



RESEARCH ARTICLE

SOLITARY MYOFIBROMA OF MANDIBLE-A CASE REPORT

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ABSTRACT

Myofibroma is a rare spindle cell neoplasm that consists of myofibroblasts (cells with both smooth muscle and fibroblastic features). Although it was originally described as a multicentric tumor process affecting infants and young children (myofibromatosis), it is now recognized that most cases of the tumor are solitary and that it can occur at any age. This report describes a case on solitary myofibroma involving posterior part of body of mandible extending to the ramus occurring in a 15 years old patient.

Key Words:

Myofibroblast,  
Biphasic Appearance,  
Storiform, Mandible.

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INTRODUCTION

Myofibroma is a benign mesenchymal neoplasm (Cargini *et al.*, 2012). 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 localizations (Behar *et al.*, 1998). The most common oral location is the mandible followed by the lips, cheek and tongue. The tumor is typically a painless mass that sometimes exhibits rapid enlargement. Intrabony tumors create radiolucent defects that usually tend to be poorly defined although some may be well defined or multilocular (Neville *et al.*, 2007). Histopathologically, myofibromas are composed of interlacing bundles of spindle cells with tapered or blunt-ended nuclei and eosinophilic cytoplasm.

Nodular fascicles may alternate with more cellular zone imparting a biphasic appearance to the tumor. Centrally, the lesion is often more vascular with a hemangiopericytoma-like appearance. The tumor cells are positive for smooth muscle actin and muscle-specific actin with immunohistochemistry, but they are negative for desmin (Neville *et al.*, 2007). Behavior of these lesions varies depending on the site. A solitary myofibroma usually treated by wide local excision to include normal adjacent tissue. 25% of the tumors treated in this manner will recur, these can be controlled with reexcision. Large expansile lesions require en block resection (Sapp Philip *et al.*, 1997). The prognosis in solitary and multicentric forms without visceral involvement is excellent, whereas the visceral involvement is potentially fatal (Cargini *et al.*, 2012). Here, we are presenting a case of 15 years old otherwise healthy female with swelling involving left posterior part of body of mandible extending to the ramus which was diagnosed as myofibroma based on clinical, radiographic, histopathological and immunohistochemical features.

**Case report:** A 15 years female patient, from a semi urban area had reported to a private clinic with the chief complain of a painless swelling involving the left side of lower jaw region since past 03 months.

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The swelling was rapidly enlarging and asymptomatic. Initially, it was small in size, but had later grown gradually to attain the present dimension. The past medical and dental histories as well as the family histories were non-contributory. The patient was of average height and weight. Extra oral examination revealed a well demarcated, large swelling involving the left side of the posterior part of the body of mandible (Fig-1). On palpation, the swelling was well localized, firm in consistency, non mobile, measuring about 3.5x3 cm in diameter, non tender, fixed to the underlying bone and non adherent to the superficial skin. No regional lymph nodes were palpable. Intraorally no abnormality was detected. No paresthesia was noted.

Orthopantomogram (OPG) revealed a large, relatively well-demarcated, unilocular radiolucency involving mandible extending from the 37, 38 region to ramus, with destruction and thinning of lower border of mandible (Fig.-2). Considering the history, clinical and radiological findings a provisional diagnosis of benign odontogenic neoplasm was made. An incisional biopsy was performed under local anesthesia after routine blood evaluation (no significant alteration was noted) and informed consent from the patient, for histopathological evaluation. Microscopic features of the H & E stained section showed biphasic stromal tumor mass proliferating in nodular pattern, composed of alternating light and dark stained zones (Fig.-3a). Peripherally, light staining zone is composed of plump myoid spindle cells (Fig.3b, 3c) with eosinophilic cytoplasm & elongated and tapering nuclei arranged in nodules. Interlacing bundles of spindle cells were also in a storiform pattern (Fig.-3d). Some foci shows extensive hyalinization (Fig.-3b). The centrally, dark staining areas are composed of round to polygonal cells (Fig.-3b) with mild pleomorphic hyperchromatic, vesicular nuclei arranged around hemangiopericytoma like vasculature (Fig.3e). Focal areas of calcification seen. The overall histopathological features were suggestive of Myofibroma. Immunohistochemical analysis was done for confirmative diagnosis.

