ISSN: 0975-833X

Available online at http://www.journalcra.com

International Journal of Current Research
Vol. 10, Issue, 04, pp.68294-68297, April, 2018

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

CASE REPORT

FIBROMYXOMA OF LEFT MANDIBLE: A CASE REPORT

Dr. Suresh Babu, P., Dr. George Skariah, Dr. Dain C Pearl, Dr. Naveen Kumar Jain and Dr. Sangeetha Shivaji

Department of Oral And Maxillofacial Surgery, Government Dental College Trivandrum, Kerala, India

ARTICLE INFO

Article History:
Received 14th January, 2018
Received in revised form
28th February, 2018
Accepted 20th March, 2018
Published online 30th April, 2018

Key words:
Fibromyxoma, Odontogenic myxoma, Odontogenic tumors, Mandibular Reconstruction

ABSTRACT

Fibromyxoma represents a rare benign neoplasm that mostly affects the posterior region of the mandible. It represents 0.5 to 17.7 % of odontogenic tumors: the third most frequently occurring type. This case report represents a patient with fibromyxoma of left mandible. A 42 year old female was referred for the evaluation and management of non-tender swelling on left face region that had gradually increased in size over 4 months duration. The examination revealed a palpable firm mass. Mild blanch mucosa with no ulceration was observed over swelling. Preoperative radiographs and computed tomography shows well defined multilocular expansile lesion. Surgical segmental resection of mandible under general anesthesia was performed via the extra oral approach followed by reconstruction with Titanium Recon plate of 2.5 mm. A histopathological examination confirmed the diagnosis of fibromyxoma. In conclusion, the radiological examination by means of CT and MRI plays an important role in the diagnosis of a fibromyxoma and in the differential diagnosis from other pathological entities such as the ameloblastoma. Patients must be monitored for at least two years postoperatively in order to diagnose possible recurrence.

Citation: Dr. Suresh Babu, P., Dr. George Skariah, Dr. Dain C Pearl, Dr. Naveen Kumar Jain and Dr. Sangeetha Shivaji, 2018. "Fibromyxoma of left mandible: A case report", *International Journal of Current Research*, 10, (04), 68294-68297.

INTRODUCTION

Myxomas of odontogenic origin were first described by Thoma and Goldman (1947), on the basis of site of occurrence, age at occurrence, association with missing teeth, and histopathological examination, which showed structural resemblance with dental mesenchyme and the sporadic presence of islands of odontogenic epithelium (Thoma and Goldman, 1947). Odontogenic fibromyxoma represents a rare slow-growing benign neoplasm, usually occurring in the 2nd and 3rd decades of life, rarely in children or adults over 50 years of age (Lo Muzio *et al*., 1996). The WHO classifies odontogenic myxoma under "Odontogenic ectomesenchyme with or without included odontogenic epithelium" (1992) (Thomas *et al*., 2011). Odontogenic myxoma is usually asymptomatic and is found incidentally on radiographs, appearing as a "soap bubble". The lesions are not encapsulated, allowing substantial infiltration into the adjacent medullary bone. Consequently, odontogenic myxoma is generally managed surgically; reports of surgical treatment of odontogenic myxoma vary from simple enucleation and curettage to segmental resection and hemimandibulectomy. Recurrence rates are reportedly high, at around 25%, especially when a more conservative approach is taken (Rocha *et al*., 2009). There are currently no clear evidence based surgical management guidelines for odontogenic myxoma. In this report, we presented a patient with fibromyxoma of left mandible. Diagnosis was confirmed through clinical examination, preoperative radiograph and CT scan, and surgical resection was conducted.

Corresponding author: **Dr. Naveen Kumar Jain,**
Department of Oral And Maxillofacial Surgery, Government Dental College Trivandrum, Kerala, India.

