Case Report

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Myofibroma of Mandible – A Rare Case Report with Review of Differential Diagnosis

Khushboo Singh[*], S Jayachandran[**], Sunita Gupta[***], Mahesh Verma[****]

Abstract

Myofibroma is rare spindle cell neoplasm that consist of myofibroblasts. Previously it was described as multicentic tumour affecting infants & young children (myofibromatosis), now recognised that most of cases are solitary & can occur at any age. Solitary intraosseous myofibroma is a rare finding. This article reports an intraosseous myofibroma involving mandible in a 19-years-old female. The clinical, radiographic and histoptahological features are also discussed.

Key words: Myofibroma; Intraosseous; Mandible.

Introduction

Myofibroma is a rare benign tumor that was originally described as a form of congenital multicentric fibroblastic proliferation by Stout in 1954 [1].The terms myofibroma (solitary) and myofibromatosis (multicentric) were adopted by WHO [2].It shows predilection for the soft tissues of head and neck and intraosseous variety being rare [3].Internal organs such as lungs, kidney, pancreas & gastrointestinal tract have been described as rare sites [3]. Clinically, tumor presents with painless mass which sometimes may exhibit rapid enlargement. As the lesion is completely benign so its extent determines the type of treatment, ranging from conservative surgical excision to the more aggressive excision.

Case Report

A 19 years old female patient reported to the department of Oral Medicine and Radiology with the chief complaint of swelling in right lower jaw region for the past 6 months. History of presenting illness revealed that patient noticed a swelling in lower right jaw in back teeth region 6 months ago which was small at that time and gradually increased to the present size with which she had reported. It was also associated with pain and functional loss of chewing. There was no history of any trauma and patient did not receive any treatment for the same. Past medical was non-contributory. Past dental history revealed history of extraction of 44, 45 due to loosening of teeth one month back. Extraoral examination revealed swelling in right mandibular body region of size 3 × 3 × 2 cm, oval in shape, diffuse margins extending anteriorly from angle of mouth, posteriorly 2 cm anterior of ramus of right mandible, superiorly to upper lip region, and inferiorly to the inferior border of mandible, slightly firm in consistency with normal overlying skin, and tender on palpation (Figure 1). Intraoral examination revealed a solitary growth present in right mandibular alveolus region of size 4 × 3cm, oval in shape, well defined margins extending antero-posteriorly from 43 to 47 region, medially to lingual sulcus, and laterally to buccal vestibule, soft to firm in consistency, overlying mucosa is erythematous in some areas and keratotic in some areas due to impingement of teeth of opposite arch, and tender on palpation (Figure 2).Based on clinical findings and examination, provisional diagnosis was made as benign soft tissue neoplasm involving right mandibular alveolus in premolar region. Differential diagnosis was given as peripheral ossifying fibroma and central giant cell granuloma. Patient was subjected to various radiological examinations. Intraoral periapical radiograph (IOPA) revealed bone

Author's Affilation: [*]Senior Resident, [***]Professor and HOD, Department of Oral Medicine and Radiology, Maulana Azad Institute of Dental Sciences, New Delhi- 110002. [**]Professor and HOD, Department of Oral Medicine and Radiology, Tamil Nadu Government Dental College and Hospital, Chennai – 600003. [****]Professor and HOD, Department of Prosthodontics, Maulana Azad Institute of Dental Sciences, New Delhi- 110002

Reprints Requests: Khushboo Singh, Senior Resident, Department of Oral Medicine and Radiology, Maulana Azad Institute of Dental Sciences, New Delhi- 110002
E-mail: khushbu2424@gmail.com

