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Oral focal mucinosis of the hard palate and gingiva

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ABSTRACT

Oral focal mucinosis (OFM) is an uncommon, asymptomatic, submucosal, slow-growing nodule representing a counterpart of the cutaneous focal mucinosis (CFM). OFM has a female predilection with the highest prevalence in the fifth decade of life. About 68% of OFMs occur in the gingiva and 14% in the palate. We present the case of a 41-year-old woman presenting a progressively growing mass on the palate, since the last 8 months. The diagnostic workup led to the diagnosis of an unusual OFM with the clinical presentation involving the gingiva and hard palate. This case report discusses the clinical and histopathological differential diagnosis.

Keywords
Mucinosis; Soft Tissue Injuries; Gingiva; Palate, Hard.

INTRODUCTION

OFM is an uncommon, submucosal, tumor-like mass, and a counterpart of CFM.[1] CFM and OFM have been regarded as non-neoplastic reactive lesions,[2] having possible correlation with trauma.[3] The etiology remains unknown; however, it has been correlated to the overproduction of hyaluronic acid by fibroblasts.[4,5]

There is some difference in the demographic features between CFM and OFM. As compared to solitary CFM, which is more prevalent in males,[2] OFM shows a predominance among women.[3] The mean age of the patients with CFM and OFM are similar, commonly found in the fifth decade of life.[2,3] The time of the evolution of the lesion from diagnosis to complete excision in both entities is variable, ranging from months to years.[6] Usually, both CFM and OFM lesions appear as a firm but loose nodule, not adhering to the deeper structures and with the same color of the skin or mucosa.[1] CFMs commonly affect the extremities, head, neck, and trunk,[2] whereas, OFMs are frequently seen in the gingiva (68%) and palate (14%).[3] The advisable treatment for CFM and OFM is complete surgical excision;[2,3,6] recurrences are unusual.[1,2,7]

The present case report shows an OFM with an unusual clinical aspect, with respect to its proportions involving the gingiva and the hard palate, followed by a discussion on its clinical and histopathological differential diagnosis.

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CASE REPORT

A 41-year-old woman was referred for the investigation of a progressively growing, large-sized lesion involving the gingiva and the palate, since the last 8 months. The patient gave a history of an oral surgery at the site of the current lesion, 8 years back; however, the patient was unaware of the histopathological diagnosis. Therefore, we could not assert if it was a recurrent or *de novo* lesion. There was no relevant medical history or any extraoral signs. The intraoral examination revealed a well-defined, lobulated nodule covered by normal mucosa, measuring 3.0 cm, extending from the palatal gingiva of the right first and second molars to the hard palate (Figure 1). The lesion was firm on palpation, painless and presented slight mobility. Additionally, there was a sign of mild trauma in the premolar region, caused by a removable partial prosthesis (Figure 1). The panoramic x-ray examination was normal.

An incisional biopsy was performed. The specimen collected was rubbery, soft to moderately firm, and white-gray colored. The histopathological examination showed a well-delimited but non-encapsulated lesion, characterized by a myxomatous connective tissue presenting spindle-shaped fibroblasts interspersed with short bundles of collagen (Figure 2A). An Alcian Blue staining (pH = 2.5) showed strong staining of the myxoid areas, suggestive of hyaluronic acid and was negative in the dense connective tissue areas (Figure 2B, 2C and 2D). Immunohistochemical reaction for S100 protein was negative, ruling out neural tumors.