Case Report

A 42-year-old female was reported to the department of oral & maxillofacial surgery, Government Dental College, Thiruvananthapuram, with complaint of painless, gradually progressive firm swelling on left side of the face of 4 months duration. The medical anamnesis of the patient did not reveal anything in relation to the pathological condition. Clinical examination revealed a diffuse swelling, measuring about 3 × 5 cm with no clear borders, over the left body region of the mandible. Skin over the swelling was normal with no local rise of temperature, ulceration, or redness (Fig. 1). Intraoral examination shows swelling extending from left first premolar to second molar region and mild blanching of overlying mucosa with no sinus/ulceration on left posterior region (Fig. 2). On palpation, the swelling was diffuse, non-tender, firm and bony hard to touch. Expansion of the buccal cortical plate and mild lingual cortical plate was palpable near the left body

of the mandible and grade I mobility of tooth #33-36. Level I B lymph node of left side was movable and tender on palpation. Orthopantomography (OPG) showed a well-defined, multilocular, radiolucent lesion extending from distal aspect of 31 to towards the angle of mandible and from upper alveolus to lower border, resorption of root seen with 33-37 with thinning of superior and inferior border of mandible (Fig. 3). Occlusal view shows multilocular expansile lesion with honey comb pattern noticed at same site (Fig. 4). Lateral oblique view shows well defined multilocular radiolucency with soap bubble appearance (Fig. 5). CT revealed well defined expansile lytic lesion involving the body and alveolar process of mandible on the left side measuring approximately 2.3 × 5.3 cm's in size with areas of soft tissue density within; lesion causes resorption of adjacent tooth, marked cortical thinning and cortical defects (Fig. 6 A, B, C, D). Needle aspiration and incisional biopsy was then performed; aspiration showing yellowish thick jelly like material suggestive of mucinous material and histopathology revealed scattered spindle shape cells in mucoid stroma, cellular and fibrous area are admixed with bony trabeculae with osteoblastic rimming confirming the diagnosis of fibromyxoma of mandible (Fig. 7). Following which, wide excision of the lesion with segmental resection and reconstruction with Titanium Recon plate of 2.5 mm was done. A submandibular incision passing 2 cm below the lower border of the left mandible with subplatysmal dissection was given to expose the lesion extraorally. Lesion was excised along with the body and angle of the mandible. Reconstruction plate was adapted and screwed to the ramus distally and to the parasymphyseal mandibular region mesially. Patient has been on periodic follow-up (Fig. 8 A, B, C, D).

