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Case Report

A rare case of odontogenic myxoma of maxilla

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ABSTRACT

Odontogenic myxoma is a benign locally aggressive rare intraosseous neoplasm, originating from the odontogenic ectomesenchyme in second and third decades of life. Clinically they are slow growing, indolent, expansile, non-metastasizing involving predominantly mandible than maxilla. Maxillary odontogenic tumor spreads more aggressively through maxilla than mandible hence the need to address the tumor early and also the reconstruction of the maxillary defect following defect is uniquely challenging. This is a report of a rare case of left maxillary odontogenic myxoma which was locally aggressive and causing significant maxillary erosion in a 35 year old female, left subtotal maxillectomy done using Weber-Fergusson approach with Dieffenbach's modification and the defect repaired with temporalis muscle flap.

Keywords: Maxilla, Odontogenic myxoma, Tumor

INTRODUCTION

Odontogenic myxoma is a rare and intriguing type of jaw tumor that represents about 3% of all odontogenic tumors.[1] It's known for its unique clinical presentation, often causing progressive swelling and sometimes leading to ulceration of the overlying mucosa.

It predominantly affects the posterior mandible but can also occur in the incisive sector, upper maxilla, and mandibular condyle.[2,3] The premolar to molar region is the site of predilection in the maxilla. It rarely crosses the midline.

The growth of this tumor can be rapid, frequently resulting in the displacement of teeth. Its characteristic appearance on radiographs, often described as 'soap bubble' or 'honeycombed,' helps in differentiating it from other conditions such as ameloblastoma and hemangioma.[4] The average age of occurrence is around 30 years, and it has slight female preponderance than male.[4,5] Histologically, it is distinguished by stellate fibroblastic cells within a mucoid matrix, lacking a capsule and showing infiltrative growth, which can make it challenging to manage. Treatment often requires radical resection for larger tumors due to their aggressive nature and high recurrence rate, while smaller lesions may be managed more conservatively but still require careful evaluation due to the risk of recurrence.[6]

Odontogenic myxoma often cause significant destruction of the involved bone and tooth displacement. The size of the lesions correlated with their locularity with size of multilocular lesions exceeding unilocular lesions in greatest dimension.[7]

Treatment strategies for odontogenic myxoma are primarily surgical, ranging from conservative methods like enucleation to more radical approaches including resection. The choice of treatment depends on the size and location of the tumor, as well as the patient's age and general health. Despite the benign nature of the tumor, there is a notable risk of recurrence, especially if the tumor is not completely removed during surgery. This highlights the importance of early detection and appropriate surgical intervention in the management of odontogenic myxomas.

CASE REPORT

A 35 years old female presented with indolent swelling in the left cheek region for 2 years, insidious onset and progressively increased to attain the present size. There was no history of trauma. On Intraoral examination revealed a swelling in the left upper jaw with maligned upper premolar and molar tooth. Mucosa over the swelling normal. Extraoral examination showed an ill-defined swelling in the left middle third of face superiorly extending from infraorbital margin to left angle of mouth inferiorly, medio-laterally extending from left nasolabial crease to zygoma, nontender and hard in consistency. Skin surface over swelling appeared smooth, stretched shiny and free from the swelling on palpation and there was no cervical lymphadenopathy.

Contrast enhanced computerized tomography of head and neck was suggestive of expansile lytic lesion with soft tissue component involving the left upper alveolus eroding the floor of left maxillary sinus. Image guided Biopsy of lesion was suggestive of odontogenic myxoma.

Under general anesthesia, Weber-Fergusson incision with Dieffenbach's extension was made to expose the lesion in its entirety. Periosteum of anterior maxillary wall, zygoma and inferior orbital rim exposed. Anterior fibers of masseter divided and osteotomies done at zygomatico-temporal junction, inferior orbital rim and posterior wall of maxilla-pterygoid plate junction. Left subtotal maxillectomy done with temporalis muscle flap used to cover defect. Patient's postoperative recovery uneventful and patient had good cosmetic outcome with temporalis muscle flap provided contour to left maxillary region.