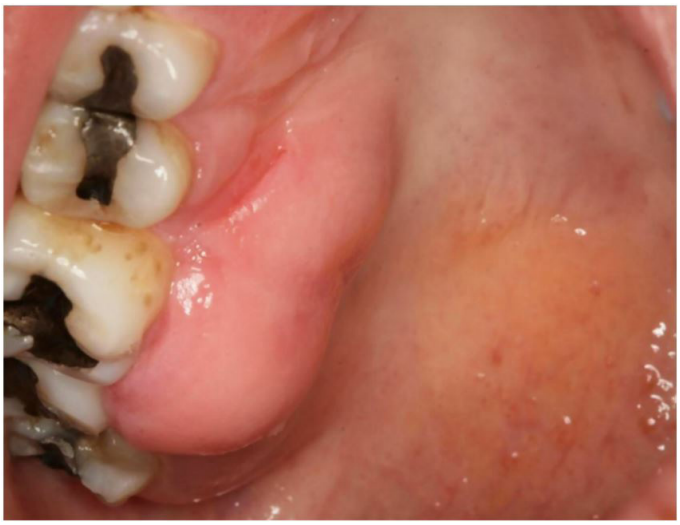

Figure 1. Intraoral examination showing a well-defined, lobulated mass covered by a smooth and superficial mucosa, measuring 3.0 cm, extending from the palatal gingiva to the hard palate.

As per the clinical, histopathological, and immunohistochemical features, the case was diagnosed as OFM, following which, the lesion was excised (Figure 3A). The patient showed no signs of recurrence in the follow-up after 8 months (Figure 3B).

DISCUSSION

OFM presents as a local gingival overgrowth, with fibroma, gingival epulis, pyogenic granuloma, and oral mucocele as the relevant differential diagnosis.[1,8,9]

In our case, the location and the size of the lesion did not immediately favor such clinical hypotheses.[9] A significant part of the lesion seemed to be correlated to the gingiva, which could support reactive injuries. However, the tumor also had a great extension toward the hard palate, favoring the hypothesis of salivary gland tumors.

The three most common reactive lesions of the gingiva are peripheral ossifying fibroma (POF), pyogenic granuloma (PG), and peripheral giant cell granuloma (PGCG).

POF is a fibro-osseous reactive lesion, exclusive to the gingiva, though it could expand to the adjacent structures, depending on the size.[10] Clinically, it is a slow-growing, nodular mass, with a smooth surface and usually presenting the same color as the surrounding normal mucosa.[11,12] Ulceration and erythematous areas may be present.[13] Although POF may be diagnosed at any age, it commonly occurs in the second decade of life. POF is more prevalent in women[14] and has a higher chance of recurrence as compared to PG and PGCG.[12]

PG occurs both on the skin and mucosa.[15] When it occurs on the mucosa, it may present as a sessile or pedunculated, reddish-purple nodule, with or without ulceration and having a natural tendency to bleeding[15,16], distinguishing it from OFM.

The clinical appearance of PGCG is very similar to POF, which also develops exclusively on the gingiva/alveolar mucosa.[14,17] The lesions tend to be less reddish than PG and more similar to OFM. A higher incidence is seen among the females aged 30-40 years

