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A solitary intraosseous myofibroma of mandible: A case report

Fereshteh Baghaii[1], Mohammad Reza Rokni[2], Sedigheh Rahrotaban[1], Maryam Jolehar[1], Azadehzeinab Titidezh[3], Samira Derakhshan[1*]

1- *Department of Oral and Maxillofacial Pathology of dentistry school, Tehran University of Medical Science, Tehran, Iran.*

2- *Private clinician, Tehran, Iran.*

3- *Department of Oral and Maxillofacial Pathology of dentistry school, Qazvin University of Medical Science, Qazvin, Iran.*

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**Corresponding author:*
Samira Derakhshan
Department of Oral and Maxillofacial Pathology of dentistry school, Tehran University of Medical Science, Tehran, Iran.

Tel: +98-21-22374866
Tel : +98-21-22374866
Fax: +98-21-22646531
Email: Samirad86@yahoo.com

ABSTRACT

Myofibroma is an uncommon benign mesenchymal neoplasm. Solitary myofibroma is common in soft tissues of head and neck, but rare in the jaw bones. Only a few sporadic cases of solitary myofibroma of mandible have been described. The aim of the present study was to present a clinical case of a patient with a myofibroma in mandible bone and its management. Differentiating this lesion from other benign and malignant neoplasms is crucial in deciding between a radical and a conservative treatment approach. We explained that diagnosis of myofibroma can be reached by a histopathologic and immunohistochemical analysis and surgical excision is the treatment of choice.

Key words: Intraosseous, Mandible, Myofibroma.

Introduction

Myofibroma is an uncommon benign neoplasm typically arises in soft tissues affecting all ages [1]. The terms myofibroma (solitary) and myofibromatosis (multicentric) were adopted by WHO to describe the benign neoplasms of contractile myoid cells arranged around thin walled vessels [2]. Myofibroma involves predominantly the head and neck region (36%) or the trunk, however cases of the jaws are rare [3]. Only a few sporadic cases of solitary myofibroma of mandible have been described. In such cases, the lesions occur more commonly in children [4]. In adults the development ofibroma of mandible have been described. In such cases, the lesions occur more commonly in children [4]. In adults the development of a solitary intraosseous myofibroma is even rarer [5]. Central myofibroma of the jaws involve the teeth and exhibit alarming clinical or radiographic features suggestive of an odontogenic cyst/tumor or other non-odontogenic lesions [1,6]. In addition, histopathologic feature of this lesion has great potential for confusion with the more aggressive spindle cell tumor [1]. because of this reasons, diagnosis of myofibroma may be a challenge for clinician and need to immunohistochemical analysis.

In this article we present a case of solitary myofibroma of mandible in a 17-year-old boy with detailed description of clinical, radiographic, histopathological, and immunohistochemical findings.

This article was approved by Ethics Committee of University of medical sciences, Dental school, Tehran, Iran. Informed consent was obtained from the patient.

Case report

A 17-year-old boy was referred to oral and maxillofacial surgeon to evaluate a mandibular lesion that his dentist had found during routine examination. The clinical examination revealed a very mild swelling in buccal aspect of right mandibular premolars. Mucosa over the swelling was clinically normal. There was no associated pain or paresthesia. No extra oral swelling was found. There is no lymphadenopathy. No noticeable past medical history was found.

Periapical radiography revealed a radiolucent lesion with well-defined sclerotic borders in pre molars area (fig 1). It seemed the lesion has caused to mild diversion of premolars roots (fig 1).

Excisional biopsy of the lesion was performed under local anesthesia and the specimen was sent for histopathologic examination. The histopathologic finding showed a well-circumscribed mass with fascicules of spindle cells with oval or round nuclei in a collagenous stroma (fig 2). Spindle cells arranged in different directions. Numerous vessels especially in the periphery of tumor mimicking the hemangiopericytoma pattern with multiple slit-like vascular spaces was noted (fig 3). Atypical cell and mitotic figures are not seen.

