Myofibroma of the Gingiva: Report of a Case

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Myofibroma is a benign mesenchymal neoplasm composed of myofibroblasts which has been described with different synonyms since the first report in 1951. It occurs most commonly as a solitary lesion of soft tissue, skin, or bone in infancy. The prognosis of oral myofibromas is excellent, and surgical excision is curative. Recurrence is rare. Awareness and recognition of this benign tumor is important to establish the correct diagnosis and avoid morbidity of unnecessary aggressive therapy. This report describes a myofibroma of the gingiva in a 14 year old girl and is reported together with the conventional histologic, and immunohistochemical findings. The tumor showed rapid increase in size and clinical features suggestive of malignancy. However, on histopathologic evaluation it was diagnosed as a benign neoplasm, and this diagnosis was supported by immunohistochemical markers. The spindle cells were immunopositive for smooth muscle actin, and vimentin but were negative for desmin and S-100 protein. The patient was treated with surgical excision, and is followed-up for 33 months without any signs of recurrence.

Keywords: *Oral myofibroma, infant, spindle cell neoplasm.*
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INTRODUCTION

Myofibroma and myofibromatosis are terms used to describe conditions characterized respectively by solitary and multicentric presentation of a benign spindle cell neoplasm composed mainly of myofibroblasts situated around thin-walled blood vessels.[1] The first description of this entity was reported in 1951 as congenital fibrosarcoma.[2] In 1954, the term congenital generalized fibromatosis was introduced, because of the lack of malignant potential[3] which later gave way to infantile myofibromatosis.[4] The name infantile myofibromatosis suggests that the lesion is considered to arise exclusively in neonates and infants, either solitary or multicentric. However, it is now apparent that adults may also be affected.[5-7] In addition, solitary myofibromas are more common than multicentric ones.[4,7]

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Etiology of myofibroma is unknown. As myofibroblasts are thought to play a role in wound healing, trauma or injury may contribute to the development of the tumour and it is believed that myofibroma is derived from myofibroblast cells.[8]

Solitary myofibromas of the head and neck region occur more commonly in children and isolated myofibromas of both children and adults have since been reported in a variety of sites, including the oral cavity.[9-11] They develop most frequently in the first decades of life, with a mean age of 22 years.[12] It has been reported in the mandible, tongue, buccal mucosa with only a few cases reported from the gingiva.[12,13]

Clinically, the appearance of the oral myofibroma is quite variable, and the tumor can enlarge rapidly and ulcerate, alarming to the clinician of malignancy. However it may regress spontaneously.[1,14] The tumour may be confused with benign lesions and low-grade malignant lesions.[6,9,14] It is however completely benign and upon complete surgical excision has an excellent prognosis. Although, the lesion behaves in a benign fashion, low recurrences can be also seen.[7]

We found only 46 cases of oral soft tissue myofibromas with well documented clinical and histopathologic data reported in the last 42 years of the English-language literature.[15-17]

The differential diagnosis includes a range of spindle cell neoplasms that have many overlapping and subtle histologic patterns which often render final diagnosis difficult and challenging.[6,11,18] Immunohistochemical investigations are usually used to support the histological diagnosis.

The aim of the present study is to report a new case of myofibroma and to discuss the difficulties in diagnosis and differential diagnosis of this benign tumor.

Case Report

A 14 year old female was referred to the the Department of Oral and Maxillofacial Surgery for evaluation of a painless mass on the mandibular gingiva. The patient and the family mentioned that the lesion grew rapidly in a 6 weeks period.

Clinical examination revealed a firm, mucosal colored mass at the back of the second right mandibular molar extending towards the vestibulum and lingual mucosa and measuring 3.2 x 2.0 x 0.8 cm. The lesion was painless and non-hemorrhagic in response to palpation but the upper surface of the lesion was ulcerated because of the crunching of the upper teeth (Fig 1). No evidence of purulence was present. The patient complained of difficulty in chewing related to huge volume of the lesion because of the pressure at the mass. The mandibular right second molar tooth showed an abnormal mobility and the mandibular first molar had an amalgam filling. The regional lymph nodes were not palpable. The patient's medical history was noncontributory, and she was not under any medication. The routine laboratory findings were within normal limits.

In the radiologic examination, orthopantomography revealed no alterations of bone in the region of the lesion, but an impacted third molar (Fig 2). The initial clinical differential diagnosis included sarcoma, lymphoma, leiomyoma and reactive processes, particularly peripheral giant cell lesion or pyogenic granuloma. The patient was scheduled for biopsy. An incisional biopsy was performed under local anesthesia.