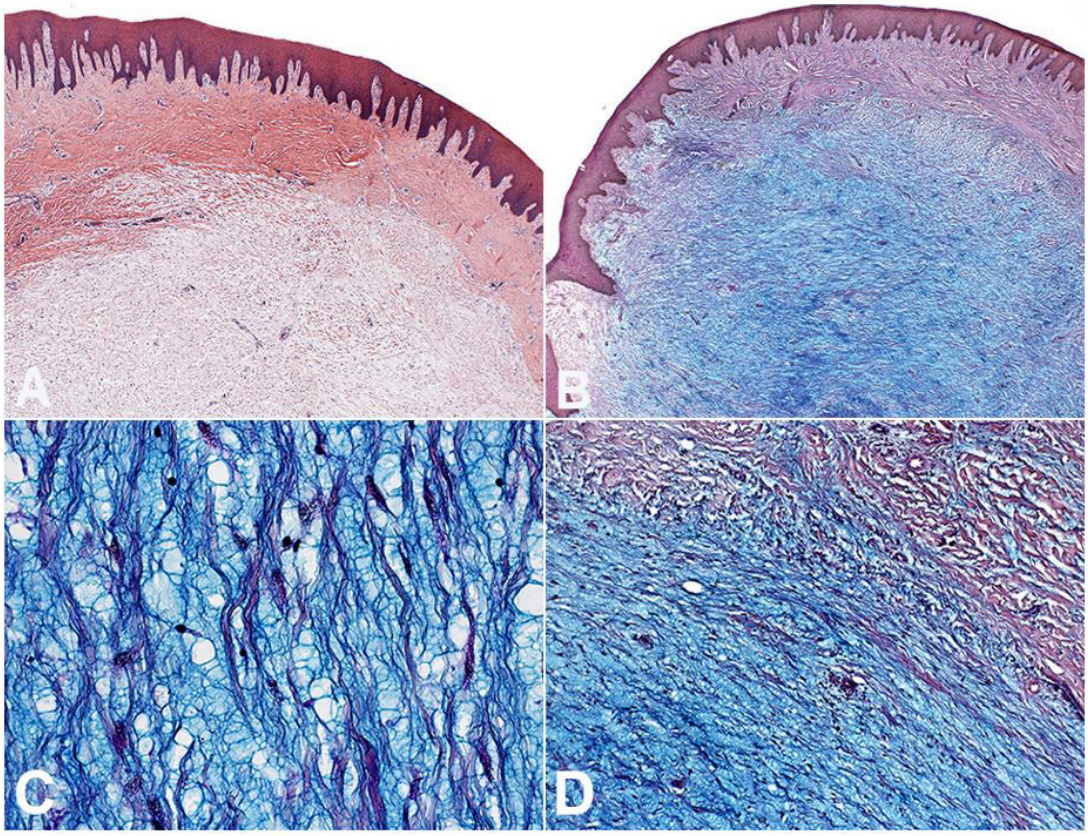

Figure 2. Photomicrography of the tumor biopsy showing: **A –** A well-circumscribed, non-encapsulated lesion, characterized by a myxomatous connective tissue presenting spindle-shaped fibroblasts interspersed with short bundles collagen (H&E; 250x); **B –** Alcian blue stain, pH = 2.5, 250x); **C –** Alcian blue stain, pH = 2,5 400x; **D –** Alcian blue satin pH 2,5, 400x myxomatous area-connective tissue interface.

