.

cell granuloma, epulis fissuratum, pyogenic granuloma), tumors (peripheral fibroma and peripheral ossifying fibroma), benign primary lesions (peripheral ameloblastoma and peripheral odontogenic fibroma) and also some malignant or metastatic gum lesions. We must emphasize that in most of the lesions the preoperative diagnosis is almost impossible because of its rarity and its similarity to other lesions of various etiologies. Treatment suitable for cases of OFM includes surgical excision of the lesion and there is no known case of relapse [1].

The etiology of focal mucinosis is unknown. According to some authors, trauma does not appear to be part of the pathogenesis of the disease. However, Reed *et al*. proposed the trauma as an etiological factor [2]. Another possible etiologic factor of cervical external root resorption of is mechanical pressure against the outer wall of the root, which is caused by tissue mass [6]. The mechanism of root resorption is a sterile inflammatory process, initiated by the application of external force [5]. The constant mechanical pressure can promote an external root resorption that can occasionally be seen in orthodontic therapy, cysts and benign tumors [5]. Indentations were also noted in regions of reported lesions. Johnson *et al*. attributed the pathogenesis of skin lesions to an overproduction of hyaluronic acid by fibroblasts, increasing collagen production, replacing it at the tissue [3].

OFM is seen mostly in adults with ages ranging from 16 to 68 years, with only one reported case of a girl aged 4 years old. The literature shows a predilection for females, with 31 cases reported in women and 18 cases in men. The mucosa that is in direct contact with the bone is particularly affected and the gum is the most common site, followed by the hard palate [3].

Clinically, these lesions present as painless masses, sessile or pedunculated, with the same color of the adjacent normal mucosa. The surface is typically flat, non-ulcerated, however, some cases showed a lobulated surface. The size varies from a few millimeters up to 2 centimeters in diameter.

OFM has no distinguishing clinical features and is often seen or considered as fibroma, pyogenic granuloma, mucocele or similar injuries [3]. A review of all cases shows that there has never been a clinical diagnosis of "oral focal mucinosis" and that the histological characteristics are the basis for the diagnosis, demonstrating the need for biopsy to be performed well and the importance of pathological examination in differential diagnosis of nodular masses in the oral cavity to complete the diagnosis [3].

We respected the guidelines of the Medical Ethics Committee of our institution.

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