The specimen was fixed in 10% neutral buffered formalin and routinely processed for paraffin embedding for histologic examination and immunohistochemistry. Histologic analysis of 5-μm sections stained with hematoxylin and eosin (HE). Microscopic examination revealed a partially ulcerated and encapsulated nodular tumor composed of spindle cells. There was no cellular atypia, but some normal mitotic figures were identified (Fig 3). Immunohistochemical reactions were positive to smooth muscle actin and vimentin (Figs 4 and 5). Other immunomarkers, including S100 protein, and desmin were all negative, thus supporting the myofibroblastic nature of the cells and consequently the diagnosis of myofibroma.

Complete surgical excision of the tumour with adjacent teeth was performed under general anesthesia. The postoperative course was uneventful, and the patient has been in follow-up for 33 months without any signs of recurrence (Fig 6).

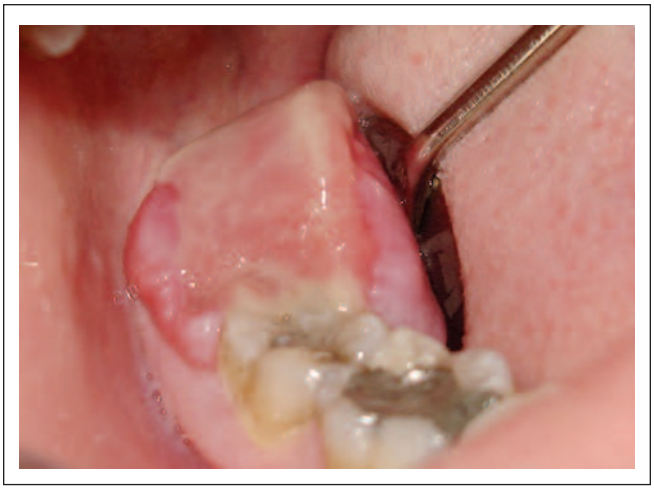

Figure 1. Clinical view of the patient.

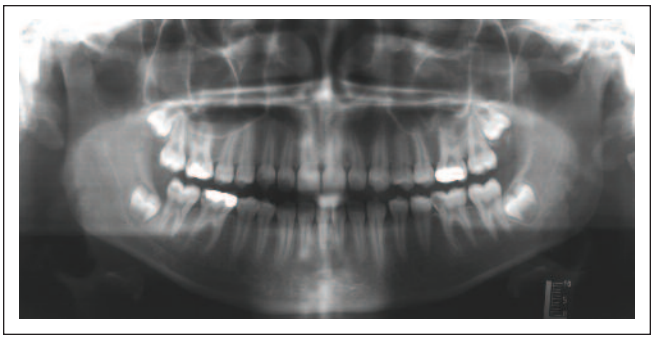

Figure 2. Orthopantomograph of the patient prior to the operation.

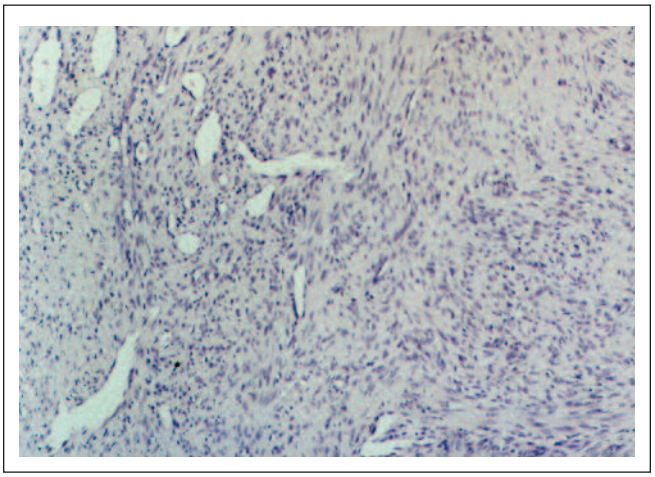

Figure 3. Myofibroma composed of fascicles of spindle cells (hematoxylin and eosin stain, original magnification x110)

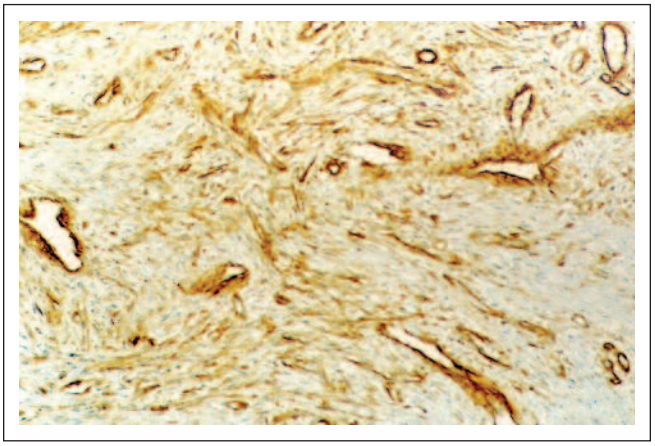

Figure 4. Positive immunohistochemical stain for smooth muscle actin (original magnification x110).

