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Solitary Adult Gingival Myofibroma : Case Report and Literature Review

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Abstract:- Myofibroma is a benign mesenchymal neoplasm frequently observed in the head-neck region. It is mostly reported as congenital forms in newborns and acquired in early childhood. Adolescents and adults are less likely to experience it. Myofibromas in the oral cavity is uncommon, and differential diagnosis must be made with various benign and malignant neoplasms of the oral mucosa either high or low grade.

Histologically, myofibroma exhibits a biphasic development pattern, with elongated spindle cells with eosinophilic cytoplasm on the edges and polygonal cells grouped in a palisading pattern with hyperchromatic nuclei in the middle.

The diagnosis of myofibroma is generally established through immunohistochemistry, which shows positivity for actin smooth muscle antibodies (α-SMA) and negative for keratin, S-100, and epithelial membrane antigen (EMA) antibodies.

The treatment is surgical, and the prognosis is generally favorable, with minimal recurrence following excision.

Herein, we present a case of myofibroma in the gingiva of the right lingual molar region of a 34-year-old male patient, describing the diagnosis and the therapeutic strategy through a literature review.

Keywords:- *Myofibroma, Soft Tissue Neoplasms, Myofibromatosis, Gingival Neoplasms.*

I. INTRODUCTION

Myofibroma is a benign mesenchymal neoplasm characterized by the proliferation of fibroblasts and myofibroblasts frequently observed in the head-neck region. It is categorized as solitary or multicentric.[1]

It is mostly seen in newborns and young children (82% under 2 years old), with congenital forms. It can be observed less commonly in adolescents and adults.[2,3]

The development of myofibroma in the oral cavity is rare, and differential diagnosis must be established with various benign and malignant neoplasms of high and low grades of the oral cavity.[4] Herein, we describe a case of gingival myofibroma of the mandible, including clinical, imaging, microscopical, and immunohistochemical features, as well as a literature review.

II. CASE REPORT

A 34-year-old male was referred to the oral medicine and surgery department of Fattouma Bourguiba University Hospital of Monastir, with a chief complaint of a rapidly growing swelling in the mandibular right posterior region for approximately 4 weeks, causing chewing difficulties and discomfort. There was no history of previous trauma or infection in this region, and the past medical history was not contributory.

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The extraoral examination had no particularities. The intraoral examination revealed a nodule in the lingual gingiva of the right mandibular molar region covered by normal-colored mucosa, partially ulcerated, measuring approximately 2,5 cm in diameter, extending from the first to the second mandibular molar of the same region (46 and 47) (Figure 1). The lesion was partially bleeding, had a firm consistency on palpation, and was painless, except for slight discomfort due to tumor growth.

No signs of local trauma or chronic irritation were noted on the physical examination.

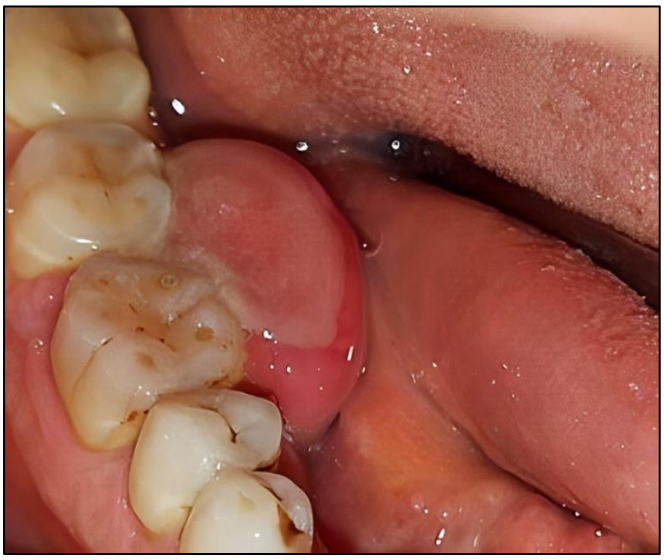

Fig 1 Pre-Operative Appearance of a 2,5 cm Nodule, with well-defined Borders, Sessile, Locally Ulcerated on the Surface, in the Right Mandibular Alveolar Ridge Region

Cone-beam computed tomography (CBCT) scanning demonstrated the presence of a superficial lytic defect without distinct cortical margins in the right posterior mandibular lingual cortical **(Figure 2)**. The initial clinical differential diagnosis included peripheral giant cell tumor, Peripheral odontogenic fibroma, schwannoma, neurofibroma, and solitary fibrous tumor.

