ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)
IJDSIR : Dental Publication Service
Available Online at:www.ijdsir.com
Volume – 4, Issue – 5, September - 2021, Page No. : 264 - 270

Surgical Management of Odontogenic Myxoma - A Case Report and Review of Literature

[1]Dr.Beena Roopak, Professor, Department of Oral and Maxillofacial Surgery, Rajarajeshwari Dental College and Hospital, Mysore Road, Bengaluru.

[2]Dr.Madhumati Singh, Professor and HOD, Department of Oral and Maxillofacial Surgery, Rajarajeshwari Dental College and Hospital, Mysore Road, Bengaluru.

[3]Dr.Anjan Kumar Shah, Professor, Department of Oral and Maxillofacial Surgery, Rajarajeshwari Dental College and Hospital Mysore Road, Bengaluru.

[4]Dr Sendhil Kumar M, Post Graduate Student, Rajarajeswari Dental College and Hospital, Mysore Road Bengaluru, India

Corresponding Author: Dr Sendhil Kumar M, Post Graduate Student, Rajarajeswari Dental College and Hospital, Mysore Road Bengaluru, India

Citation of this Article: Dr. Beena Roopak, Dr. Madhumati Singh, Dr. Anjan Kumar Shah, Dr Sendhil Kumar M, "Surgical Management of Odontogenic Myxoma - A Case Report and Review of Literature", IJDSIR- September - 2021, Vol. – 4, Issue - 5, P. No. 264 – 270.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Odontogenic myxoma (OM) is known to be an uncommon, benign, locally invasive, non-metastasizing neoplasm arising from the odontogenic ecto mesenchyme that usually affects in the tooth bearing areas of the jaws.Odontogenic myxoma commonly occurs in age groups ranging from 20 to 40 years with a female predilection, and the common site of occurrence is the mandible. The purpose of this article is to describe the unique one step surgical approach in which tumour resection and zygomatic implant insertion in a 21yr old male patient with odontogenic myxoma in the left maxillary region.

Keywords: Ectomesenchyme, Myxoma, Neoplasm, Maxillectomy, zygomatic implants.

Introduction

World Health Organization classifies odontogenic myxomas (OMs) as non-metastasizing, locally invasive benign tumours occurring both in maxilla and the mandible. They are relatively rare and represent around 3% of all the tumours of odontogenic origin and are frequently seen in the tooth bearing areas of the jaws. Their occurrence varies however; they are most frequently seen in second and third decades of life and are more common in females as compared to males. [4] Several studies have been performed in order to define its precise nature, but at present there is no universally accepted theory about its probable histogenesis. Based on the presence of occasional small islands of odontogenic epithelium, its occurrence almost exclusively in the jaw

bones and the histomorphological similarity to the mesenchymal component of the developing tooth, an odontogenic origin has been proposed and particularly from the dental follicle or the periodontal ligament. Some tumours may exhibit rapid growth. [5]Cortical expansion and perforation are common findings, however maxillary lesions tend to obliterate the maxillary sinuses as an early feature. 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 there are currently no clear evidence based surgical management guidelines for odontogenic myxoma. [3]

Case Report

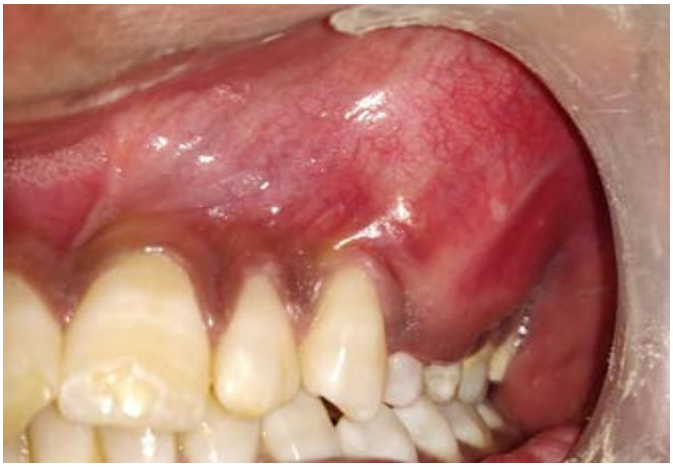

Figure 1: Intraoral appearance of the swelling.