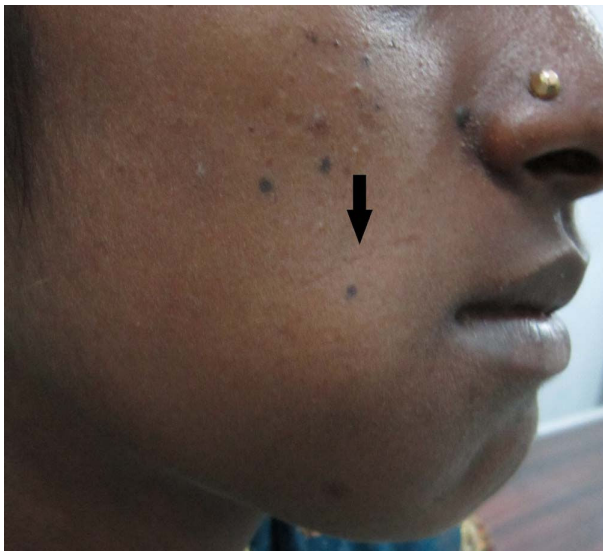

Fig. 1 Photograph showing extraoral lesion

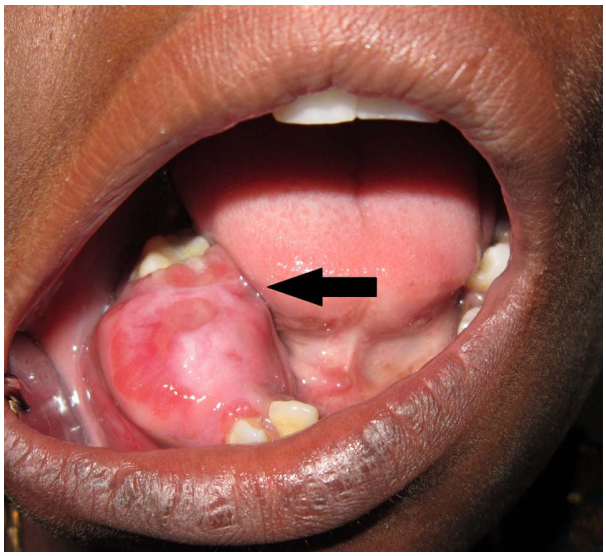

Fig. 2 Photograph showing intraoral lesion

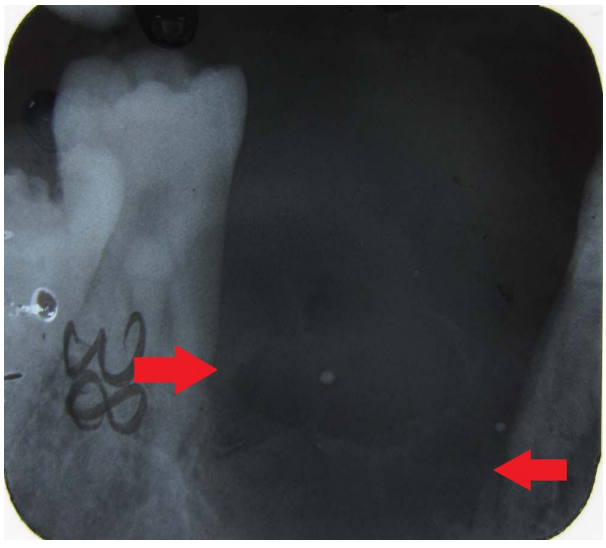

Fig. 3 IOPA radiograph showing radiolucent lesion

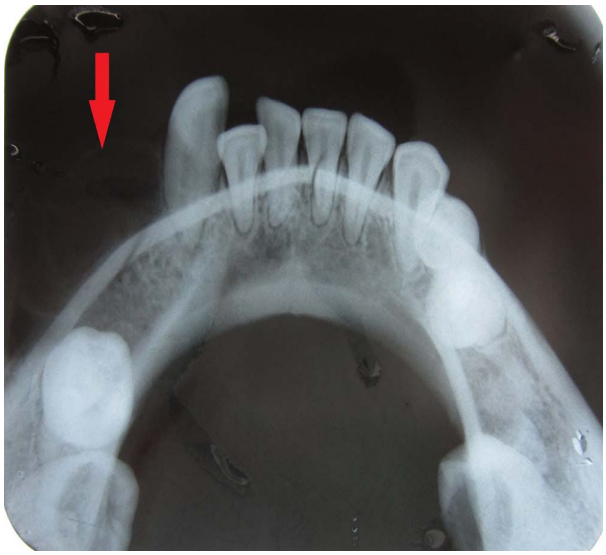

Fig. 4 Mandibular occlusal radiograph showing soft tissue shadow

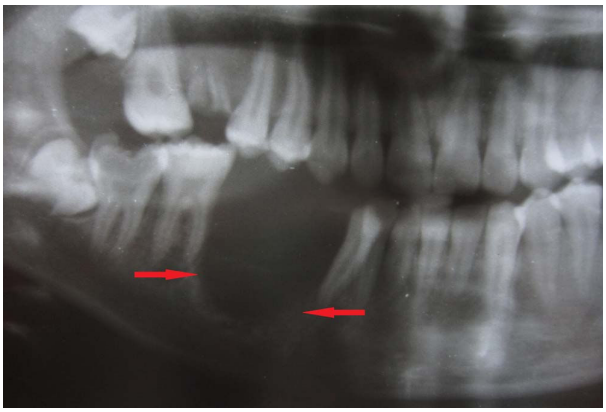

Fig. 5 Orthopantamogram showing well defined radiolucent lesion

loss in 44, 45 region with incomplete delineation of the lesion (Figure 3). Mandibular occlusal radiograph revealed soft tissue shadow having sclerotic borders in premolar region (Figure 4). Orthopantamogram showed arc shaped bone loss extending from root apex of 43 to apex of mesial root of 46 (Figure 5).Computed tomography (CT) scan showed expansile lytic lesion in the body of right mandible involving alveolar process.Overlying cortex appears thinned out& lost in most of the areas. No