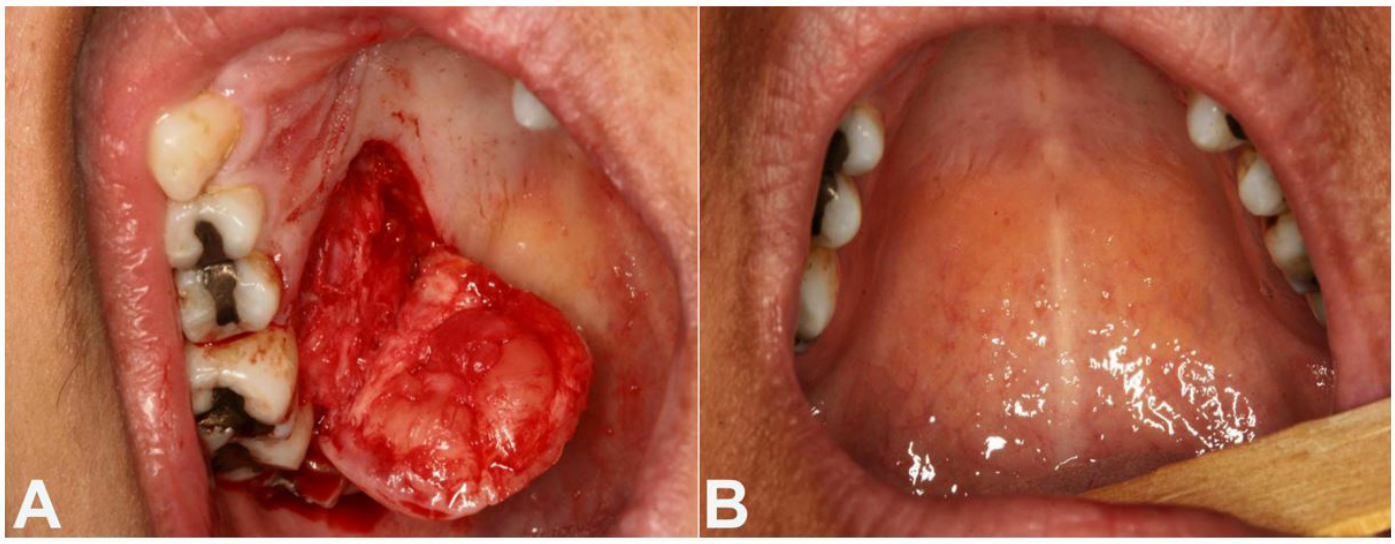

Figure 3. A – Gross view of the intra-operative oral focal mucinosis (OFM) excision; **B –** 8 months' post-operative oral examination.

and a superficial "cupping" representing alveolar bone resorption is often seen in the radiographs;[17] this aspect is not found in OFM.[9,18]

Differential diagnosis of lesions of the palate includes salivary gland tumors and mesenchymal tumors. Pleomorphic adenoma (PA) is the most common tumor among the benign minor salivary glands. Usually, it appears as a painless, firm, and slow-growing mass with a smooth surface, often lobulated on the posterior lateral of the palate. The lobulated appearance seen in

the present case is not a common finding in OFM and it usually appears as a dome-shaped nodule.[9] The peak incidence of PA is around 40-50 years of age, with a slight female predominance. Generally, PA involving the palate does not reach large dimensions as seen in parotid gland due to the impairment of speech and swallowing.[19] The recurrence of PA usually occurs when an incomplete excision is performed causing violation of the tumor pseudocapsule.[20] Recurrences in OFM are rare.[1,6]

Mesenchymal tumors such as neurofibroma and schwannoma should also be added to the differential diagnosis. Among these, schwannoma (neurilemoma) is a benign neural tumor arising from the Schwann cells of any peripheral nerve. Similar to the neurofibroma, it can occur as a solitary lesion or associated with type II neurofibromatosis. The schwannoma presents as an isolated, slow-growing, well-demarcated, encapsulated, and usually asymptomatic tumor,[21] occurring mostly in the 4th decade of life, with no gender predominance.[21] Intraorally, the most common affected site is the tongue, followed by the palate, floor of the mouth, buccal mucosa, gingiva, lips, and vestibular mucosa.[22] The neurofibroma is a benign peripheral nerve sheath tumor characterized by the presence of Schwann cells, perineural-like cells, and fibroblasts.[23] This tumor can occur as a solitary lesion or associated with neurofibromatosis type I, and can be found in hard or soft tissues. The tongue and the buccal mucosa are the most frequent involved sites in the oral cavity, but other sites such as the palate, lip, and gingiva have also been reported. Clinically, it represents a well-demarcated, sessile, slow-growing mass, which blends with the adjacent normal mucosa and is usually painless.[24]

Microscopically, CFM and OFM show the same pattern. They present as a non-encapsulated but well-circumscribed pool of myxoid connective tissue with scattered spindle cells, which do not stain for anti-S100, CD34, smooth muscle actin, desmin or CD68;[5-7] however, they stain positively for Alcian blue (pH = 2.5).[5-7] Based on the hematoxylin-eosin (HE)-stained sections, the histological differential diagnosis for OFM were mainly myxoid lesions such as myxoid neurofibroma, neurothekeoma, and angiomyxoma.[9,25] When an incisional biopsy is performed other tumors, which can eventually demonstrate a myxoid stroma cannot be ruled out. In contrast to most neural tumors, OFM does not express the S100 protein.

To summarize, this case of OFM presented an unusual clinical aspect, being located on the gingival and hard palate with a lobulated appearance, thus, demonstrating that OFM should be included in the clinical differential diagnosis of lesions of the gingiva/palate.

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The patient provided an informed consent declaration to permit the use of images and medical information.

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