Investigations

Orthopantomogram of the jaws revealed mixed radiolucency and radio opacity associated with the left maxillary sinus [Figure 2]. Computed tomography of the region showed a hypo dense lesion with irregular zathinned out margins extending from the maxillary left alveolus to the infra orbital rim obliterating the entire maxillary sinus. Destruction of the medial and lateral margins of the maxillary antrum along with partial occlusion of the nasal passage was also evident. Erosion of the medial and anterio-lateral wall of the maxillary sinus was evident. [Figure 3] In the 3D reconstruction of the CT images thinning of the palatal was noted. Blood investigations showed serum calcium, phosphorus, magnesium and alkaline phosphatase within normal limits. There by a clinical differential diagnosis of AOT, ossifying fibroma, fibrous dysplasia and peripheral giant cell granuloma was made. An incisional biopsy was performed, which showed spindle shaped cells in a loose myxomatous background in the histopathological analysis suggestive of odontogenic myxoma.

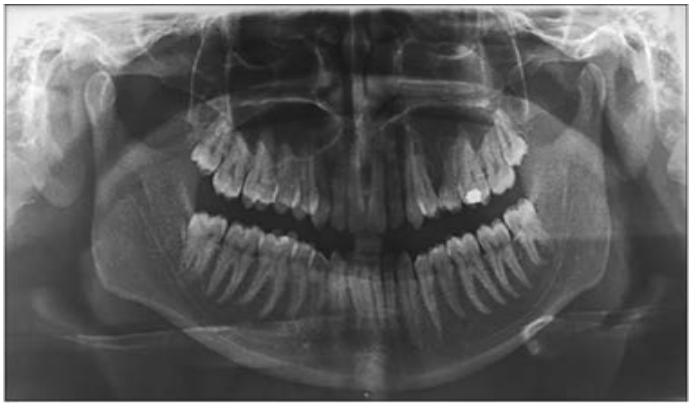

Figure 2: Orthopentamograph showing mixed radiolucency in relation to 24 & 25.

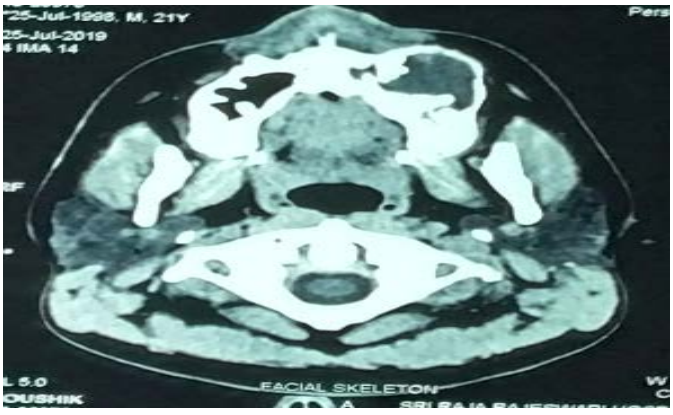

Figure 3: Computed Tomography images showing hypo dense lesion occluding the left maxillary sinus.

Under general anaesthesia the tumour was exposed through intra oral paramedian midface degloving incision extending from mesial aspect of 14 to distal aspect of 27 region region .[Figure 4] Osteotomy was performed around the encapsulated mass with a 1centimeter clearance sparing the 21 and 27 after the exposure the

tumour.[Figure 5] Left maxillectomy was done [Figure 6] and haemostasis achieved. Simultaneously a zygomatic implant of size 50mm implant was placed. The wound was closed and a prefabricated palatal plate with acrylic teeth was adapted to the operated site [Figure 7]. Postoperatively the healing was uneventful and under regular follow ups of every 3 months [Figure 8] till date where no recurrence has been noted. Further implant supported permanent prosthesis was fabricated.