evidence of septation, calcification within mass (Figure 6 & 7). Incisional biopsy was performed. Histopathological section revealed Fibrocellular stroma with dark & light staining areas. Dark staining areas are made up of round cells & immature cells surrounding hemangiopericytoma like blood vessels. Light staining areas are made up of spindle shaped cells & foamy macrophages in some areas, overall features were suggestive of "Myofibroblastic tumour

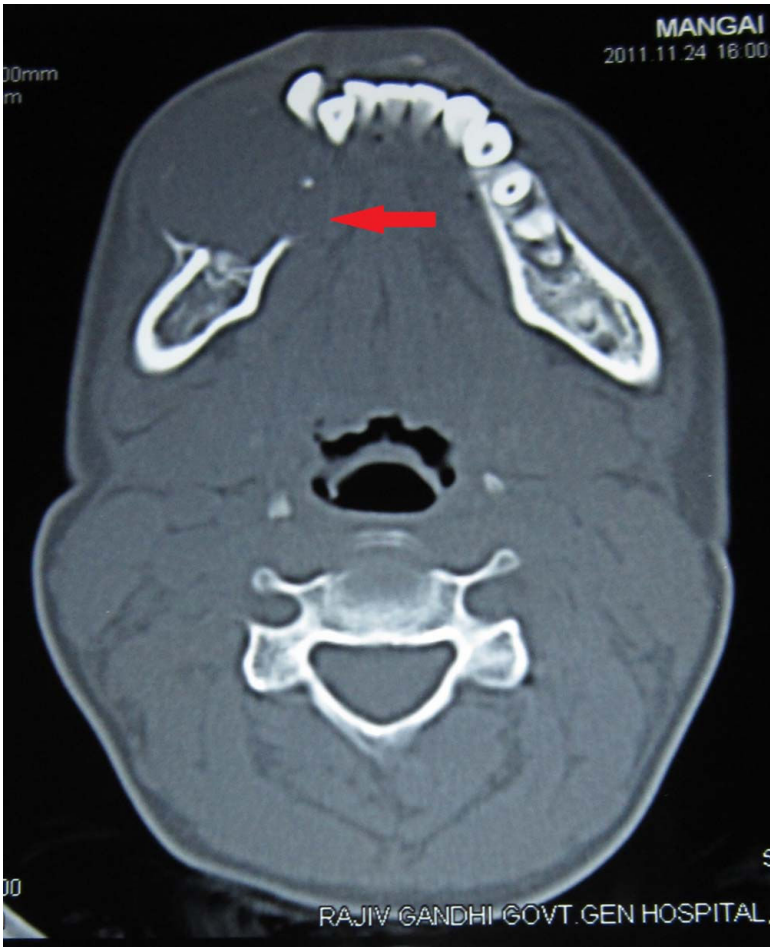

Fig. 6 Axial CT scan section showing homogenous, hypodense lesion

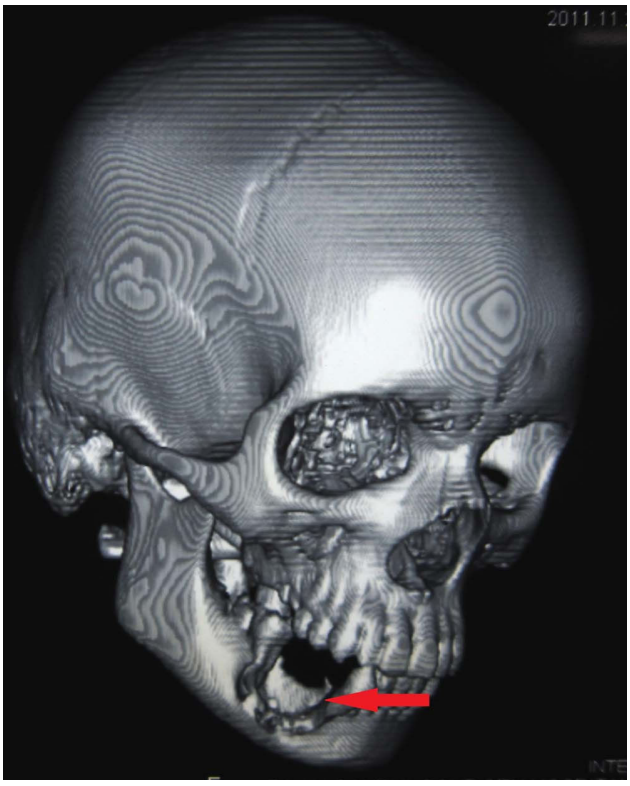

Fig. 7 3D-CT scan section showing osteolytic lesion

(Myofibroma)". Based on this, final diagnosis was given as Myofibroma involving right mandibular alveolus in premolar region. Patient was referred to the department of Oral & Maxillofacial Sugery for further management and Peripheral osteotomy along with surgical excision of the lesion was performed.

