Case Report

Myofibroma of the body of mandible: A case report of a solitary lesion

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Abstract

Myofibromas are rare benign lesions and are often found as solitary entities. The treatment of the lesion is complete excision and the recurrence is rare. A 56-year-old female presented to the Oral and Maxillofacial Surgery clinic for further evaluation and management of a solitary lesion of the right body of the mandible that was first noticed incidentally by her referring dentist. An incisional biopsy was first performed in the clinic with the diagnosis of myofibroma. The patient was then treated with complete excision of the lesion and extraction of the adjacent teeth. The final biopsy report confirmed the initial diagnosis of myofibroma. Intraosseous solitary lesion of the mandible is a rare lesion with a handful of reported cases. Uniquely, the diagnosis of myofibroma in this 56-year-old is the oldest reported case of myofibroma. There are distinct histopathological features of the lesion that distinguishes this entity from other closely resembling lesions.

Keywords: Excision, intraosseous, lesion, mandible, myofibroma

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INTRODUCTION

In 1989, the term myofibroma was first used to describe solitary lesions arising in adults,[1] that were histopathologically identical to multifocal infantile myofibromatosis lesions commonly found in infants.[2] Myofibroma presents as solitary or multicentric and is recognized as a separate entity distinct from multifocal myofibromatosis.[3] Solitary myofibroma have been reported to appear predominantly in the soft tissues of the head and neck in adults,[4] including on the tongue, palate, gingiva, and retromolar pad region.[4] Rarely, solitary myofibroma have been described in the literature to be located centrally in the mandible.[4] These lesions are typically diagnosed in children, usually in the first decade of life.[3] On exceptionally rare occasions, intraosseous myofibroma has been documented in adults.[5] Since the first documented solitary adult intraosseous myofibroma of the mandible in 2002, only a few cases have been reported.[2]

Microscopically, myofibroma is a rare benign neoplasm of contractile spindle cell myofibroblasts,[6] and displays ultra-structural features of myofibroblastic differentiation, cytoplasmic filaments with dense bodies, basal lamina, pinocytotic vesicles, and nuclear invaginations. Immunohistochemical studies similarly reveal myofibroblastic typia: Actin reactive, focal desmin expression, non-reactive with S100 and keratin.[3] Clinical presentation of the lesion includes an asymptomatic

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buccolingual mandibular swelling with occasional associated intraoral mucosal swelling.[3] Radiographically, myofibroma are typically unilocular, radiolucent lesions with well-defined borders.[7] Due to the rare presentation of intraosseous myofibroma coupled with a nebulous clinical, immunohistochemical, and radiographic interpretation, more concerning misdiagnoses and subsequent treatment may ensue.

The purpose of this case report is to describe a rare occurrence of intraosseous solitary myofibroma in a 56-year-old female, the oldest reported patient (to the best of the authors' knowledge) with regard to the clinical, radiographic, and immunohistochemical features, as well as the surgical treatment of the lesion.

CASE REPORT

A 56-year-old female was originally seen by her general dentist in the midsummer of 2020. A radiolucent lesion of the right mandible was noted on the orthopantomogram. Her dentist then appropriately referred the patient to Indiana University Oral and Maxillofacial Surgery (IU OMFS) outpatient clinic for further evaluation and definitive management. The patient's medical history included hypertension, asthma, and osteoarthritis but no previous history of any bony lesions or malignancy. On clinical examination, the patient denied any history of pain, paresthesia, tingling, or burning sensation to her mandible or tongue. Furthermore, she denied any history of a foul taste or smell intraorally. Some buccal bony expansion of the mandible was noted, and teeth #29, and 31 demonstrated class II mobility but tested positive to routine vitality "cold" testing.

The panoramic image from November 2020 [Figure 1] revealed a radiolucent lesion of the right posterior mandible with well-defined borders. The lesion was pyramidal in shape and extended mesiodistally from the root apex of tooth #29 to the apex of #31, measuring approximately 23 mm in the A-P dimension. The superior margin of the lesion approximated the crest of the alveolus and measured 11 mm in height.