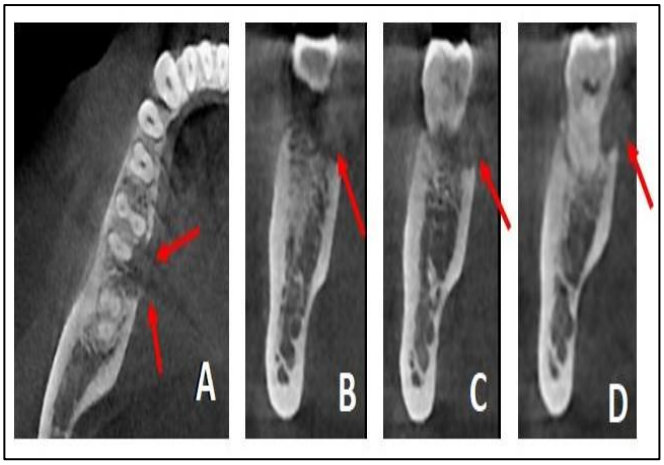

Fig 2 Imaging Studies. CBCT Scan of the Right Mandibular Region in Transverse View (A) and Coronal Views (B-C-D) Showing Alveolar Bone Loss between 46 and 47.

An entire lesion exeresis was performed under local anesthesia (**Figures 3A,3B, and 3C**). After proper site curettage and bleeding control, a simple suture was made, and the patient was dismissed with a prescription of antibiotics, analgesics, and an antiseptic mouth-wash. The biopsy specimen was submitted for histopathological analysis.

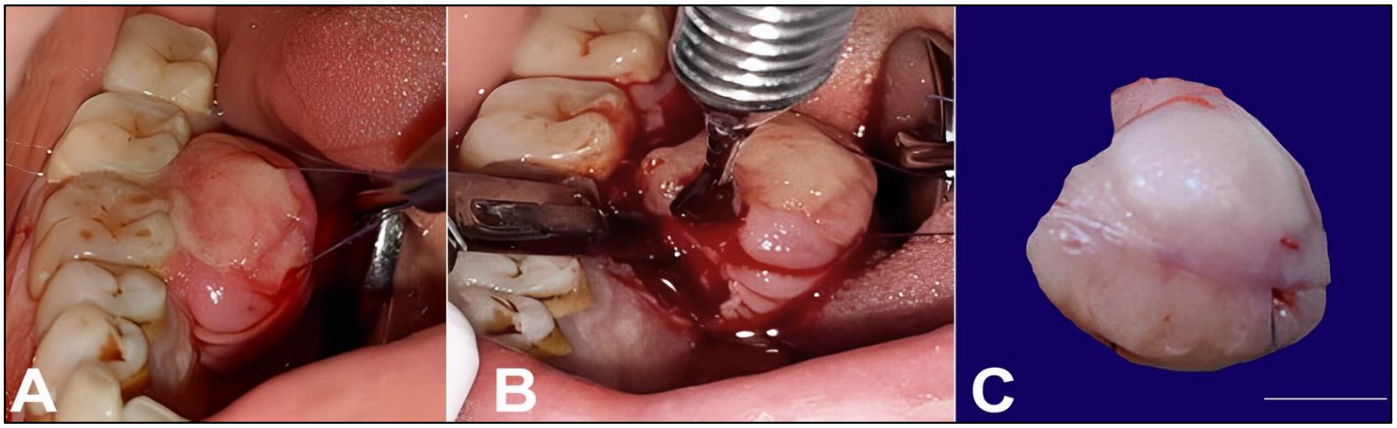

Fig 3 Intraoral View of the Surgical Procedure. A and B - Showing the Nodule Resection; C - The Specimen Measuring 2.4 × 1.7 × 1 cm (scale bar = 1 cm).

Gross examination revealed a specimen measuring 2.4 × 1.7 × 1 cm with a firm consistency and a grayish surface. Histopathological examination revealed a focally ulcerated squamous surface epithelium. The nodule was composed mainly of spindle-shaped cells grouped in long crossed bundles. The appearance of these cells is suggestive of myofibroblastic differentiation. Their cytoplasm is sparsely eosinophilic, and their nuclei are monomorphic with fine chromatin. Mitosis is a rare occurrence. At high magnification, there are 3 mitoses / 10 fields. Numerous branching vessels with a hemangiopericytoma appearance separate the bundles (**Figures 4A and 4B**).

The myofibroblastic nature of tumor cells was confirmed by an immunohistochemical panel, which showed a positive reaction for α-smooth muscle actin (α-SMA) (**Figure 4C**) and h-caldesmon (h-CD) (**Figure 4D**) and negative for Desmin, SOX 10, S100, and CD 34.