Figure 4: Intraoral paramedian midface degloving incision

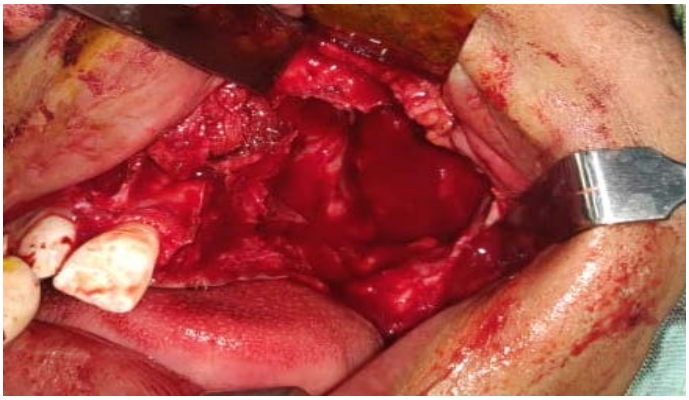

Figure 5: Partial maxillectomy sparing 21 and 27

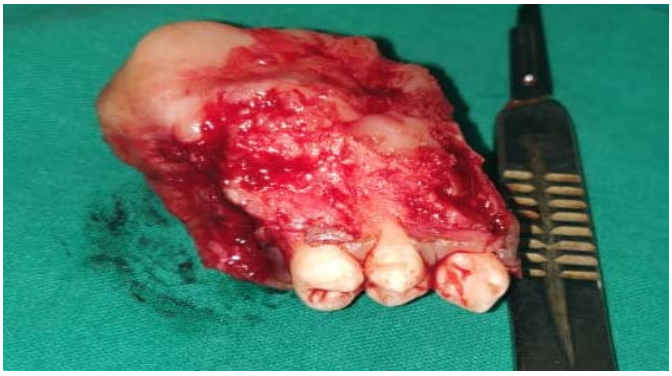

Figure 6 : Resected tumour.

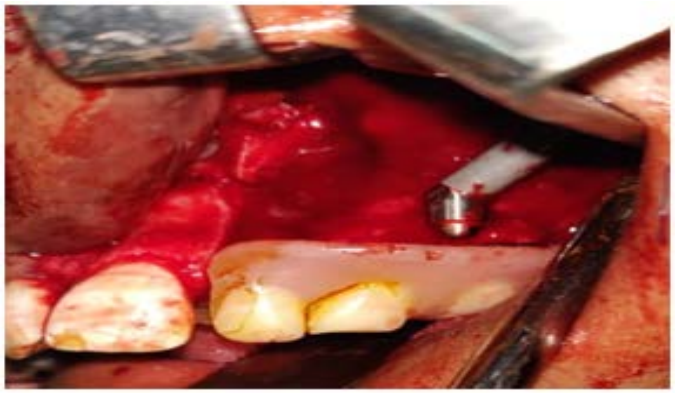

Figure 7 : Zygomatic implant and temporary acrylic plate with acrylic teeth in place.

