



Oral Myofibroma: Clinical Case Report

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Abstract

Myofibroma is a rare mesenchymal benign spindle cell tumor consisting of myofibroblasts. In most cases it is solitary, and it can occur in any age group but it is more common until the fourth decade of life. It is a tumor most commonly originated from soft tissues, bones or internal organs, with predilection for the head and neck region. Clinically, the tumor appears as a painless mass with rapid growth, which in some cases can simulate malignancy. However, it is benign and the treatment of choice consists of surgical excision, with low chances of recurrence. Spontaneous regression can occur. The clinical case report in question was developed in a 79-year-old Caucasian female patient who underwent an incisional biopsy of a painless nodule in the mandibular vestibular gingiva. Histopathologically, it appeared as a tumor composed of interlacing bundles of spindle cells with tapered nuclei and eosinophilic cytoplasm. The tumor cells were positive for smooth muscle actin and vimentin, but negative for desmin in immunohistochemistry. The histopathological exam, with the aid of the immunohistochemistry technique, was conclusive for oral myofibroma. Mesenchymal lesions are infrequent in the mouth, especially at the age of the patient, which makes the case a histopathological rarity. The purpose of this case report is to elucidate and better understand the clinical characteristics, treatment and prognosis of this tumor. This report also discusses the diagnostic method and the expected characteristics in histopathological and immunohistochemical exams.

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Introduction

Myofibroma is a rare benign spindle cell neoplasm consisting of myofibroblasts. In most cases it is solitary, but it can also be multicentric and occur in any age group [1], being more common until the fourth decade of life, occurring slightly more often in males [2]. The exact etiology is unknown [1], but it is suggested that it may be sporadic or have a genetic origin and run in families with autosomal dominant inheritance pattern or recessive traits [3,4].

It is a tumor most commonly originated from soft tissues, bones, internal organs and even intracranial areas [3] with a predilection for the head and neck region [5], followed by the trunk and extremities [3]. Cases involving the oral cavity are rare. In the intraoral region, the mandible is the most common location [1], followed by the tongue and by the buccal mucosa, rarely seen in the gingival [6]. In younger patients, it is more likely to demonstrate bone involvement [2].

Normally this neoplasm presents a limited growth potential and may have a spontaneous resolution. In radiographic features they usually appear as a unilocular radiolucent area with undefined edges [1]. In histopathological exams, it appears as a spindle cell tumor consisting of myofibroblasts, arranged in intertwined bundles with elongated nucleus and eosinophilic cytoplasm [2].

Clinically, the tumor presents itself as a painless mass with slow [3] or rapid growth that can mimic malignancy [7]; however, it is completely benign. The treatment of choice is surgical excision [7], with low chances of recurrence. Spontaneous regression can occur in some cases, exhibiting their benign nature [3] and may be mediated by apoptosis [8].

Myofibromas are commonly not considered as a main diagnostic hypothesis in most cases and they are usually considered as a differential diagnosis during investigation [7], due to their rarity. The

purpose of this article is to report a case of oral myofibroma involving the mandible gingiva and to describe its clinical, histopathological and immunohistochemical features.

Case Presentation

A 79-year-old Caucasian female patient attended the Stomatology Clinic of the Army Dental Service for evaluation of a painless oral nodule noticed one month before the consultation. During the anamnesis, no abnormalities were noticed. Intra-oral examination revealed a submucosal nodule with a normochromic fibrous consistency in the mandibular vestibular gingiva, located between teeth 31 and 32 (Figure 1).

The diagnostic hypotheses initially were traumatic neuroma and solitary fibrous tumor. An incisional biopsy of the lesion was performed and sent to the Pathology Service (Figure 2). After a week, before the results were ready, the patient returned to the clinic complaining of difficulty eating. Upon clinical examination, a rapid growth of the lesion was observed, requiring a new incisional biopsy, which was performed to relieve the patient symptoms.

Histopathological analysis of 5-µm sections stained with Hematoxylin and Eosin H&E showed a spindle cell tumor consisting of myofibroblasts, arranged in intertwined bundles with elongated nucleus and eosinophilic cytoplasm (Figure 3).

Immunohistochemistry analysis showed positivity for anti-smooth muscle actin (Figure 4A) and anti-vimentin (Figure 4B) and negativity for antibodies anti-pan cytokeratin AE1/AE3, anti-CD34 and anti-desmin (Figure 4C).

The histopathological report with the aid of the immunohistochemistry technique was conclusive of myofibroma. The treatment of choice was surgical, which the patient underwent a new surgery for total excision of the lesion. The patient is being followed up with no signs of recurrence (Figure 5).



Figure 1: *Clinical aspect* - Nodule in the mandibular vestibular gingiva before excisional biopsy.

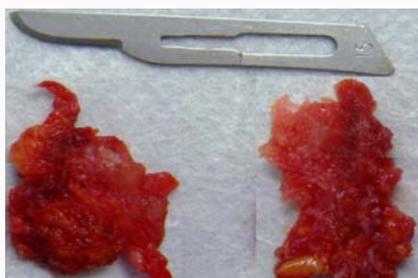


Figure 2: *Macroscopic aspects* - Two fragments of soft tissue, with irregular shape and surface, fibrous consistency and measuring together 3 cm x 2 cm x 1 cm.