Fig. 1. Extraoral Examination

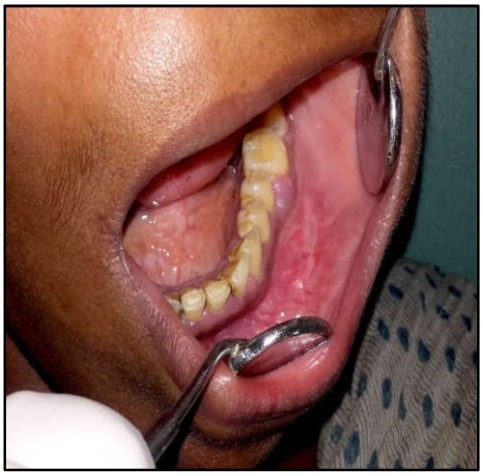

Fig. 2. Intraoral

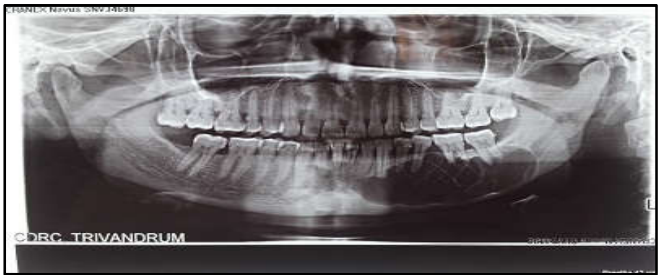

Fig. 3. Orthopantogram

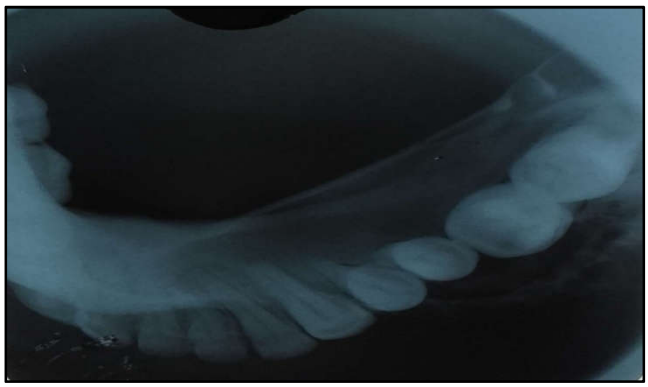

Fig. 4. Occlusal view mandible

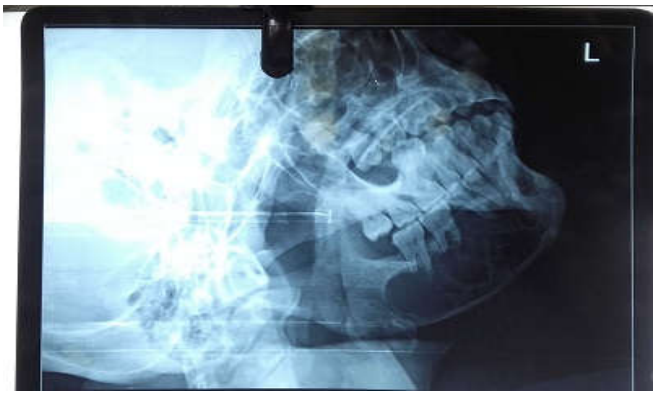

Fig. 5. Lateral oblique view mandible

DISCUSSION

Fibromyxoma is classified as a specific type of myxoma with a higher fibrous/myxoid tissue ratio than myxoma. According to Dutz and Stout, the term myxoma was first used by Virchow in 1863, but the term fibromyxoma was described by Marcove *et al*. in 1964 who reported extragnathic locations of fibromyxoma (Dietrich *et al*., 2011). Odontogenic myxoma is generally regarded as a rare benign tumors of mesenchymal origin that occurs in tooth-bearing areas of the mandible and maxilla, and is characterized by its slow growth and bony invasions, resulting in painless facial deformity (Rotenberg *et al*., 2004). Myxomas of the head and neck region occur mainly in the jaw bones, with a very small minority occurring in the pharynx, larynx, paranasal sinuses, and other soft tissues (Moore *et al*., 2008). Traditionally, the myxoma of the maxilla and mandible has been considered to be a neoplasm of odontogenic origin. Although the evidence is mainly circumstantial, support of an odontogenic origin has been perpetuated by its almost exclusive occurrence in the tooth-bearing areas of the jaws, its common association with an unerupted tooth or a developmentally absent tooth, its frequent occurrence in young individuals, its histologic resemblance to dental mesenchyme, especially the dental papilla and the