Given the size of the lesion and its presentation, a cone beam CT scan was obtained to further delineate the dimensions of the lesion and the presence of any cortical perforations or involvement of the inferior alveolar nerve canal [Figure 2].

Following a discussion with the patient on the clinical and radiological findings, an incisional biopsy of the lesion was recommended to determine the pathological diagnosis prior to definitive surgical intervention.

During the biopsy procedure, the lesion was appropriately de-roofed with careful removal of a bony window to avoid perforating the lining. An adequate sample of tissue was obtained, placed in formalin solution, and submitted to the Oral Pathology department at Indiana University School of Dentistry for histopathological examination.

Histology confirmed the diagnosis of myofibroma with interlacing bundles of spindle cells with tapered nuclei and eosinophilic cytoplasm. Furthermore, no odontogenic epithelium was reported. [Figures 3-5].

The immunohistochemical profile of the lesion with strong reactivity to Alpha- smooth muscle actin and vimentin, and negative for Desmin and S 100 confirmed the final diagnosis of myofibroma. No additional head and neck intra-bony lesions were noted based on clinical and radiological examination; therefore, myofibromatosis was excluded. The patient was then scheduled for follow-up and planned for definitive excision of the lesion under general anesthesia. Intra-operatively, the lesion was completely excised and a peripheral ostectomy was

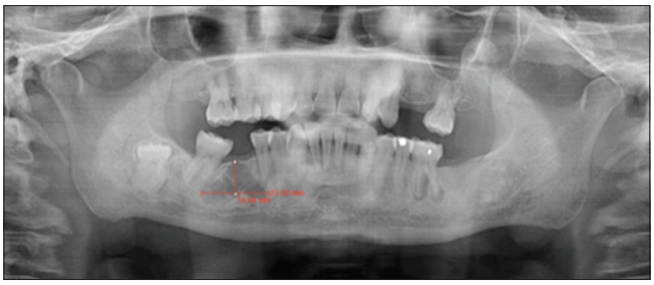

Figure 1: Orthopantomogram of patient taken at initial consultation visit demonstrating a well-defined radiolucent lesion of the right body of the mandible

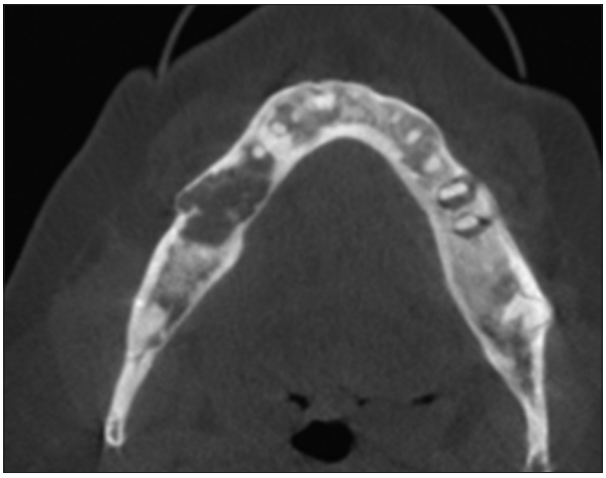

Figure 2: Axial cone beam CT scan slice of the mandible demonstrating the right mandibular lesion. Note the buccal and lingual cortical thinning and mild buccal cortex expansion

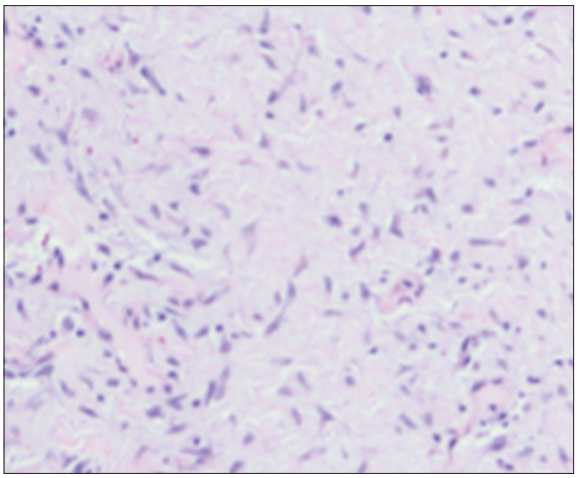

Figure 3: Photomicrograph with H&E stain depicting the nature of myofibroma with bundles of spindle cells with oval nuclei. (Photomicrograph courtesy of Dr. Ritchie. 4x magnification)