On macroscopic examination biopsy specimen showed a lobulated partly encapsulated myxoid glistening lesion measuring 6.5×5.8×4 cm involving entire maxilla. Cut section showed myxoid, firm to soft glistening and homogenous. Microscopic examination showed sheets of monotonous hypocellular areas having stellate shaped cells with elongated cytoplasmic process and small nuclei. Some of odontogenic epithelial rests in loose mesenchymal myxoid background visible, Suggestive of odontogenic myxoma. Patient is on close observation and is now disease free in 6th month follow up with no radiological signs of recurrence.

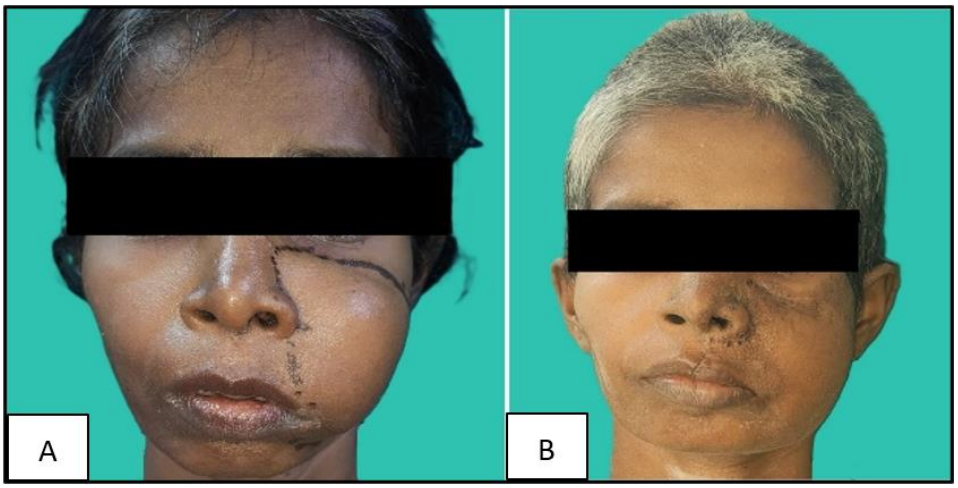

Figure 1 (A and B): Preop and postop clinical image.

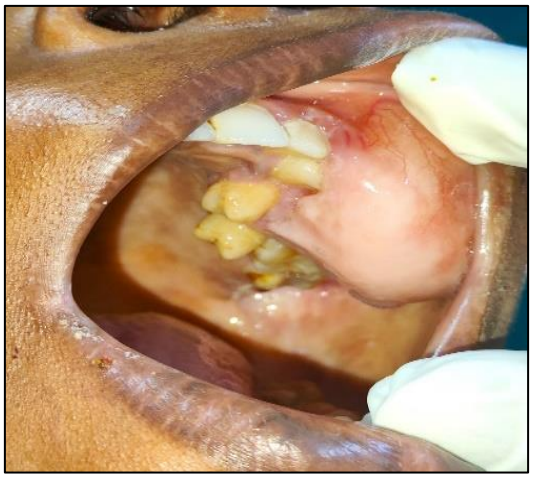

Figure 2: Intraoral examination of lesion.