Immunohistochemical staining was carried out for desmin, beta catenin, Ki67, S100, αSMA, CD34, and C-kit (CD117). Positive immunoreactivity was observed for beta catenin, Ki67 and αSMA and negative immunoreactivity for S100, desmin, CD117 and CD34, thus confirming the myofibroblastic nature of the tumor (fig4).

After more than one year follow up, there is no clinical and radiographic evidence of recurrence (fig5).

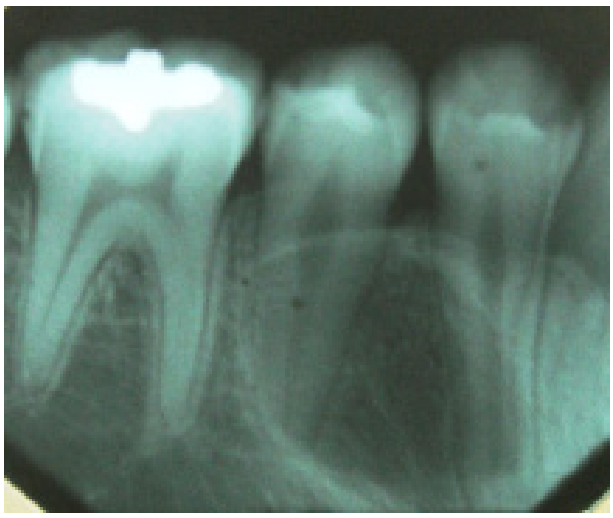

Figure 1. Periapical radiography shows a radiolucent well-defined lesion.

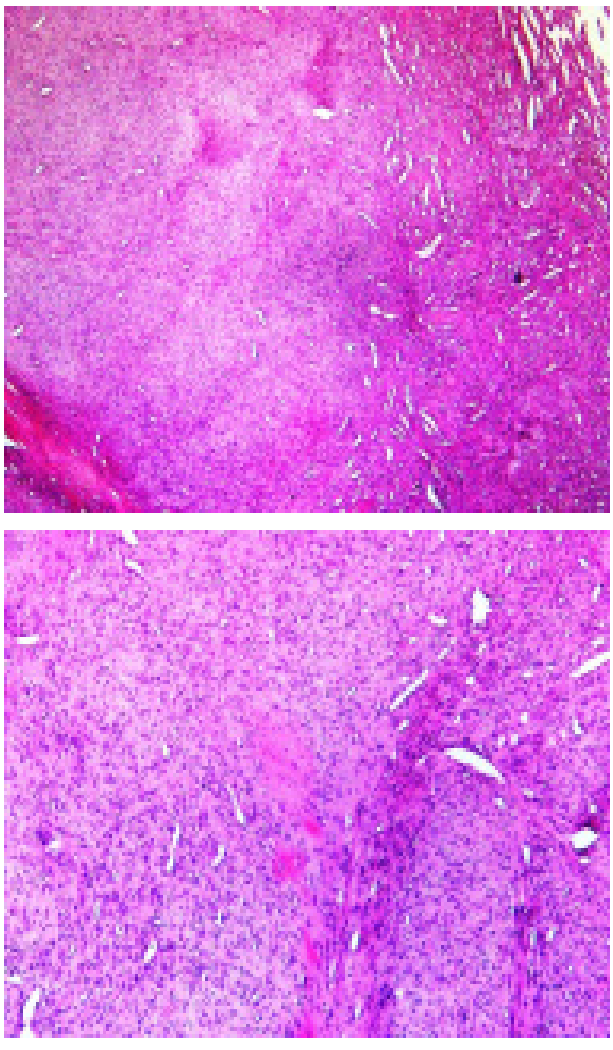

Figure 2. Histopathologic picture of the lesion. a)benign biphasic tumor (H-E staining, original magnification ×10). b) cells with small, round nuclei and spindle cells forming short fascicles or whorls (H-E staining, original magnification ×40).

