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Gingival myofibroma. A case report

ABSTRACT

Background *The Myofibroma (MF) is a benign mesenchymal tumor frequently observed in the skin and subcutaneous tissue of the head-neck region. It is described mainly in infants with congenital forms and, in early childhood, with acquired forms. Less often, it can be observed in adolescents and adults. The location of a MF in the oral cavity is occasional and differential diagnosis must be established with other benign and malignant neoplasms, high or low grade, of the oral mucosa. The histology of the myofibroma shows a biphasic growth pattern: elongated spindle cells with eosinophilic cytoplasm, in the borders, polygonal cells arranged in a palisading pattern, with hyperchromatic nuclei, in the central portions. The diagnosis of MF, usually made after excision of the neoformation, is obtained by means of immunohistochemistry, in which there is positivity for vimentin and αactine smooth muscle antibodies and negativity for keratin, S-100, EMA (Epithelial Membrane Antigen) antibodies. The treatment is surgical; the prognosis is generally good with low rates of recurrence after excision.*

Case report *The authors describe a case of MF in a 16-year-old male subject, that came to their observation for the growth of a considerable mass attached to the gingival mucosa, describing the therapeutic strategy.*

Keywords *Gingival disease; Gingival neoplasm; Myofibroma*

Introduction

Myofibroma (MF) is a benign mesenchymal neoplasm, observed in the skin and subcutaneous tissues of the head and neck region. Congenital forms are described, especially in newborns and infants, but it is also observed in adolescents. The immunohistochemical pattern is characterised by high positivity for antibodies anti-vimentin, anti αactin smooth muscle antibodies, negativity for antibodies anti-keratin, anti-S-100 and anti-EMA.

Three forms are known: solitary (the most common one, more than 50% of all cases), multicentric (less than 30% of all cases) and multicentric with visceral involvement (less than 15% of cases). The solitary form involves skin, subcutaneous tissues, fascial planes, muscle and bone structures. In the multicentric type are found multiple, synchronous or metachronous, myofibromatosis areas, with sometimes (multicentric variant with visceral involvement) pulmonary, cardiac, gastrointestinal, and CNS localisations [Behar et al., 1998]. The prognosis in solitary and multicentric forms without visceral involvement is excellent, whereas the visceral involvement is potentially fatal.

Males are more frequently affected. Clinically, MF is generally an esophytic sessile mass, with homogeneous surface, but sometimes superficial erosions caused by occlusal trauma can be observed. Microscopically the MF consists of myoid cells, distributed around the walls of small vessels, forming a morphological continuum with myopericytes. The microscopic aspect is characterised by single or multiple nodular areas, rich of spindle shaped myofibroblasts, which are well differentiated and arranged in lines, with lengthened slightly eosinophilic cytoplasm. In the central portion, the degree of differentiation becomes less marked, and the cells can be arranged in a palisading pattern, with hyperchromatic nuclei around small vessels (haemangiopericytoma-like pattern). The low mitotic activity contributes to the low rate of recurrence and metastatic potential of this disease.

The immunohistochemistry highlights positivity for vimentin and αactin and negativity for S-100, EMA and keratin. Although, as mentioned above, the head and neck region is frequently affected by forms of MF, the oral localisation is infrequent and few cases are described in the literature.

The rarity of the gingival form of myofibroma compelled us to describe the case, along with the clinical, histological and therapeutic aspects.

Case report

The patient, a 16 years old male, was referred to the Maxillofacial Surgery unit of the San Salvatore Regional Hospital (L'Aquila, Italy), about 3 years ago, for a sessile mass in the right inferior first molar region.

From the medical history it emerged that tooth 46 had been uprooted, 3 months before the observation, during a traumatic event. The mass was roughly oval-

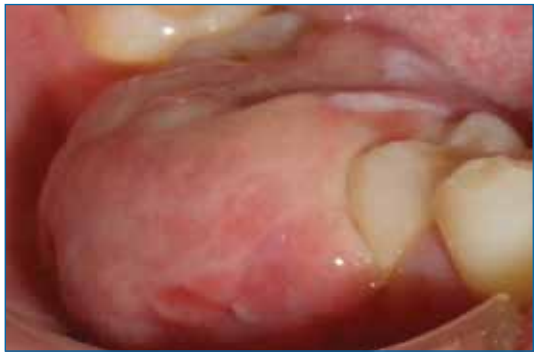


FIG. 1 Intraoral view of the gingival neoplasm.

shaped, red-greyish, with a regular surface, fibrous consistence, and it was quite painful to palpation (Fig. 1). The lesion increased rapidly over the two weeks after the first observation, with very painful symptoms and bleeding episodes. The patient reported that in a previous dental visit, both anti-inflammatory and antibiotic therapy was prescribed, without any benefit. There was no evidence of fever or regional lymph nodes swelling.

The blood tests, routinely performed at admission, showed no significant alterations. Afterwards an ortopantomography was taken, which highlighted vertical bone loss, in the traumatic uprooted tooth region that extended mesially to tooth 47 and distally to tooth 45; it was also seen the presence of residual roots of teeth 36 and 15.

The treatment consisted in the complete surgical excision of the mass up to the bone plane, which resulted unaffected, under local anaesthesia. After control of bleeding was obtained, a buccal suture was made, and the patient was dismissed with antibiotic and anti inflammatory therapy (Amoxicillin + Clavulanic Ac cpr 1 g 2/die and Nimesulide 100 mg 2/die).