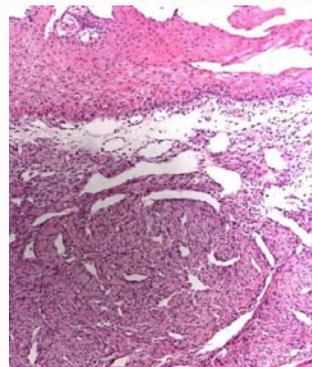


Figure 3: *Histopathological features (HE 10x)* - Proliferation of spindle-shaped cells with both fibroblastic and smooth muscle features.

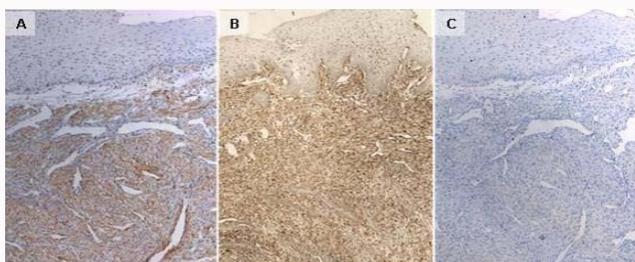


Figure 4: *Immunohistochemical features (IHC 10x)* - **A** Smooth muscle actin positive in tumor cells; **B** Vimentin intensely positive; and **C** Desmin expression negative in tumor cells.



Figure 5: *Follow-up* - Clinical view after two months.

Discussion

The reported case represents a rare benign neoplasm, oral myofibroma. Mesenchymal lesions are infrequent in the mouth, especially in the age group of the patient, being more common in younger age groups [9]. Although the location of the myofibroma in the mandibular vestibule gingiva has already been described in literature [6,7], it is a less common area of appearance for this type of tumor.

The patient presented a solitary lesion, which is the most common presentation form of myofibromas [9]. The neoplasm also showed a typical behavior of its type, which is rapid growth, since shortly after the partial removal of the lesion for the incisional biopsy, the patient returned to the medical unit having difficulty eating, due to the increased lesion. There are cases in literature describing myofibromas that started to accelerate growth after incisional biopsy [2], which often mimics a malignant tumor and, in the past, it has been commonly misdiagnosed by one.

There were no complaints of pain in the area affected by the

myofibroma, which is to be expected, because they are normally painless [10,11]. This, in many cases, prevents patients from perceiving it until the tumor enlarges sufficiently to become visible or appears on radiographic examination [12]. The patient did not report any similar cases in her family medical history, suggesting that the tumor has a sporadic etiology.

Histopathological exams associated with immunohistochemistry are necessary for the diagnosis of this type of tumor [12], mainly due to its rarity and the clinical and histopathological similarities to benign mesenchymal lesions, being normally an uncommon differential diagnosis. The main histological similarities of this type of lesion are with sarcoma, leiomyoma, fibromatosis, and myofibromatosis [2]. Myofibromatoses have the same histologic features as myofibromas, although the former consists of multiple tumors appearing several years apart in different locations, and their incidence is higher in children and very rare in adult or elderly patients [3].

Immunohistochemistry should be requested to support histopathology and establish a definitive diagnosis [5]. Immunohistochemistry is expected to be positive for smooth muscle actin and negative for desmin, as occurred in the case reported [9,12]. The immunohistochemistry along with the histological diagnosis mentioned fulfill the criteria for the diagnosis of oral myofibroma.

For the histological diagnosis, the biopsy must be incisional and for the treatment of the typical solitary lesion, a simple excision is recommended [12]. Local recurrence has been reported in some documented cases [2,13] though, most are associated with the anatomic restraints during surgery. These high recurrences are most likely to be found in cases in which surgical access is difficult [14].

Assuming that myofibroblasts that are influenced by various temporal and spatial factors become more resistant to apoptosis as the patient ages, lesions of myofibroma of the oral soft tissues that generally develop in children and older patients do not tend to regress spontaneously, occurring more commonly in infants [6]. Chemotherapy and radiation are rarely used, with the exception of some cases with recurrence or a non-resectable lesion [15]. The prognosis of this tumor is usually excellent in solitary cases [1] after resection. However, in cases with multicentric visceral lesions, they are more aggressive and sometimes may be fatal [2,12].

Conclusion

Despite having a good prognosis in most cases, it is fundamental that oral myofibroma tumors should be correctly and early diagnosed. Awareness and recognition of this benign neoplasm in the medical and dental communities, along with its inclusion in the differential diagnoses of oral cavity lesions are important to establish the correct diagnosis and avoid the morbidity of an unnecessary aggressive therapy.

Due to the similarity of the clinical and histopathological aspects,

with risk of possible overlap with benign mesenchymal lesions and malignant spindle cell tumors, immunohistochemistry is essential for establishing the definitive diagnosis and should be requested along with the histopathological exam of the incisional biopsy.

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