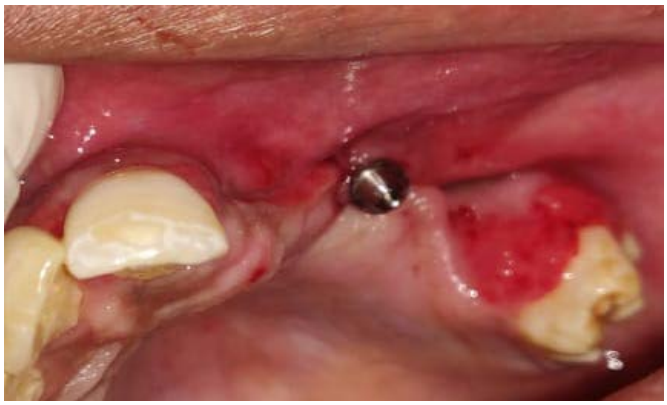

Figure 8 : Postoperative healing

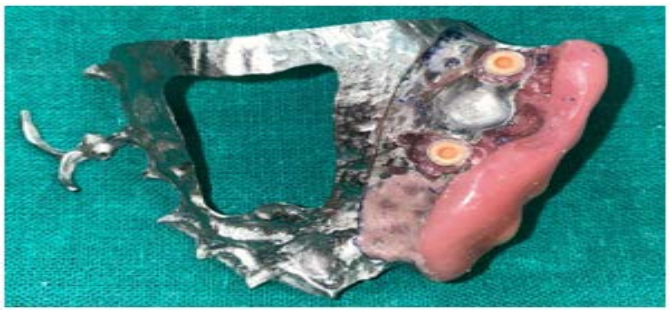

Figure 9: Implant supported Cast partial denture

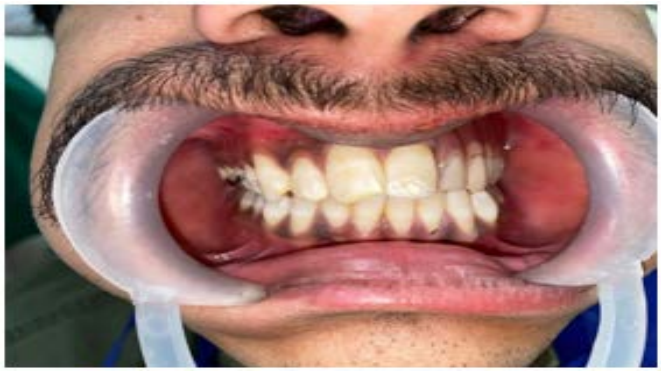

Figure 10: Final prosthesis in place

Discussion

Thoma and Goldman first described odontogenic myxoma of the jaw in 1947. Odontogenic myxoma is generally regarded as a rare benign tumour 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. [3]

This neoplasm occurs almost exclusively in the jaw bones and comprises 0.2% to 17.7% of odontogenic tumors. It may present at any age but is most frequently discovered in the 2nd to 4th decades. It occurs more frequently in the mandible than in the maxilla with ratio of 3: 1. The male to female ratio is 1:1.5. The neoplasm probably arises from the primitive mesenchymal structures of a developing tooth including the dental follicle, dental papilla, or periodontal ligament, and is therefore classified as an odontogenic tumor. Displacement of teeth by the tumor was a relatively common finding, but root resorption is rare. Differential diagnosis includes lesions showing typical multilocular radiolucency such as ameloblastoma, central haemangioma, aneurysmal bone cyst, central giant cell granuloma, giant cell lesions of hyperparathyroidism, cherubism, and metastatic tumors in the jaws. [6]

Previous theories stress that the lesion derives from the neural sheath or is the result of degeneration of fibromas, lipomas and so forth, due to the chronic irritation and the degenerative processes following tissue anoxemia . Recent studies advocate that myxomas/fibromyxomas arise from the mesenchymatous tissue of the dental follicle, thus being described as odontogenic with fibroblasts playing the major role in cell dispersal. [2]

On histopathology, the tumor consists of rounded, spindled, and stellate cells arranged in a loose, myxoid stroma with few collagen fibrils and delicate fibrous connective tissue as seen in our case. Immunohistochemistry studies suggest that the spindle-shaped cells constituting this neoplasm have a combined fibroblastic and smooth muscle typing, suggesting that it is of myofibroblastic origin. Odontogenic myxoma especially maxillary ones show little encapsulation and often extends into the surrounding soft tissue and intratrabecular space beyond the bony border, so that complete resection is difficult and recurrence is common.[6]

Radiographically, OMs appear as unilocular or multilocular radiolucency, sometimes showing a fine soap bubble or honey comb appearance occasionally with fine trabeculations. Root resorption is rarely seen although displacement of teeth is relatively common. Further, it frequently displays aggressive infiltration of the adjacent tissues as well as tendency to re-occur after surgical removal. [9]

Recommended therapy varies from curettage to radical excision. Curettage is associated with a high recurrence rate. Complete surgical removal can be difficult as the lesion is not encapsulated and because the myxomatous tissue infiltrates adjacent bone tissue. The treatment of choice is surgical excision ranging from segmental resection with clear bony margins of up 1.5 cm to prevent recurrence. Reconstruction can be immediate or delayed, and can include an autologous bone graft from the anterior or posterior iliac crest. Fibula free vascular osteocutaneous bone graft and distraction osteogenesis are other reconstructive modalities. Immediate postoperative follow up is weekly for approximately one month, then monthly for the next five months and twice a year for the next five years .[6]

Resection of malignant or benign tumors of the jaw causes emotional, functional, and aesthetic complications if not diagnosed and treated properly. Rehabilitation consists of different options for bone grafting, type and

origin of the graft, and the use or not of fixed prostheses and implants. [1]