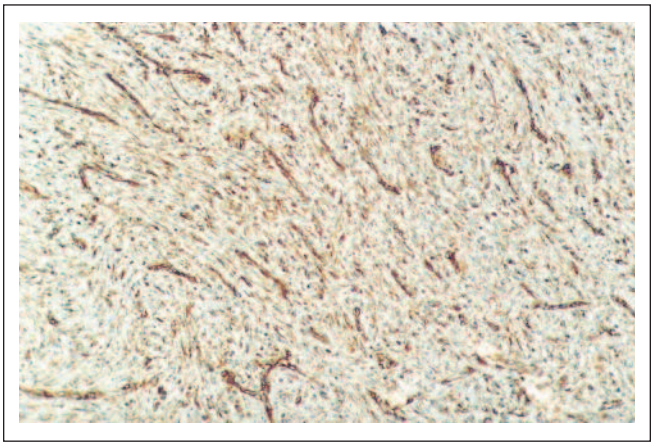

Figure 5. Positive immunohistochemical stain for vimentin (original magnification x40).

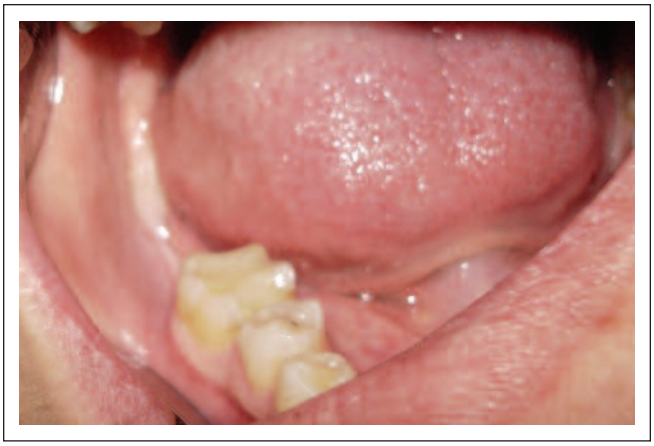

Figure 6. Clinical view after 33 months.

DISCUSSION

Solitary myofibromas are uncommon benign lesions and these tumours show a predilection for the head and neck region and particularly for the oral cavity.[4]

Oral myofibromas have been reported occurring in the mandible, tongue, buccal mucosa, hard palate, floor of mouth and gingiva.[5-7,10,11,15,17] To date, there are 9 reported cases of gingival myofibroma in the English language literature with only 6 cases reported in children.[5,6,10,13,17] Table 1 shows the details of 10 cases of gingival myofibroma with 7 cases, including the current case, occurring in children.

Oral myofibromas are usually unencapsulated but circumscribed as also observed in our case. Histologically, they have a monomorphic spindle cell appearance but a characteristic feature is their biphasic, zonal architecture. Some areas are composed of spindle cells with tapered or oval nuclei and vesicular nuclear chromatin, arranged in a rather haphazard, fascicular pattern.

Table 1. Reported cases of gingival myofibroma in adults and children.

Authors	Age	Gender	Size (cm)	Duration	Initial diagnosis
Beham et al[5]	60	M	0.5	NI	NI
Montgomery et al[6]	50	M	2.2	NI	NI
Jones et al[10]	70	F	0.8x0.5	4 months	Atypical cellular smooth muscle tumour
Jones et al[10]	8	F	1.5x0.7	2 weeks	Myofibromatosis
Jalil et al[13]	8	F	3.0x2.5	3 weeks	Peripheral giant cell granuloma
Jalil et al[13]	14	F	3.5x2.0	NA	Pyogenic granuloma ? Malignant tumour
Jalil et al[13]	7	M	3.0x2.0	2 months	NA
Jalil et al[13]	7	F	2.5x2.0	2 months	Pyogenic granuloma
de Souza et al[17]	9	F	2.0	3 months	Peripheral ossifying fibroma Peripheral giant cell granuloma Neurofibroma
Present Case	14	F	3.2x2.0	6 weeks	Peripheral giant cell granuloma Malignant tumour

F: female, M: male NI: not indicated, NA: not available

The differential diagnosis of myofibroma is wide and includes other benign lesions and malignant neoplasms, including benign and malignant mesenchymal lesions which may be of fibrous, muscle, neural, or vascular tissue origin.[6,7,15,18] In our case, differential diagnosis included conditions commonly observed in young patients as rhabdomyosarcoma, fibrosarcoma, leiomyosarcoma, and leiomyoma.