occasional presence of sparse amounts of odontogenic epithelium (Ashoka *et al*., 2016). The usual age of occurrence of odontogenic myxoma is between 10 and 40 years of age, with a peak incidence in the third decade of life. Simon *et al*. report a marked bias toward female sex, with a male: female ratio of 1:2, whereas other researchers have reported ratios ranging from 1:1.5 to 1:4. Shafer *et al*. stated that there is no sex predilection, whereas Zhang *et al*. reported a slight predominance in males (Shivashankara *et al*., 2017). Myxomas/fibromyxomas are usually located intraorally most often in the posterior regions of the mandible, its angle and ramus and rarely extraorally. The maxilla and anterior region of the mandible are rarely affected. In the present case, the lesion was on left side in the body of the mandible which is the most usual location of odontogenic myxoma consistent with the report of Li *et al* (2006), Speight (2013) (Shivashankara *et al*., 2017). Odontogenic myxoma is usually a slow-growing mass with late-appearing symptoms, primarily due to the mass effect. Symptoms include pain, paresthesia, ulceration, and tooth mobility, (Gonzalez-Garcia *et al*., 2006) we found mobility of the tooth in the present case. Although benign, odontogenic myxoma is invasive into surrounding normal bone, sometimes breaking through its boundaries[11]. This invasiveness has been attributed to the expression of matrix metalloproteinases 2 and 9, which degrade the extracellular matrix (ECM). These enzymes purportedly cause tumor cells to penetrate the bony trabeculae by acting on the ECM, thus aiding tumor growth (Miyagi *et al*., 2008). Immunohistochemistry and ultrastructural studies suggest that the cells of odontogenic myxoma have a myofibroblastic origin (Shivashankara *et al*., 2017). On gross examination, odontogenic myxoma appears as a mucous or gelatinous grayish-white tissue that replaces the spongy bone and displaces the cortical plates of the jaws. Root displacement and resorption may be present. It may refer to hard and also to soft tissues (Dietrich *et al*., 2011). In present case root resorption was present with the involved teeth. The tumor may have a minimal true capsule or may be uncapsulated and poorly demarcated from surrounding tissues (Shivashankara *et al*., 2017). In the present case, there was no capsule. Histologically, the myxoma is composed of loosely arranged spindle, rounded and stellate-shaped cells with a lightly eosinophilic cytoplasm in a mucoid rich (myxoid) intercellular matrix. The myxoid matrix is rich in hyaluronic acid and chondroitin sulphate (Thomas *et al*., 2011). In our case, the radiological appearance was of a well-defined, multilocular, radiolucent lesion with tennis-racquet appearance on OPG, honey-comb pattern on occlusal view and soap bubble appearance on lateral oblique. The radiological appearance of odontogenic myxoma has been variably described as always radiolucent, usually radiolucent, or mixed radiolucent-radiopaque (Altug *et al*., 2011). Zhang *et al*. classified the radiological appearance of odontogenic myxoma into six groups (Zhang *et al*., 2007). The appearance is variably described as mottled, soap-bubble, tennis racquet, or honeycombed. The honeycomb appearance seems to be restricted to mandibular lesions (Shivashankara *et al*., 2017). On computed tomography (CT) images, the low density area may correspond to the area with the abundant mucoid component. The solid portion of the tumor is reported to be rich in collagen fiber and may correspond to the enhanced area seen on CT images (Koseki *et al*., 2003). Differential diagnoses suggested based on radiological appearance of odontogenic myxoma include ameloblastoma, intraosseous hemangioma, aneurysmal bone cyst, glandular odontogenic cyst, central giant cell granuloma, cherubism, metastatic tumor, simple cysts, odontogenic keratocyst, and osteosarcoma (Chrcanovic *et al*., 2010). Initial treatment for odontogenic myxoma is surgery, but there appears to be some controversy about the best approach to take and evidence-based management guidelines have not been established. Conservative treatment was defined as enucleation, curettage, and marginal resection; radical treatments was defined as segmental or block resection, and hemimandibulectomy requiring reconstruction with the treatment protocol depending on the site and size of the tumor (Kawase-Koga *et al*., 2014). According to Chiodo *et al*, radiotherapy should not be considered as a standard therapy because these tumors are benign, occur in young patients, and are easily excised, avoiding the late risk of radiation induced tumors (Chido *et al*., 1997). Mandibular lesions can be managed primarily using a reconstruction plate followed by immediate or delayed vascularized fibular free flap, iliac crest graft, costochondral graft or scapular osteocutaneous free flap (Thomas *et al*., 2011). Our case was primarily managed with a segmental resection of mandible followed by titanium plate reconstruction. Odontogenic myxoma is notorious for a high recurrence rate of up to 25% after curettage. A minimum follow-up period of 5 years without recurrence is recommended by some researchers before performing reconstructive surgeries (Shivashankara *et al*., 2017). Bone and soft tissue reconstruction or osseointegrated implant should be reserved for those patients who remain free of the disease for 3 to 5 year after surgery or until the surgeon is confident that the patient is free of recurrence (Thomas *et al*., 2011).

REFERENCES

Altug, HA., Gulses, A. and Sencimen, M. 2011. Clinico-radiographic examination of odontogenic myxoma with displacement of unerupted upper third molar: review of the literature. *Int. J. Morphol.,* 29(3):930–3.