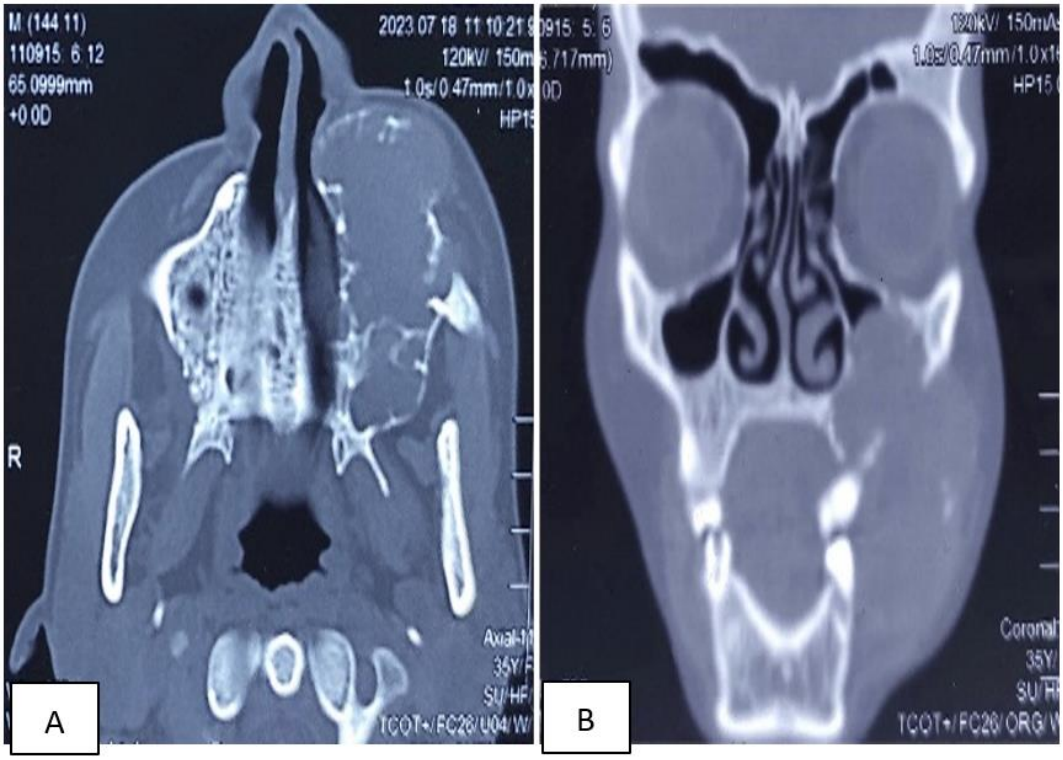

Figure 3 (A and B): Computerised tomography of paranasal sinus showing erosion of inferior wall of maxilla.