The maxilla skeleton profile has both aesthetic and functional roles. Maxilla defects can therefore cause devastating functional and outcomes. The complicated three dimensional contouring of the maxilla makes autologous tissue transfer difficult; therefore, the use of a prosthesis provides an alternative for maxillary defect rehabilitation. However, most approaches use prostheses that are retained and supported by remaining natural teeth. This strategy increases the risk for various issues, including poor retention; poor mastication; abrasive ulcerations caused by dentures,overloaded abutment teeth, leakage of saliva, liquid, or food; and problems with speech . With the introduction of the zygomatic implants prosthesis retention has greatly improved, which should translate into better stability overall [8]. Zygomatic implantology (ZI) has been first mentioned by Aparicio et al. in 1993 then proposed by Brånemark in order to overcome bone availability after maxillectomy. Commonly, this option is offered as delayed procedure after tumour resection. Later, ZI has been employed in non-neoplastic, severely atrophic maxilla. [14]

Implant-supported prosthesis (ISP) seems to be the best alternative for these cases due to the unfavorable features of the grafted area, extreme mobile mucosa and lack of vestibule. Moreover, compared to removable prostheses, ISPs offer better masticatory capacity, patient comfort, and speech improvement, among others. [1]

. Our case and approach was compared with others reported in the literature, which was identified by a PubMed search using the term “odontogenic myxoma” of Maxilla , Articles without full text or with missing data were excluded the extent of tumour ,age /sex predilection ,surgical and rehabilitative strategies followed and recurrence on long term follow up were noted.

Table 1

Sn.	Author Details	Age	Sex	Region	`Treatment	Recurrence
1.	Haroon Rashid	22	M	Right Maxilla	Resection (Bulb Obturator)	No
2.	G. Siva Prasad Reddy , Surya Kumar	12	F	Left Mandible	Enucleation And Curettage	No
3.	YING LIU, BO HAN2	37	M	Maxilla	Right Radical And Left Partial Maxillectomy	No
4.	Abhishek Jaswalavik Kumar Jana	28	M	Left Maxilla	Resection (Removable Prosthesis	No
5.	Arul AJ, Verma S	25	F	Right Maxilla	Resection (Removable Prosthesis)	No
6.	Eva-Maria Dietrich, Styliani P	46	M	Right Maxilla	Enucleation	No

Conclusion

We have reported a rare case of maxillary Odontogenic myxoma in a 21-year-old male. These lesions are usually slow growing benign neoplasms. But OMs can turn out to be aggressive and locally invasive due to its unspecific nature, a sound knowledge of this lesion with proper

correlation of clinical, radiological and histopathological findings is a pre requisite to treat these patients appropriately. Resection with wide margins is the treatment of choice and follow-up during the first two years postoperatively is highly recommended as this is the period of highest reported recurrences planned single step procedures like what is done in this case will benefit the patient the most.