Smooth Muscle Actin (SMA) was positive in tumor cell (Fig.4a) in Tram Track Pattern (Fig.4b) & in normal vessels wall (Fig.4b) but Desmin was negative in tumor cells. Therefore, a final diagnosis of "Myofibroma" was made on the basis of clinicopathological and immunohistochemical parameters. Based upon the above diagnosis the patient was referred to the department of Oral and Maxillofacial Surgery for further management and treatment.

## DISCUSSION

In the *WHO Classification of tumours of soft tissue and bone* revised in 2013, myofibroma was classified as a perivascular tumor, as well as angioleiomyoma (Fletcher, 2017). Solitary myofibroma is nonaggressive, benign myofibroblastic proliferations that are relatively common in the head and neck. The clinical appearance is nonspecific, such as enlarging mass that may or may not be painful; and secondary ulceration of the mucosal surface (Poon Chiu-Kwan and Kwan Po-Cheung, 2005). In the present case, the patient reported with slow enlarging, firm, non mobile, painless swelling involving the left side of lower jaw region without any surface ulceration. Radiographically, the lesions appear as unilocular or multilocular radiolucencies with a variably thick sclerotic rim (Allon *et al.*, 2007). Some cases show thinning of the cortical

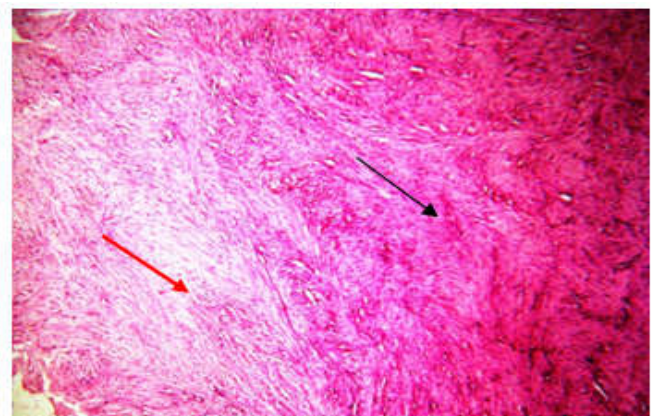
plate and displacement of adjacent teeth (Sugatani *et al.*, 1995).



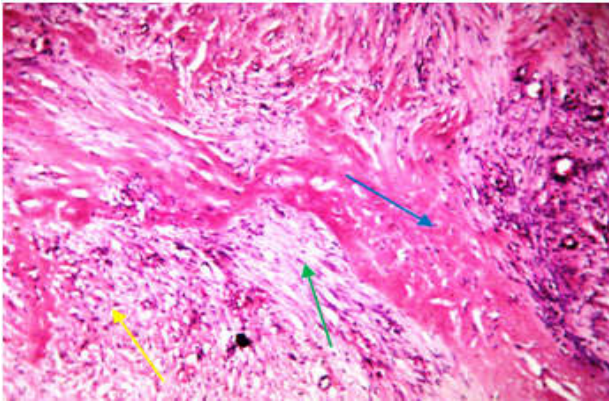
**Fig. 1. Extra oral photograph showing large swelling involving the left side of the posterior part of the body of mandible**



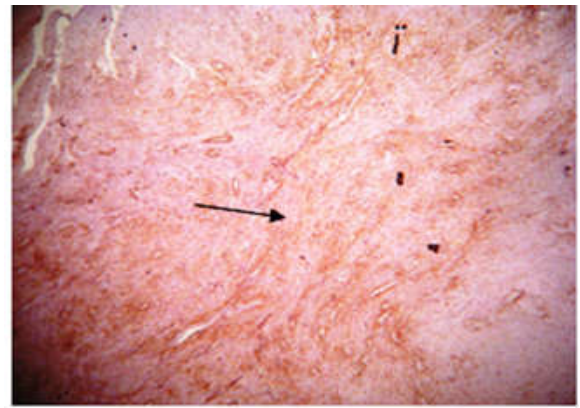
**Fig. 2. Orthopantomogram (OPG) showing a large, relatively well-demarcated, unilocular radiolucency involving mandible extending from the 37, 38 region to ramus, with destruction and thinning of lower border of mandible**