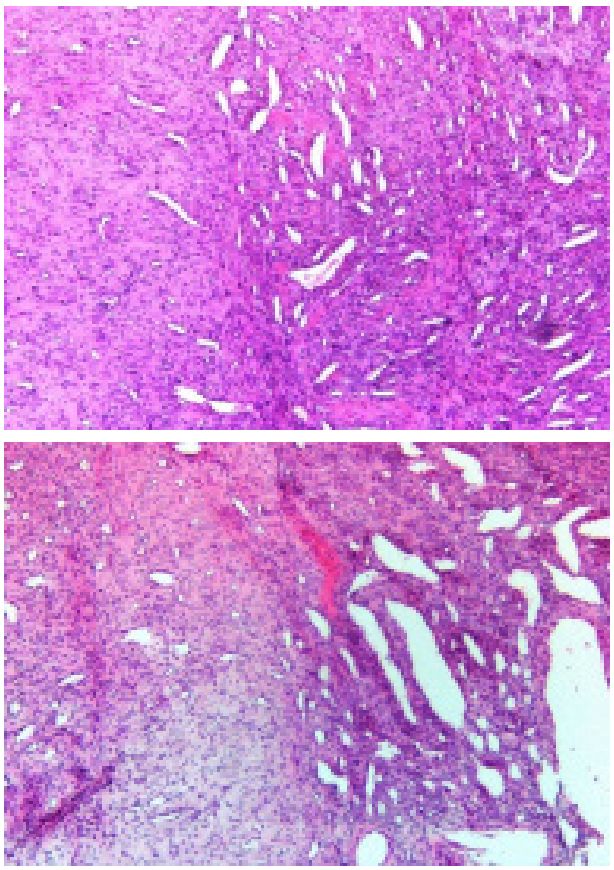

Figure 3. Prominent vascular pattern mimicking the appearance of hemangiopericytoma with multiple slit-like vascular spaces in different sizes. (H-E staining, original magnification ×40).

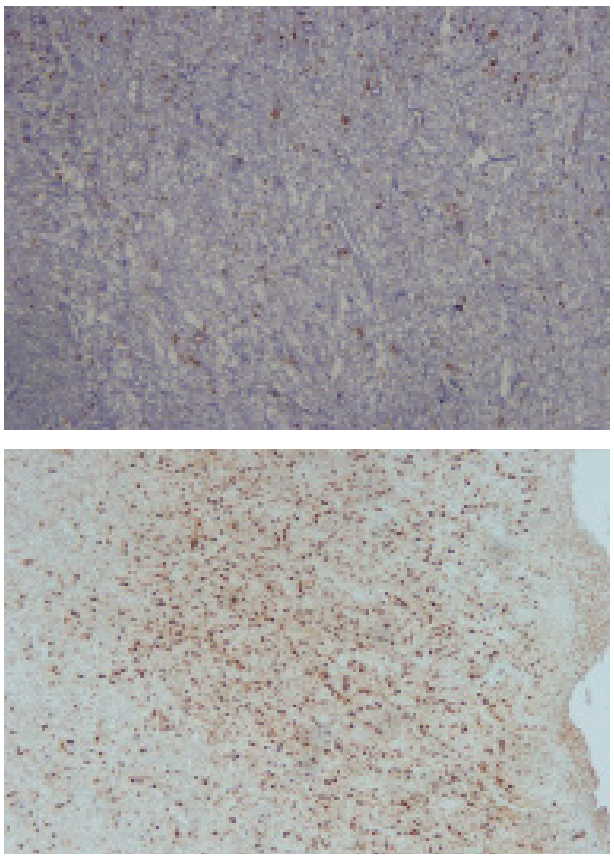

Figure 4. a) Positive immunoreactivity of 10% of **tu**moral cells with Ki67 (original magnification ×10). b) Positive immunoreactivity of tumoral cells with anti-SMA antibody (original magnification ×10).

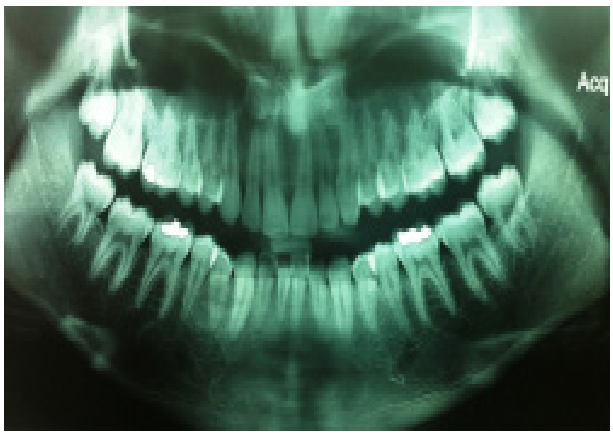

Figure 5. Panoramic view of patient 1 year after excision with bone formation and no signs of lesion recurrence.