Myofibromas commonly infiltrate and entrap the adjacent normal tissue and have a relatively high mitotic activity; therefore, they may also be easily misinterpreted as malignant or aggressive lesions and treated inappropriately. As most of these lesions are found in infants and children, the differential diagnosis is usually with the infantile form of fibrosarcoma, which carries a better prognosis than the adult counterpart. Fibrosarcoma was included in the differential diagnosis of the present case owing to the rapid enlargement and ulcerated surface. Fibrosarcoma of the soft tissues occur mostly in persons between 30 and 50 years of age.[19] Histologically, it has an undifferentiated appearance. Cells in the tumour have irregular nuclei. Despite little cellular pleomorphism, fibrosarcomas usually presents prominent mitotic activity and nuclear hyperchromasia, features not commonly observed in myofibromas. Myofibromas may have a relatively high mitotic rate without atypical mitotic figures.[17]

Submucosal examples of myofibromas with superficial ulceration, as also seen in our case, may also cause diagnostic problems. The acquired inflammatory component, consisting of plasma cells and other mixed elements, broadens the differential diagnosis considerably, from inflammatory myofibroblastic tumor on the benign end of the spectrum to low-grade fibrosarcoma on the malignant end. Superficial biopsy specimens of ulcerated mesenchymal lesions should be interpreted with caution. Accurate diagnosis needs an evaluation of the overall pattern, which is best assessed in the deep, non-ulcerated component. General surgical pathologists may be unaware of this lesion in the differential diagnosis of tumors of the mouth and jaw, in which it is probably somewhat more common than the medical literature would indicate.[6]

Low-grade myofibroblastic sarcoma is a recently described distinct entity representing a rare mesenchymal malignant neoplasm with myofibroblastic differentiation that occurs predominantly in adults with a predilection for the soft tissue of head and neck.[16,20,21] Its microscopic appearance includes a cellular infiltrative neoplasm composed of spindle cells arranged in fibrosarcoma- like fascicles, sheets, or storiform whorls that show at least focally moderate atypia and slightly increased proliferation rate,[16, 20,21] overall features not observed in myofibromas, that usually present a monotonous appearance of a biphasic growth pattern.

Wavy and blunt-ended nuclei seen in neurofibroma and leiomyoma/leiomyosarcoma, respectively, were not observed in the present case.[9,18,21,22]

Differentiation from solitary fibrous tumor may also be difficult because of the hemangiopericytoid appearance shared by both lesions; nevertheless, the biphasic pattern present in the present case is not observed in solitary fibrous tumor, described as a patternless proliferation of spindle cells, alternating hypercellular areas and hypocellular areas rich in dense keloid-type collagen.[18,23,24]

Benign fibrous histiocytoma may also present hemangiopericytoma-like areas,[1] but it is better characterized by a biphasic cell population of fibroblast-like spindle cells and histiocyte-like rounded cells arranged in a storiform pattern.[11,22-24]

Even though there are subtle morphologic differences between all of these benign and malignant mesenchymal lesions immunohistochemistry is a valuable, precise, and reliable method for reaching accurate diagnosis of oral myofibroma and, in particular, for the differential diagnosis of spindle-cell neoplasms arising in the oral cavity. Myofibromas are characterized by staining with vimentin. Immunoistochemical negativity for S100, and desmin excludes Immunohistochemical tumors of neural, and smooth-muscle origin, respectively.

The solitary myofibroma is typically a painless mass. Although the tumour pursue a benign course and may even regress spontaneously,[1] sometimes it exhibits rapid enlargement as also seen in our case. In this case, the lesion grew rapidly in a 6 weeks period. 2 weeks after the first incisional biopsy, the patient was scheduled for the surgical excision of the tumour. It was observed that the size of the tumour showed further enlargement within 2 weeks.

Solitary myofibromas are usually treated by surgical excision. As these benign tumours commonly infiltrate and entrap the adjacent normal tissue, wide surgical excision is essential. In our case, complete surgical excision of the lesion with adjacent teeth was preferred to prevent the recurrence. A small percantage of tumours will recur after treatment, but typically, these can be controlled with reexcision.

CONCLUSION

Myofibroma is a benign tumor of scant incidence in the oral cavity, and with a good prognosis, though it must be included in the differential diagnosis of other oral mucosal lesions. Awareness of this benign tumour is important and may avoid misdiagnoses, unnecessary aggressive therapy. The treatment of choice is wide surgical excision with adequate safety margins in all cases and careful post-operative observation shall be continued. In this case of myofibroma, no post-operative complications and no recurrence were observed in 33 months period.

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