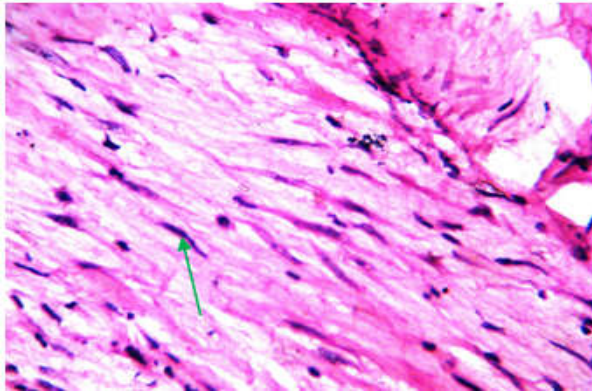
**Fig. 3a. Photomicrograph showing light (red) & dark (black) staining biphasic pattern zones of tumor (H&E, 10X)**



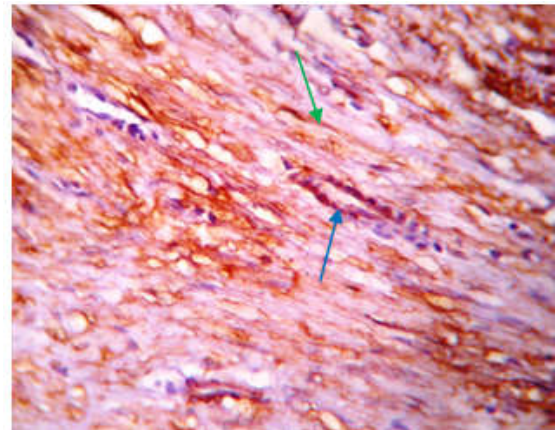
**Fig. 3b.** Photomicrograph showing myoid spindle cells (green) central round cells (yellow) hyalinization (blue) (H&E, 10X)



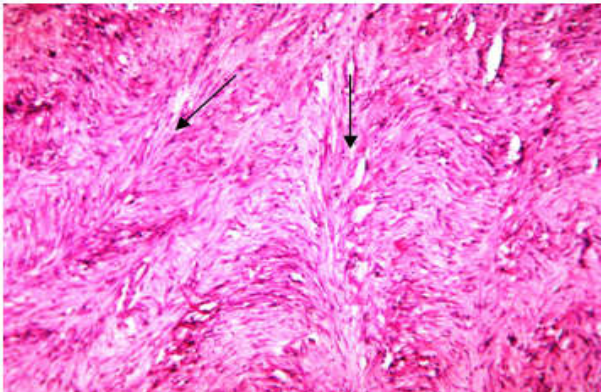
**Fig.4a.** Photomicrograph showing SMA positive tumor spindle cells (black) (H&E,10X)



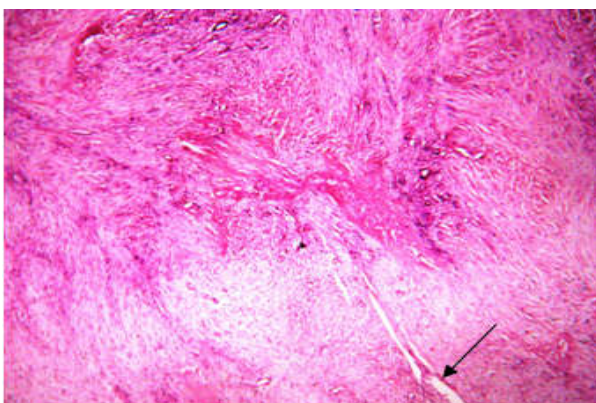
**Fig. 3c.** Photomicrograph showing myoid spindle cells with eosinophilic cytoplasm & elongated and tapering nuclei (H&E, 40X)