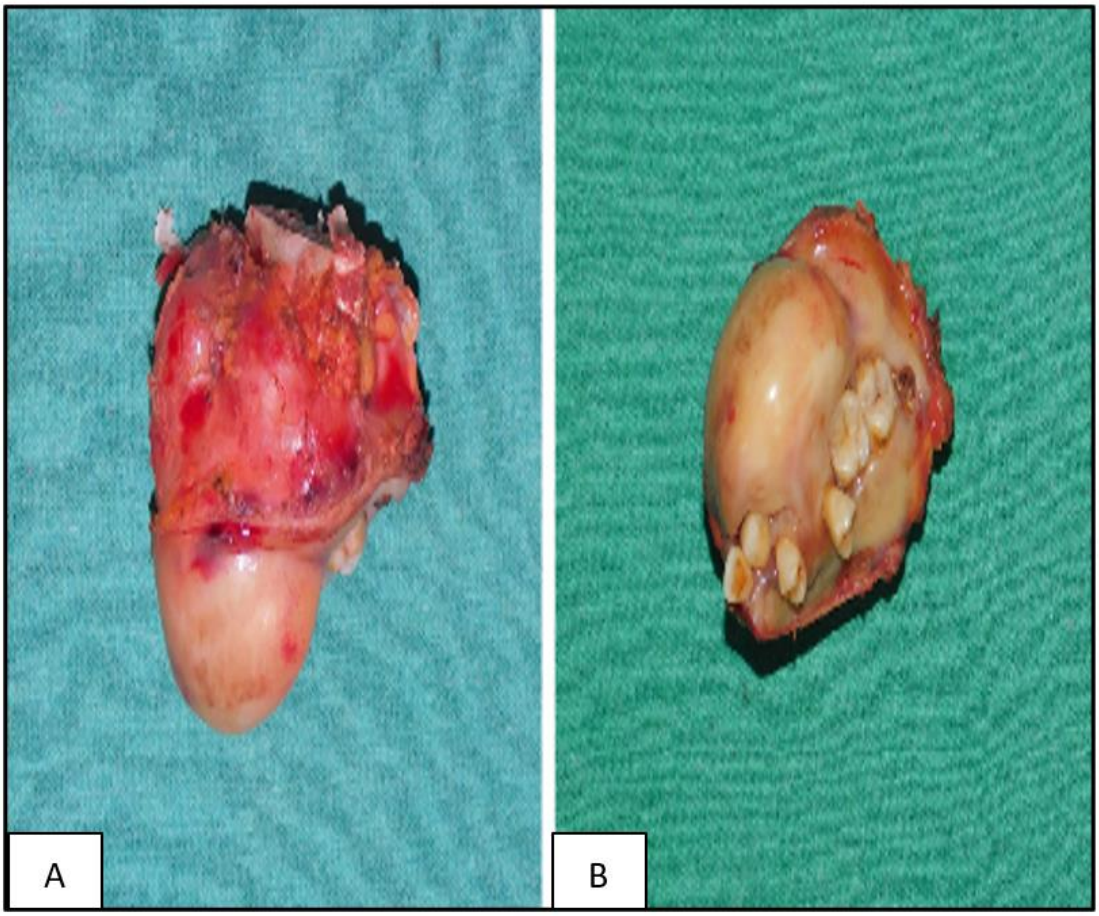

Figure 4 (A and B): Left subtotal maxillectomy specimen-anterior and inferior view.

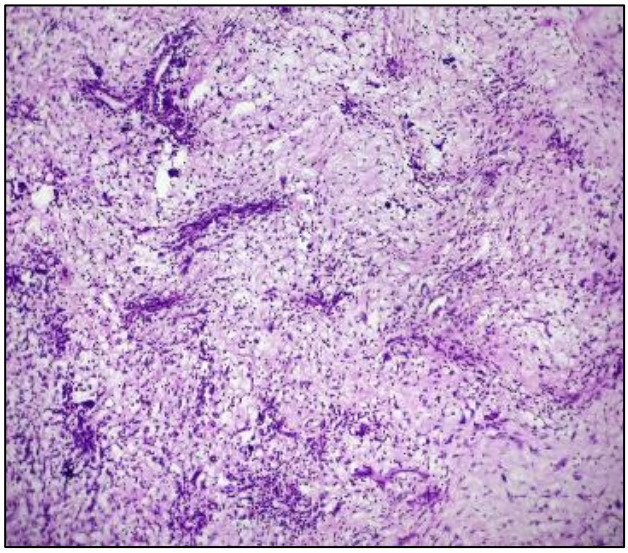

Figure 5: Hypocellular areas showing stellate cells in myxoid background.

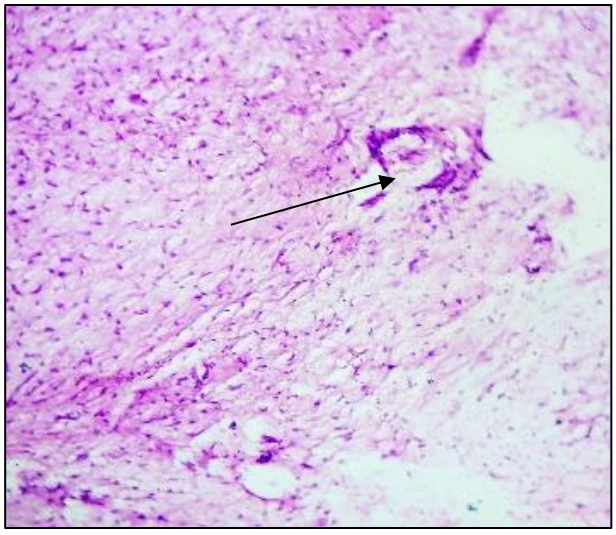

Figure 6: Odontogenic cell rests noted with stellate cells showing elongated cytoplasmic process and small nuclei.