Ashoka, CA. 2016. Odontogenic Myxoma of Mandible: Report of a Rare Case with Review of Literature. IOSR *Journal of Dental and Medical Sciences,* 15(1):35-40.

Chido, AA., Strumas, N., Gilbert, RW. and Birt, BDB. 1997. Management of Odontogenic Myxoma of Maxilla. *Otolaryngol Head Neck Surg.,* 117(6):S73-76.

Chrcanovic, BR., do-Amaral, MB., de-Andrade, Marigo H. and Freire-Maia, B. 2010. An expanded odontogenic myxoma in maxilla. Stomatologija. 12(4):122–8.

Dietrich, EM., Papaemmanouil, S., Koloutsos, G., Antoniades, H. and Antoniades, K. 2011. Odontogenic Fibromyxoma of the Maxilla: A Case Report and Review of the Literature. Hindawi Publishing Corporation Case Reports in Medicine. Article ID 238712, doi:10.1155/2011/238712.

Gonzalez-Garcia, R., Rodriguez-Campo, FJ., Naval Gias, L., Munoz-Guerra, MF., Sastre-Perez, J. and Diaz-Gonzalez, FJ. 2006. Mandibular odontogenic myxoma. Reconstructive considerations by means of the vascularized fibular free flap. *Med. Oral Patol. Oral. Cir. Bucal*., 11: E531–E535.

Kawase-Koga, Y., Saijo, H., Hoshi, K., Takato T and Mori Y. Surgical management of odontogenic myxoma: a case report and review of the literature. *BMC Research.,* Notes 2014, 7:214.

Koseki, T., Kobayashi, K., Hashimoto, K. *et al*., 2003. Computed Tomography of Odontogenic Myxoma. *Dentomaxollofacial Radiology,* 32(3):160-5.

Lo Muzio, L., Nocini, P., Favia, G., Procaccini, M. and Mignogna, MD. 1996. Odontogenic myxoma of the jaws: a clinical, radiologic, immunohistochemical, and ultrastructural study. Oral Surgery, Oral Medicine, Oral Pathology, *Oral Radiology and Endodontics,* 82(4):426–433.

Miyagi, SP., Hiraki, KR., Martins, MD. and Marques, MM. 2008. Expression of matrix metalloproteinases 2 and 9 in odontogenic myxoma in vivo and in vitro. *J. Oral Sci*., 50(2):187–192.

Moore, BA., Wine, T., Burkey, BB., Amedee, RG. and Butcher, II RB. 2008. Sphenoid sinus myxoma: case report and literature review. *Ochsner J.,* 8:166–171.

Rocha, AC., Gaujac, C., Ceccheti, MM., Amato-Filho, G. and Machado, GG. 2009. Treatment of recurrent mandibular myxoma by curettage and cryotherapy after thirty years. *Clinics,* 64(2):149–2.

Rotenberg, BW., Daniel, SJ., Nish, IA., Ngan, BY. and Forte, V. 2004. Myxomatous lesions of the maxilla in children: a case series and review of management. *Int J Pediatr Otorhinolaryngol,* 68(10):1251–6.

Shivashankara, C., Nidoni, M., Patil, S. and Shashikala, KT. 2017. Odontogenic myxoma: A review with report of an uncommon case with recurrence in the mandible of a teenage male. *The Saudi Dental Journal,* 29(3):93-101.

Thoma, KH. and Goldman, HM. 1947. Central myxoma of the jaw. *Am. J. Orthod Oral Surg.,* 33(7):B532–40.

Thomas, PS., Babu, S., Mishra, C., Anusha, RL. and Shetty, S. 2011. Odontogenic Myxoma: Report of Two Cases with Review of Literature. *Journal of Indian Academy of Oral Medicine and Radiology*, 23(2):143-6.

Zhang, J., Wang, H., He, X., Niu, Y. and Li, X. 2007. Radiographic examination of 41 cases of odontogenic myxomas on the basis of conventional radiographs. *Dentomaxillofacial Radiol,* 36(3):160–7.

****