Discussion

Myofibroma is a rare benign tumor presenting as a solitary or multiple lesions with a predilection for soft tissues of the head and neck region. It occurs less common within the jaws. It is thought to represent a benign proliferation of the myofibroblast, a cell with a phenotype of both fibroblast and smooth muscle cell, as demonstrated by immunohistochemical, histomorphologic, and ultrastructural studies [7]. Myofibroma of the mandible is commonly diagnosed in children in the first decade of life and shows a definite male predilection. Clinically, lesions present as an asymptomatic jaw swelling and rarely as a soft tissue mass when there is cortical plate perforation [8]. According to Kauffman and Stoutthe prognosis of myofibroma depends on the lesion location [9]. Lesions with a good prognosis affect the skin, subcutaneous tissue, or skeleton, and those with a poor prognosis affect the soft tissue, muscles, bone or internal organs [9]. Our case treated by surgical excision of the lesion without any evidence of recurrence.

Although radiologically, myofibromas are usually unilocular radiolucent lesions with well-defined borders but it can show a multilocular appearance [8]. Our case also revealed a unilocular osteolytic lesion. When myofibroma shows an ill-defined borders, aggressive lesions like desmoplastic fibroma and Ewing's sarcoma shoud be noted [10]. Occasionally, there may be evidence of cortical expansion and/or perforation, which may be seen only on CT examination [4,6,10].

Histologic diagnosis of myoifibroma because of its similarity to other spindle-cell lesions is difficult. It

must include the tumors of muscle and neural origin such as lieomyoma and neurofibroma [11] and certain tumours like desmoplastic fibroma, fibromatosis and low-grade fibrosarcoma [8].

Immunuhistochemistry is done to confirm the diagnosis. In Myofibromas the cells are immunoreactive for vimentin and the smooth muscle actin, but negative or inconsistently positive for desmin, S-100, CD34 and CD68. In the present case, immunohistochemical staining was decisive for establishing the correct diagnosis, as reported previously for other cases [3,4,8,11,12,13,14 and 15]. Spindle cell lesions of nerve tissue origin is immunopositive for S100, which is absent in myofibroma. Leiomyoma of bone are rare, and can be excluded on the basis of their immunoreactivity for desmin, which is negative in myofibroma.

Aggressive fibromatosis and fibrosarcoma distinguish from myofibroma by more monophasic growth pattern comprising long fascicles of spindle cells among abundant wavy collagen fibrils in fibromatosis [4,16] and "herring bone" pattern, nuclear atypia and high mitotic counts including abnormal mitoses in fibrosarcoma [4,10,17].

Differentiation from solitary fibrous tumor may also be difficult because of hemangiopericytoid appearance in both lesions [8]. Solitary fibrous tumor has less proliferation of spindle cells and hypercellular and hypocellular areas rich in a dense keloid type of collagen [8,18]. In addition, it is immunopositive for CD34 and CD99 which is negative in myofibroma [19].

The infiltrative and destructive growth pattern of desmoplastic fibroma (DF) and the absence of hemangiopericytoma-like vascular pattern can help in differentiation between DF and myofibroma [8].

Treatment of myofibroma of the mandible is usually conservative excision [8]. Some myofibromas, especially in the young involute without treatment [20]. In the present case, the small extent and easy detachment of the lesion from the mandibular bone allowed conservative surgical excision with preservation of the tooth. The outcome until now (more than 1-year follow-up) is excellent.

In conclusion, myofibroma presents a wide range of differential diagnosis, including benign and malignant neoplasms. Therefore, accurate diagnosis may avoid an unnecessarily aggressive therapy. This aim can be reached by combination of histopathologic and immunohistochemical analysis.

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