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Based on clinical, histopathological, and immunohistochemical findings, a diagnosis of myofibroma was established.

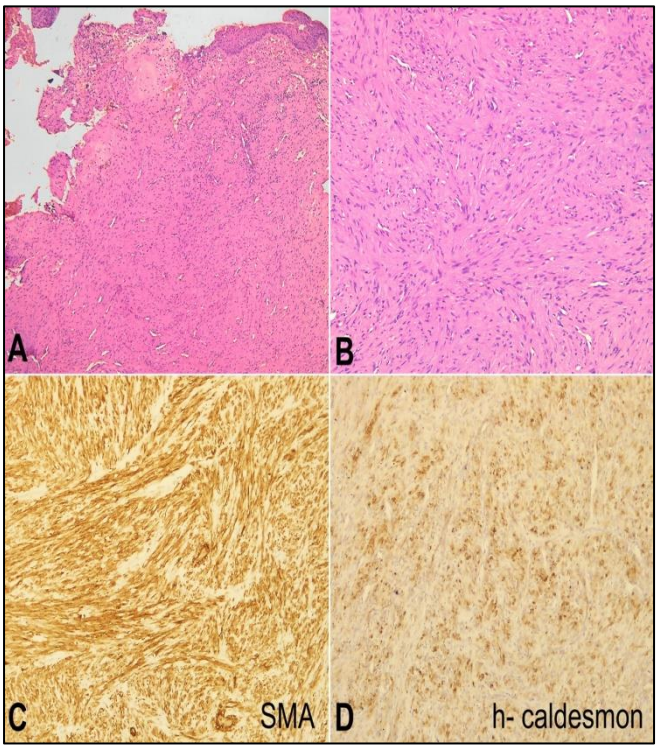

Fig 4 Photomicrographs of the Surgical Specimen. A-Myofibroma at low Magnification; B - Fusiform Cell Bundles with Abundant Extracellular Matrix of Collagen; C-Positivity of Alpha-Smooth Muscle Actin (α-SMA); D-Positivity of h-Caldesmon

The patient returned seven days after surgery. The stitches were removed, and the wound was healing. In his post-operative recovery, the patient reported no discomfort.

After a year of observation, the area examination revealed the persistence of the bone defect between the first and second molars without signs of recurrence (**Figure 5**).

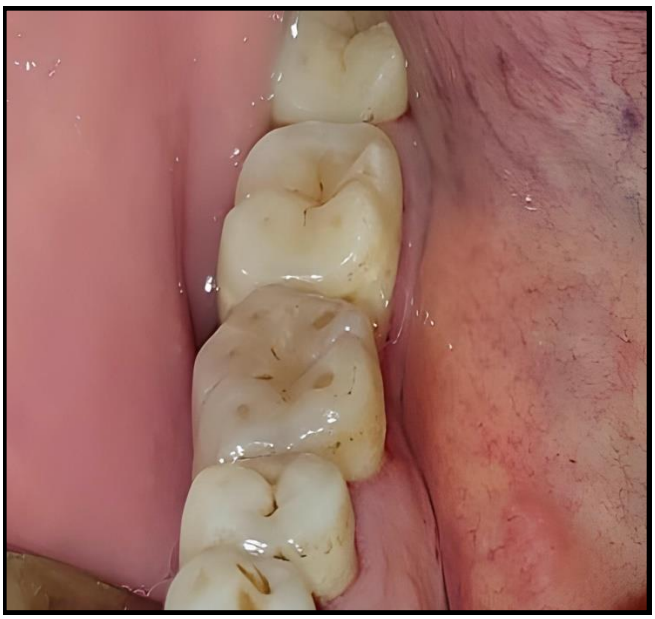

Fig 5 Postoperative Intraoral View of the Resection site after 1 Year

III. LITERATURE REVIEW

Literature was carried out through a search of PubMed (MEDLINE) and EBSCO Host electronic databases using the following Boolean formula ("myofibroma" AND ("gingiva" OR "gingival" OR "maxilla" OR "maxillary" OR "mandible" OR" mandibular")). No time restrictions were applied; only cases of gingival solitary myofibromas in the adult population were included.