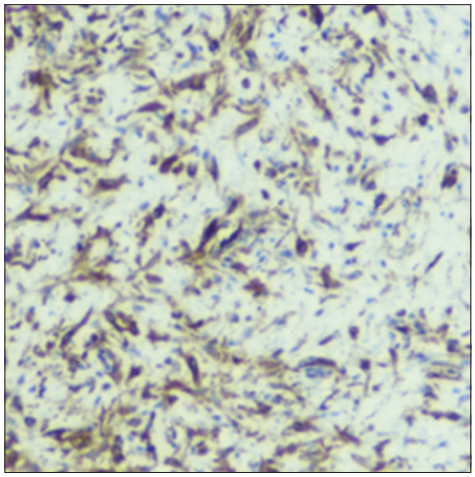

Figure 4: Photomicrograph with Alpha- Smooth Muscle Actin (SMA) with strong immunoreactivity. (Photomicrograph courtesy of Dr. Ritchie. 20x magnification)

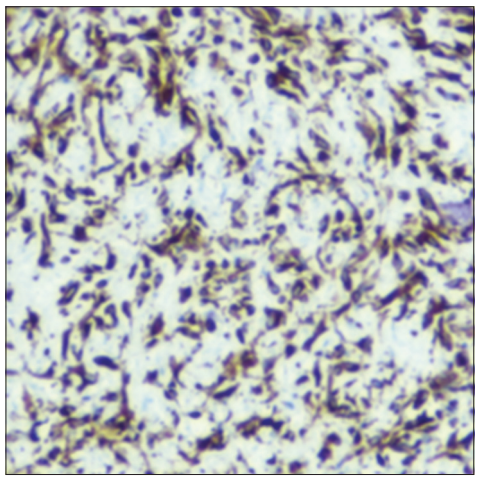

Figure 5: Neoplastic cells of myofibroma reveal positive reaction with vimentin stain. (Photomicrograph courtesy of Dr. Ritchie. 20x magnification)

completed around the lesion. The inferior alveolar nerve was not visualized. Teeth #29, and 31 were assessed to be mobile, with a poor prognosis, and were extracted to ensure complete removal of the lesion. Tooth #19 was also extracted as it was previously deemed to be unrestorable and with periapical pathology. The patient has been seen for routine 2-week and 8-week follow-up and has followed an uneventful post-op course with minimal but improving paresthesia of the right mental nerve distribution. She is scheduled for six months follow-up and further imaging to assess bony infill. Subsequently, the pathology report also confirmed the initial diagnosis.

DISCUSSION

Myofibroma is a rare benign lesion. It is most commonly reported as a solitary lesion; however, it is also known to be part of myofibromatosis. The solitary lesion is treated by complete excision and reported recurrence appears to be low and is attributed to incomplete excision.[3]

Intra-bony myofibroma in the head and neck region are rare entities. There have been a handful of cases reported in the head and neck region, with the majority occurring in the mandible. Mandibular myofibromas tend to be asymptomatic and may first be noted incidentally on routine panoramic imaging. Table 1 provides a summary of reported cases in recent litrature. Characteristically, the lesion exhibits a slow growth rate, but cortical expansion has been reported in the literature. Typically, myofibroma presents as a well-defined radiolucent lesion with a distinct corticated border, which may cause root displacement.[7,8]