References

1. de Souza JG, Claus JD, Ouriques FD, Gil LF, Gil JN, Cardoso AC, Bianchini MA. Treatment of odontogenic myxoma: a multidisciplinary approach-6-year follow-up case. Case Rep Dent. 2014;2014:795808. doi: 10.1155/2014/795808.
2. Dietrich EM, Papaemmanouil S, Koloutsos G, Antoniades H, Antoniades K. Odontogenic fibromyxoma of the maxilla: a case report and review of the literature. Case Rep Med. 2011;2011:238712. doi: 10.1155/2011/238712. Epub 2011 May 2. PMID: 21629853; PMCID: PMC3099198.
3. Kawase-Koga Y, Saijo H, Hoshi K, Takato T, Mori Y. Surgical management of odontogenic myxoma: a case report and review of the literature. BMC Res Notes. 2014 Apr 5;7:214. doi: 10.1186/1756-0500-7-214. PMID: 24708884; PMCID: PMC4028019.
4. Rashid H, Bashir A. Surgical and prosthetic management of maxillary odontogenic myxoma. Eur J Dent 2015;9:277-83.
5. Miranda Rius J, Nadal A, Lahor E, Mtui B, Brunet L. Unusual presentation of localized gingival enlargement associated with a slow-growing odontogenic myxoma. Int J Oral Sci. 2013 Sep;5(3):172-5. doi: 10.1038/ijos.2013.27. Epub 2013 May 31. PMID: 23722914; PMCID: PMC3967326.
6. Jaswal A, Jana AK, Biswas M, Sikder B, Nandi TK. Recurrent odontogenic myxoma of maxilla: a diagnostic and operative dilemma. Indian J Otolaryngol Head Neck Surg. 2008 Mar;60(1):41-4. doi: 10.1007/s12070-008-0013-1. Epub 2008 Apr 3. PMID: 23120497; PMCID: PMC3450724.
7. Lahey E, Woo SB, Park HK. Odontogenic myxoma with diffuse calcifications: a case report and review of the literature. Head Neck Pathol. 2013 Mar;7(1):97-102. doi: 10.1007/s12105-012-0387-y. Epub 2012 Jul 27. PMID: 22837042; PMCID: PMC3597162.
8. Qu XZ, Wang MY, Ong HS, Zhang CP. Post-operative hemimaxillectomy rehabilitation using prostheses supported by zygoma implants and remaining natural teeth. Clinics (Sao Paulo). 2016 Oct 1;71(10):575-579. doi: 10.6061/clinics/2016(10)04. PMID: 27759845; PMCID: PMC5054771.
9. Arul AJ, Verma S, Arul AJ, Verma R. Infiltrative odontogenic myxoma of the posterior maxilla: Report of a case. J Nat ScBiol Med 2013;4:484-7
10. Reddy GS, Kumar BS, Muppa R, Regonda SK, Tvs HK. Odontogenic fibromyxoma of maxilla: a rare case report. Case Rep Dent. 2013;2013:345479. doi: 10.1155/2013/345479. Epub 2013 Mar 4. PMID: 23533825; PMCID: PMC3603617.
11. Guo YJ, Li G, Gao Y, Ma XC. An unusual odontogenic myxoma in mandible and submandibular region: a rare case report. DentomaxillofacRadiol. 2014;43(8):20140087. doi: 10.1259/dmfr.20140087. Epub 2014 Oct 1. PMID: 25270061; PMCID: PMC4240259.
12. Liu Y, Han B, Yu T, Li L. A large odontogenic myxoma of the bilateral maxillae: A case report. Oncol Lett. 2014 Sep;8(3):1328-1332. doi: 10.3892/ol.2014.2243. Epub 2014 Jun 12. PMID: 25120718; PMCID: PMC4114706.
13. Salvatori P, Mincione A, Rizzi L, Costantini F, Bianchi A, Grecchi E, Garagiola U, Grecchi F.

Maxillary resection for cancer, zygomatic implants insertion, and palatal repair as single-stage procedure: report of three cases. MaxillofacPlastReconstr Surg. 2017 May 25;39(1):13. doi: 10.1186/s40902-017-0112-6. PMID: 28603707; PMCID: PMC5443715.

14. Oliveira SV, Rocha AC, Ceccheti MM, Gallo CB, Alves FA. Odontogenic myxoma in a child treated with enucleation and curettage. Autops Case Rep [Internet]. 2018;8(3):e2018042. https://doi.org/10.4322/acr.2018.042
15. Ram H, Mehta G, Kumar M, Lone P. Odontogenic myxoma in a 52-year-old woman. BMJ Case Rep. 2014 May 23;2014:bcr2013202416. doi: 10.1136/bcr-2013-202416. PMID: 24859552; PMCID: PMC4039872.
16. Kapoor P, Gandhewar TM, Andrade NN, Desai RS. Peripheral myxoma of the infratemporal region: An unusual case report. J Oral Biol Craniofac Res. 2015 Sep-Dec;5(3):232-5. doi: 10.1016/j.jobcr.2015.07.004. Epub 2015 Aug 8. PMID: 26605148; PMCID: PMC4623249.
17. Raubenheimer EJ, Noffke CE. Peripheral odontogenic myxoma: a review of the literature and report of two cases. J Maxillofac Oral Surg. 2012 Mar;11(1):101-4. doi: 10.1007/s12663-011-0194-0. Epub 2011 Apr 19. PMID: 23449257; PMCID: PMC3319828.
18. Manne RK, Kumar VS, VenkataSarath P, Anumula L, Mundlapudi S, Tanikonda R. Odontogenic myxoma of the mandible. Case Rep Dent. 2012;2012:214704. doi: 10.1155/2012/214704. Epub 2012 Jul 9. PMID: 22830060; PMCID: PMC3399348.