To our knowledge, only eight cases of solitary gingival myofibromas affecting adults have been reported in the English-language literature, including the current case. Data were collected and prepared for analysis and tabulation (**Table 1**).[2,5-10]

Table 1 Summary of Literature Review on Adult Solitary Gingival Myofibromas

Ref	Age/G	Time	Symptom	Size cm	Bone loss	Histology	Ihc	Treatment	Outcome
2	60M	-	PS	0.5	-	Spindle cells with eosinophilic cytoplasm	(+) for α-SMA and HHF - 35 muscle-specific actin. (-) for desmin and S-100 protein	Excision	unknown
5	70F	4 m	-	0.8	-	Spindle cell neoplasm with an ill-defined biphasic pattern	-	Excision	-
6	50M	2 m	PS	2.2	-	Fusiform cells with a slight nuclear enlargement	(+) for α-SMA	Excision	Unknown
7	34M	2 m	PS	3	yes	spindle-shaped cells with an interlacing bundles and	(+) for α-SMA (-) for desmin, S-100 and CD34.	Excision with 5 mm security	-

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						eosinophilic cytoplasm		margins	
8	52F	1 m	FGM	2.0 then 3.5	No	Interlacing bundles of spindle-shaped cells	(+) for vimentin; HHF-35 muscle-specific actin; α-SMA; CD34	Excision with security margins	No recurrence 34 months after surgery
9	53F	4 w	PfBS	2	No	-	-	Excision	No recurrence 36 months after surgery
10	40F	5 m	PSwB	2	No	fusiform cells resembling fibroblasts	(+) for HHF-35 muscle-specific actin, vimentin and α-SMA; (-) for desmin, S-100 protein, CD34	Excision with security margins	No recurrence 1-year after surgery
CC	34M	4 w	PBS	2	yes	Fusiform cell bundles with extracellular matrix of collagen	(+) for α-SMA; H-caldesmon; (-) for Desmin, SOX 10, and CD 34	Excision	No recurrence 1-year after surgery

F: female; FGM= fast-growing mass; G: gender; G=Gingiva; IHC: immunohistochemistry; LG= lingual Gingiva**;** m= month; M: Male; PG= palatal Gingiva; PBS=painless bleeding swelling; PfBS= Painful bleeding swelling; PS Painless swelling; Ref= reference; α-SMA: *α*-smooth muscle actin; w= week, -: not available data; (+): positive reaction(-): negative reaction.

IV. DISCUSSION

In 1989, Daimaru et al.[11] and Smith et al.[12] described the first series of solitary myofibromas in adults, defining separate lesions constituted of myofibroblasts as indicated by immunohistochemistry and electron microscopy.[2] Similar lesions had been mentioned previously in 1981 by Chung et al.[13] while describing 'infantile myofibromatosis'. Infantile myofibromatosis or myofibromas appear as single or numerous nodules during the first decade of life, frequently before the age of two years. The lesions can be cutaneous or reach deeper tissues, including muscle, bone, and even visceral organs, posing a later risk of death.[14]

Adult myofibromas, on the other hand, are rare, solitary, superficial, and behave in a completely benign manner. Our 34-year-old male case, is in agreement with the literature. Fletcher et al.[1] concluded that males are twice as likely to be implicated.

Although the head and neck area is predominantly involved, cases affecting the oral cavity are infrequent and have a wide differential diagnosis. In recent research by Aiki et al.,[7] myofibromas were found to affect the mandible (33%), gingiva (23%), as in our case, tongue (15%), oral mucosa (12%), palate (8%), lip (4%), and other locations (5%), in that order.

Myofibroma has an unknown etiopathogenesis. A probable inheritance pattern has been suggested in some studies.[14-16] However, since myofibroblasts and scar tissue have histological similarities, myofibroma may result from a post-traumatic excessive reactionary response. Nevertheless, this theory does not quite explain the neonatal forms of myofibromatosis.[15]

According to our review, the gingival location of myofibroma is quite variable; generally, it is a painless exophytic sessile mass, ranging in size from 0.5 cm to 3 cm with a homogeneous surface, sometimes exhibiting rapid enlargement and superficial ulceration caused by occlusal trauma like in our case.[2,7,8] This clinical presentation lacks distinctive features unique to myofibromas. It can prompt consideration of alternative soft tissue neoplasms, with epulis being the most frequently encountered, along with conditions such as pyogenic granuloma, gingival hyperplasia, peripheral giant cell tumor, peripheral odontogenic fibroma, schwannoma, neurofibroma, and solitary fibrous tumor.

In a study published by Abramowicz et al.,[17] myofibroma arising within the jawbones in children can have two types of growth patterns: one type is an aggressive exophytic type associated with an ulcerated gingival mass of rapid growth, and the other type is a non-aggressive intra-osseous type that may be found incidentally.

Radiologically, intraosseous myofibromas often display a clear-cut radiolucent appearance, while gingival lesions can range from having no observable bone loss to exhibiting evident alveolar bone erosion and destruction, sometimes leading to floating teeth.[18-20] Surface erosion of bone was seen in our case.