DISCUSSION

Odontogenic myxoma is characterized by a painless, slow enlargement of the involved bone, which may cause tooth displacement or loosening as it grows. While it typically presents without symptoms, large lesions can be symptomatic due to bony erosion and invasion into soft tissues. This patient had large lesion in intraoral soft tissue extension with maxillary erosion and significant tooth displacement. This maxillary sinus involvement caused thinning of cortical bone with areas of destruction except the inferior wall of orbit.

Odontogenic myxoma has variable radiographic presentation and overlapping radiographic features with other benign and malignant neoplasms make radiological differentiation challenging.[8]

While giant cell lesions associated with hyperparathyroidism are more commonly found in the anterior mandible and may present with pain and swelling, multilocular myxomas typically occur in the posterior jaw or maxilla and exhibit a characteristic fine trabeculation within the lesion. Aneurysmal bone cysts, on the other hand, can be differentiated by their clinical presentation of tenderness and pain. These differentials were hence ruled out in this case and the possibility of ameloblastoma or odontogenic myxoma were considered and core biopsy from the intraoral soft tissue component revealed the diagnosis.

Despite being benign, the high recurrence rate is often attributed to the tumor's ability to infiltrate the cancellous bone, which may not be immediately apparent on radiographic imaging.[9] This insidious invasion, coupled with the lack of a clear encapsulation, makes complete surgical removal challenging. Since the patient had extensive involvement of maxilla, subtotal maxillectomy done adequate bone and mucosal margins and the ensuing large defect repaired with temporalis muscle flap.

CONCLUSION

Odontogenic myxoma of maxilla behaves more aggressively than that of the mandible with local spread occurring rapidly posing a significant challenge to the surgical management. In this case the timely diagnosis allowed en bloc removal of tumor with the defect being large needed temporalis muscle flap for reconstruction. The patient is now disease free 6 months after surgery with good cosmetic outcome.

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Ethical approval: Not required

REFERENCES

1. Plastic Surgery, Vol. 5: Tumors of the Head, Neck, and Skin by Stephen J. Mathes. 2005.
2. Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. Illinois: Quintessence Publishing Co Ltd. 2004.
3. Lu Y, Xuan M, Takata T, Wang C, He Z, Zhou Z, et al. Odontogenic tumors: A demographic study of 759 cases in a Chinese population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998;86:707-14.
4. Watkinson J, Gilbert RW. Stell and Maran's Textbook of Head and Neck Surgery and Oncology, Fifth edition, Hodder Arnold publications. 2012;248-9.
5. Gunhan O, Erseven G, Ruacan S, Celasun B, Aydintug Y, Ergun E, et al. Odontogenic tumours: A series of 409 cases. Aust Dent J. 1990;35(6):518-22.

6. Zimmerman DC, Dahlin DC. Myxomatous tumors of the jaws. Oral Surg Oral Med Oral Pathol. 1958;11(10):1069-80.
7. Kaffe I, Naor H, Buchner A. Clinical and radiological features of odontogenic myxoma of the jaws. Dentomaxillofac Radiol. 1997;26(5):299-303.
8. Noffke CE, Raubenheimer EJ, Chabikuli NJ, Bouckaert MM. Odontogenic myxoma: review of the literature and report of 30 cases from South Africa. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;104(1):101-9.
9. Pahl S, Henn W, Binger T, Stein U, Remberger K. Malignant odontogenic myxoma of the maxilla: Case with cytogenetic confirmation. J Laryngol Otol. 2000;114(7):533-5.

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