Discussion

Myofibroma is characterized by benign proliferation of myofibroblast, a cell having properties of both fibroblast and smooth muscle cell. Myofibroma/myofibromatosis shows predilection for the soft tissues of head and neck, trunk, and extremities, but rarely within bone [4].The etiology of myofibroma is unknown. Few authors have proposed that it is inherited in an autosomal dominant [5] or autosomal recessive trait [6]. To the best of the author's knowledge, less than 40 cases of solitary myofibroma of the mandible have been reported so far in the literature [7]. Myofibroma of the mandible commonly occurs in the first decade with male predilection which is contrary to our case. Intraosseous myofibroma appears as a slow enlarging, asymptomatic, expansile mass and may cause cortical expansion as well as perforation, teeth displacement and root resorption. Radiographically it presents usually as unilocular radiolucent lesion with well-defined borders as presented in our case. However intraosseous myofibroma presents with various radiological features. Allon et al. [7] studied intraosseous myofibromas and stated that they were found commonly solitary radiolucent lesions in the mandible of which 70% were unilocular, 30% were multilocular and 67% had well defined borders. Table 1 shows the radiological differential diagnosis of intraosseous myofibroma [7,8]. Histological features of myofibroma are as given below [9]:

- Typical nodular biphasic pattern with micronodular "Zoning" phenomenon with alternating light and dark areas.
- Light stained areas – fascicles of myofibroblasts with abundant extracellular matrix

  – cells are spindle to ovoid with pale cytoplasm

Table 1 Radiographic differential diagnosis of Myofibroma

Radiographic presentation	Lesion as differential diagnosis
Well defined, unilocular radiolucency	- Unicystic ameloblastoma - Ameloblastic fibroma
Ill defined, radiolucency	- Desmoplastic fibroma - Ewing's sarcoma
Multilocular radiolucency	- Keratocystic odontogenic tumor - Central hemengioma - Ameloblastoma - Central giant cell granuloma - Aneurysmal bone cyst

Table 2 Histopathological differential diagnosis of Myofibroma

Lesion	Differentiating features
• Tumors of neural origin	- S100 immunopositivity present in lesions of neural origin and absent in myofibroma
• Leiomyoma • Leiomyosarcoma	- Desminimmunopositivity present in leiomyoma, leiomyosarcoma& absent in myofibroma - More cellular pleomorphism& higher mitotic rate in leiomyosarcoma - Blunt ended cigar shaped nucei& cells are arranged in long fascicles intersecting at right angle in leiomyoma &leiomyosarcoma
• Solitary fibrous tumor	- Patternless proliferation of spindle cells with alternating hyper- & hypo-cellular areas - Dense keloid type of collagen - CD34 & CD99 immunopositivity present in solitary fibrous tumor & absent in myofibroma
• Desmoplastic fibroma	- Infiltrative & destructive pattern - Absence of hemengiopericytoma like vascular pattern - Monomorphic growth pattern
• Fibrosarcoma	- "Herring bone pattern" - Nuclear atypia - High mitotic rate & abnormal mitoses

▶ Dark stained areas – smaller, densely packed, round to spindle shaped myofibroblasts with intense eosinophillic cytoplasm

- hemengiopericytoma like vascular pattern

In myofibroma the cells are immunoreactive for vimentin and the smooth muscle actin, but negative or inconsistently positive for desmin or S-100 protein.Table 2 shows the histological differential diagnosis of myofibroma [9].Conservative treatment is the choice for 75% of patients [7].Local surgical excision with a margin of about 1 cm is the treatment of choice. In the present case, localized extent of the lesion allowed conservative surgical excision. Although myofibroma is a benign lesion but recurrences are still reported hence long term follow up is necessary. Chung and Enzinger [10] reported a 10% recurrence rate for the lesions they reviewed.

Conclusion

Myofibroma is a benign tumor with very good prognosis. A careful histological examination is mandatory to differentiate it from certain benign as well as malignant spindle cell neoplasms to arrive at correct diagnosis hence avoiding aggressive surgical procedures.

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