**Fig.4b.** Photomicrograph showing, Tram Track pattern positivity of spindle cells (green), normal vessels (blue) (H&E, 40X)



**Fig.-3d** Photomicrograph showing spindle cells are arranged in a storiform pattern (H&E, 10X)



**Fig. 3e.** Photomicrograph showing hemangiopericytoma like vasculature (H&E, 10X)

The present case was imaged as a relatively well-demarcated, unilocular radiolucency involving mandible extending from the 37, 38 tooth region to ramus, with destruction and thinning of lower border of mandible. Histologically myofibroma exhibits a biphasic pattern of light and dark-stained areas. The light area mainly consists of spindle cells with eosinophilic cytoplasm and tapering or cigar-shaped nuclei, arranging in short fascicles or whorls and nodules, at the periphery of the lesion. However, sometimes these cells are distributed haphazardly throughout the lesion. In contrast, the dark stained area located more centrally, consisting of round cells or small spindle cells arranged around thin-walled, irregularly branching, hemangiopericytoma-like blood vessels. These cells have basophilic nuclei, small eosinophilic cytoplasm and indistinct cell margins (Weiss, 2001). Immunohistochemically, myofibroma cells express SMA, specific muscle actin, vimentin and are negative for desmin, S-100 and CD34 (Andreadis, 2012). In the present case, Smooth Muscle Actin (SMA) was positive in tumor cell in Tram Track Pattern & in normal vessels wall but Desmin was negative in tumor cells. In our case, histopathological and immunohistochemical findings fulfill the criteria for a diagnosis of myofibroma. Histopathologic differential diagnosis of myofibroma of oral soft tissues includes leiomyoma, schwannoma, nodular fasciitis, benign fibrous histiocytoma and solitary fibrous tumor, desmoid type of fibromatosis and infantile fibrosarcoma. The neoplastic cells of vascular leiomyoma are positive for desmin in contrast to negative in myofibroma. Schwannoma and neurofibroma do not contain hemangiopericytoma like blood vessels and are negative for SMA (Andreadis, 2012).

Nodular fasciitis demonstrates extravasated red blood cells, myxoid stroma, and chronic inflammatory cells, features that are absent in myofibroma (Vered *et al.*, 2007). Benign fibrous histiocytoma consists of fibroblast-like spindle cells and histiocytic-like cells arranged in a storiform pattern (Andreadis, 2012). Solitary fibrous tumor may be differentiated from myofibroma because its neoplastic cells express CD34 (Westra *et al.*, 1994). Unlike myofibroma, desmoid-type fibromatosis is not characterized by biphasic cellular populations. Finally, infantile fibrosarcoma is characterized by the presence of uniform spindle cells that form fascicles, focal necrosis and hemorrhages, features that are not normally seen in myofibroma (Weiss, 2001). The recurrence rates for myofibromas are quite low, ranging from 0% to 12.5%. Recurrences can be attributed to tumors with difficult surgical access or incomplete removal. Smaller lesions can regress spontaneously, and observation might be warranted in those cases (Lee Yong-Moon, 2014). In our case, the patient was also advised for complete excision to reduce the post operative morbidity as patient is of very young age group and referred to the oral surgery department for further management and treatment.

### Conclusion

An important clinical aspect is represented by the rapid growth of myofibroma in a young subject may suggest the diagnosis more aggressive and proliferative neoplasm. The clinical approach requires a careful clinical evaluation of symptoms and signs, followed by an accurate diagnosis by means of histopathological and immunohistochemical examinations. Awareness of myofibroma arising in the mandible also may lower the possibility of misdiagnosis and it will allow for the selection of the best individual therapeutic approaches, increasing the treatment efficacy in patients diagnosed with this lesion. Thus, the Clinicopathological, diagnostic and treatment modalities of myofibroma, in general and a lesion involving the mandible, in particular is discussed herewith.

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