Given its rarity, it is not surprising that myofibroma is not routinely included in the differential diagnosis of any radiolucent lesion of the mandible. Other benign lesions and even low-grade sarcomas may be included in the differential diagnosis. This is due to the lytic nature of the lesion and its occasional rapid clinical growth rate. Benign lesions like nodular fasciitis, fibrous histiocytoma, neurofibroma, and haemangiopericytoma could be considered in the differential diagnosis.[9]

The differential diagnosis in this reported case included odontogenic keratocyst and fibrous dysplasia.[3] No malignancy was included in the differential diagnosis due to the absence of pain, paresthesia, root resorption, or cortical perforation. Additionally, the inferior alveolar nerve canal which was in close proximity to the lesion appeared intact without any frank encroachment.

Histologically, myofibromas have been classically described with a series of characteristic features. They have been

Table 1: Summary of reported solitary myofibroma of mandible

Study	Gender	Age	Site	Treatment
Oliver *et al*.[6]	F	34	Ramus of mandible	Surgical excision and bone graft
Sedghizadeh *et al*.[9]	M	20	Body of mandible	Surgical excision and bone graft and titanium mesh
Ramadorai *et al*.[8]	F	32	Body of mandible	Surgical excision and plate
Brierley *et al*.[4]	F	43	Anterior mandible	Curettage of lesion and extraction
Current case	F	56	Body of mandible	Surgical excision and extraction of mobile teeth

described as "biphasic" with zones of polygonal and spindle cell arrangement.[5] Reportedly, the biphasic nature is due to longitudinal and transverse sectioning of the specimen.[1] These arrangements are well demonstrated in Figure 6.

As reported by Brierley *et al*.,[4] the margins of myofibroma may exhibit infiltrative properties with concerns for malignancy and in particular resembling leiomyosarcoma. However, in contrast to myofibroma, leiomyosarcoma exhibits cytological atypia and desmin positivity.[8] Furthermore, several pathological lesions appear to share the spindle cell nature which may lead to the misdiagnosis of myofibroma with other lesions like intraosseous fibromatosis and desmoplastic fibroma.[5] All three of these lesions are positive for smooth muscle actin but have different definitive management approaches. While there is a triad of histological characteristics associated with myofibroma, some of these features are shared with other lesions making diagnosis somewhat challenging. In the case of this reported patient, several attributes of myofibroma were identified, which provided the basis for accurate diagnosis and subsequent management. These included: the presence of spindle cells, S100 Negative, SMA Positive, Vimentin positive, and Desmin negative.[5]

With respect to an anatomical location in the head and neck region, intra-bony myofibroma of the mandible is a very rare occurrence. Only a handful of cases have been reported with ages ranging from 20-43 years, which makes this case the oldest reported case of intra-bony myofibroma of the mandible.

Surgical excision of the tumor is the mainstay of treatment and reported recurrence in the literature is low and may be attributed to incomplete excision.[3]

In summary, myofibroma are rare lesions, especially as it relates to occurrence in the mandible. Accurate diagnosis and exclusion of other lesions that may share similar characteristics are important to guide the appropriate course of treatment and follow-up.

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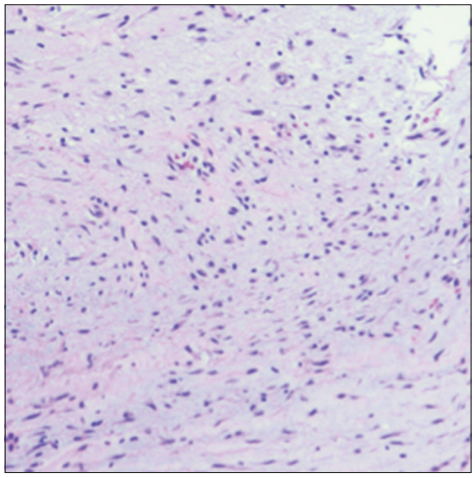

Figure 6: Hematoxylin and Eosin staining of lesion showing polygonal cell areas and spindle cells with elongated football shaped nuclei (Photomicrograph courtesy of Dr Ritchie. 4x magnification)

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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