Although adult patients are less likely to demonstrate bone involvement,[8] the current case is an example of extraosseous myofibroma with some underlying bone involvement primarily attributed to the localized expansion of the lesion.

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The histological features of oral soft tissue myofibroma, according to Vered et al.,[21] are similar to those documented for myofibroma in other body areas. Yet, although necrosis is a typical sign in tumors affecting other anatomical locations, necrosis has only been seen in four cases of oral soft tissue myofibroma.[21] It has been proposed that necrosis is linked to myofibroma's spontaneous regression.[22] Apoptosis, which is found in many normal embryonic and postnatal developmental processes, may be involved in the spontaneous regression of myofibromas. This could explain why congenital myofibromas or those that emerge shortly after birth, tend to regress spontaneously.[22] As the patient ages, it is reasonable to expect that myofibroblasts, impacted by numerous temporal and geographical variables, become more resistant to apoptosis. As a result, unlike in newborns, myofibromas of the oral soft tissues that form in children or adults do not tend to resolve spontaneously.

Myofibromas are generally well-defined but unencapsulated masses and exhibit a biphasic pattern of bright and dark-stained regions on histology. The bright areas are mainly made up of spindle cells with eosinophilic cytoplasm and conical or cigar-shaped nuclei, grouped in short fascicles or spirals and nodules at the lesion's margin. However, these cells can also be randomly distributed throughout the lesion. In the more stained areas, which are placed more centrally, round or tiny spindle cells are grouped around thin-walled, irregularly branching, hemangiopericytoma-like blood vessels. Basophilic nuclei, tiny eosinophilic cytoplasm, and unclear cell borders characterize these cells.[5]

The bright and dark areas are often not separated, and both cell subpopulations are mixed.[21] Mitotic figures are rare, although deep-seated lesions are frequently ill-defined and tend to infiltrate the surrounding tissue.[5]

However, since other tumors, such as leiomyoma, schwannoma, nodular fasciitis, benign fibrous histiocytoma, solitary fibrous tumor, desmoid-type fibromatosis, and infantile fibrosarcoma mimic the histopathologic findings of myofibroma, they had to be considered in the differential diagnosis.

Immunohistochemistry becomes an invaluable tool in this condition to achieve a definite diagnosis. Analyzing a panel of markers, including α-SMA, vimentin, desmin, S100, and CD34+, helps the differentiation of these neoplasms.

Very strong positivity to α-SMA in most areas and to vimentin in a few areas is noted in myofibromas. Leiomyoma shows positive staining to desmin, neurofibroma to S100, and solitary fibrous tumor to CD34+. Faint positivity to α-SMA is also noted in nodular fasciitis.[1,9] In our case, the neoplasm showed strong positivity to α-SMA. It was evident even in the walls of the blood vessels. Vimentin was also positive in a few areas but was negative for desmin, S100, and CD34+. As for h-caldesmon positivity, several studies have demonstrated that most of myopericytomas co-express α-smooth muscle actin (SMA) and h-caldesmon.[23,24] Expression of h-caldesmon has been reported in myofibromas,[25] but needs to be validated in studies conducted at a larger scale. Based on the IHC findings in the present case, leiomyoma, neurofibroma, nodular fasciitis, and solitary fibrous tumor were ruled out, and a definitive diagnosis of myofibroma was made.

The treatment of myofibroma is the excisional biopsy. Wide surgical resection is crucial since these benign tumors frequently infiltrate and implicate the adjacent healthy tissue. Local recurrence has been documented in 7% to 31% of cases,[15] although an incomplete excision of the lesion mostly causes relapses. In rare cases of chronic recurrence or unresectable lesions, chemotherapy or radiotherapy might be a treatment option.[16] Our patient received an extensive local excision, along with alveolectomy, to address the pressure-induced resorption of the alveolar bone.

To conclude, Myofibroma is a benign tumor with a good prognosis that occurs infrequently in the oral cavity. It must be included in the differential diagnosis of other oral mucosal lesions. It is critical to be aware of this benign tumor to avoid misdiagnoses and unnecessarily invasive treatments. In all situations, extensive surgical excision with appropriate safety margins is the ideal therapy, and careful post-operative surveillance should be continued. In this example of myofibroma, there were no postoperative problems or recurrence after one year.

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- *Ethics Statement* -
  Informed consent was obtained from the patient. The treatment was conducted following the Helsinki Declaration.

- *Conflict of Interest.*
  The authors declare that there is no conflict of interest.

- *Financial Support. None*

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