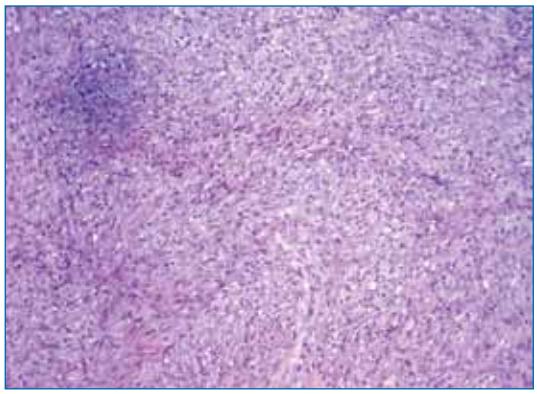
The excised material was sent to the Department of Anatomical Pathology for the histological examination. The suture was removed after 10 days. The first follow up, at 30 days, showed that the surgical site was in good condition: no signs of phlogosis or swelling were observed and no symptoms were reported by the patient; the follow-up at 12 and 24 months confirmed the complete healing and the total anatomic, morphological and functional restoration of the site, with no signs or symptoms of recurrence.

Discussion

The localisation of MF in the maxillofacial district is not uncommon, while the oral localisation is unusual [De Souza et al., 1999; Lingen et al., 1995]. The MF is a benign neoplasm, in which a biphasic proliferation is observable. Cells similar to mature smooth muscle cells, myofibroblasts, are spread in the outer tissue (Fig. 2, 3). In the central portions polygonal cells can be observed that tend to assume a palisading pattern around small vessels in intimate relationship with myopericytes (haemangiopericytoma-like pattern) [Scheper et al., 2005].

The malignant transformation and metastatic ability is low. The long-term prognosis is good, but aggressive forms (dermoid myofibroma), for which treatment is chemotherapic and surgical combined, are recognised [Seper et al., 2005; Shah et al., 1998; Conley et al., 1966].

The aetiopathogenesis of MF is unknown. For some authors the pathogenesis resides in a minor trauma, in the same site, occurred at least two months prior (as in the present case). Due to the histological similarities between myofibrolasts and scar tissue it can be assumed that MF might be due to an exuberant reactive response to a traumatic event. On the other hand, it is possible that a traumatic event could stimulate a tissue in which myofibromatosis loci are already present. Nevertheless the hypothesis of exuberant tissutal response, subsequent to a trauma, does not explain the neonatal forms of myofibromatosis [Kassenoff et al., 2004]. The lack of specificity in the clinical presentation, the



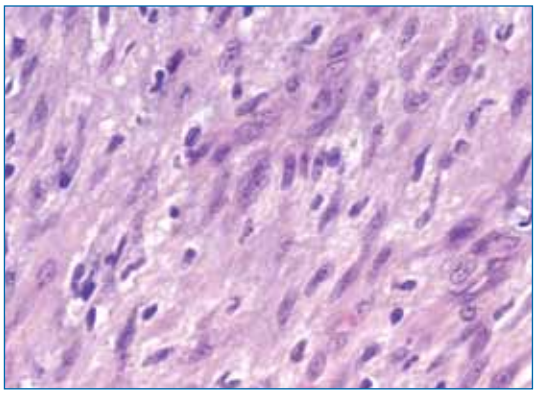


FIG. 2 Histology of myofibroma at low magnification (EE 10x). FIG. 3 Spindle cells with large nuclei and eosinophilic vesicular cytoplasm (EE 40x).

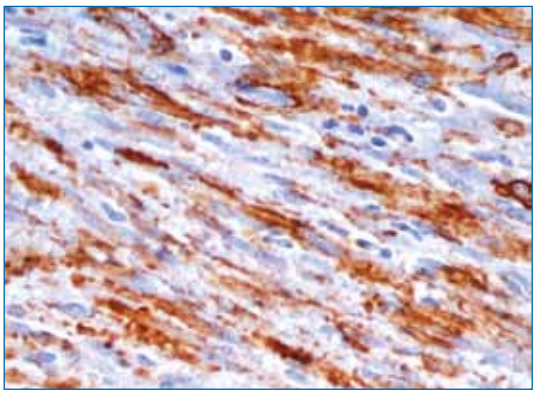


FIG. 4 Immunohistochemical view of MF, positivity for Vimentine (magnification 40x).

macroscopic aspect and the unusual site of presentation of the MF, make differential diagnosis with other benign tumors difficult; therefore a histopathological evaluation is needed. Immunohistochemical studies show a characteristic pattern: positive for anti-vimentin (Fig. 4), anti αactin smooth muscle antibodies, negative for anti-S-100, anti-EMA and anti-keratin antibodies. Due to these particular immunohistochemical characteristics, the differential diagnosis must be established with fibrosarcoma (which is characterised by high cellular malignancy) and haemangiopericitoma.

Conclusion

In conclusion, it is possible to state that the MF is a benign neoplasm whose treatment of choice is the surgical excision [Beck et al., 1999] with low recurrence rate [Liu et al., 2001].
An important clinical aspect is represented by the rapid growth of this lesion that in a young subject may suggest the diagnosis of more aggressive and proliferative neoplasms.
The clinical approach requires a careful clinical evaluation of symptoms and signs, followed by an accurate diagnosis by means of histopathological and immunohistochemical examinations.

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