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Odontogenic Myxofibroma of Mandible and Submandibular Region

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ABSTRACT: Odontogenic myxomas are benign but locally aggressive, slow growing neoplasms found almost exclusively in the jaws and occasionally only arise in other bones. We present a rare case of odontogenic myxoma occurring in the mandible and submandibular region of a 38-year-old male patient with a brief review of clinical and radiological features, and diagnostic and operative dilemmas in managing the same.

KEYWORDS: Odontogenic myxoma, Myxofibroma

I. INTRODUCTION

Odontogenic myxoma (OM) is a benign, locally aggressive, slow-growing and non-metastasizing neoplasm of the jawbone.[1] According to the World Health Organization (WHO 2017), OM is classified as a benign tumour of Mesenchymal origin with or without odontogenic epithelium. Odontogenic myxoma is the third most common odontogenic tumour after ameloblastoma and odontomas.[2] Rudolph Virchow, the German scientist probably was the first to describe the histologic features of myxofibroma in 1863, although the lesions of jaws were not particularly mentioned. Thoma and Goldman 1947 first described myxomas of the jaws. Since then, odontogenic myxoma has been a subject of continuous scientific debate.[3]Myxomas of the head and neck region are rare. They account for 3–6% of all odontogenic tumours. Clinically occurs in individuals who are between their 2nd to 4th decades of life, have a slight predilection for females and are rarely found in children and the elderly.[4]

Radiographically, it appears as unilocular pericoronal radiolucency with variable trabecular pattern giving rise to a soap bubble, tennis racquet or honeycomb appearance. However, other radiological appearances like "Sun-ray" appearance have also been reported in the literature, root resorption is rarely seen although displacement of teeth is relatively common. It frequently shows aggressive infiltration of the adjacent tissues as well as the tendency to re- occur after excision.[5]

In this article, an unusual OM is presented. The myxoma located in the mandible and the submandibular region was characterized by large size, short duration and bone involvement.

II. CASE REPORT

A 38- year- old male patient presented to the Department of Oral and Maxillofacial Surgery, Regional Dental College and Hospital, with a chief complaint of painless swelling in his right body of the mandible and submandibular region for 6 months.

The patient presents with a history of extraoral swelling, which was initially small in size, and gradually increased to its present size. There was no history of pain, pus discharge, bleeding, and trauma. Medical history and family history were non-contributory. On general physical examination, the patient was moderately built and nourished.

On extraoral examination, solitary diffuse swelling was present on the right mandible and submandibular region measuring approximately 5×5cm in dimension, extending Superioinferiorly from the superior border of the body to the submandibular region at the level of the hyoid bone.The surface of the swelling appeared smooth and slightly glossy, the surrounding skin was stretched and no visible pulsation was seen [Figure 1]. On palpation, the swelling was firm to bony hard in consistency and non-tender. Right Submandibular

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lymph nodes are palpable, mobile, non-tender, and oval with approx. 1x1cm in size.

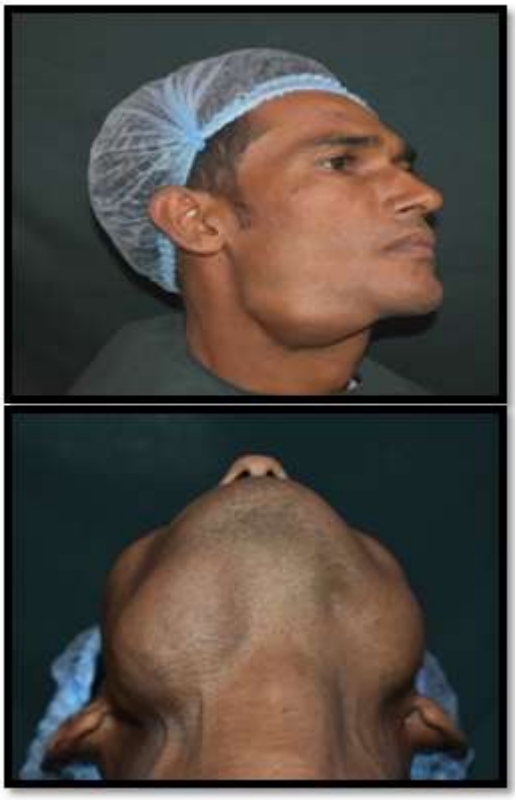

Figure 1: Diffuse swelling present on the right side of the face involving right submandibular region.

Intraorally, a well- defined swelling was present on the lower right back tooth region measuring about 2 × 3 cm, extending anteroposteriorly from the gingival margin of 45,46 to the buccal vestibule of 45,46, and mediolaterally from the distal aspect of 45 to mesial aspect of 47. Mucosa overlying the swelling appeared normal [Figure 2]. On palpation, the swelling was firm to hard in consistency, non-tender with a smooth, well- defined margin. Other findings include periodontal pocket present 45,46. Hard tissue examination revealed grade 1 mobility i.r.t 44,45 and 47 with tenderness on percussion, supragingival calc and generalized extrinsic stains.

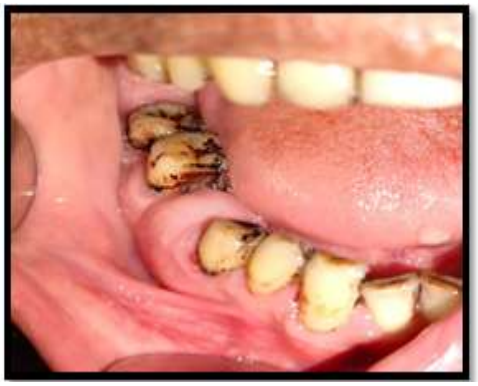

Figure 2: Well defined swelling was present on lower right back tooth region 45,46

Based on history and clinical examination, a provisional diagnosis of Ameloblastoma i.r.t right mandible along with the differential diagnosis of ameloblastoma, intraosseous hemangioma, giant cell granuloma, osseous aneurismatic cyst and metastasis Ameloblastic fibroma, Odontogenic myxoma.

Radiographical investigations of orthopantomography and CECT of the oral cavity and neck were done. Orthopantomograph revealed well- defined radiolucency involving from distal to 43 to the right ramus of the mandible extending to the inferior border of the mandible with fine trabeculation along with inter bony septa present. Also, resorption of the roots is present from 43 to 48 teeth. [Figure 3 (A) &(B)]

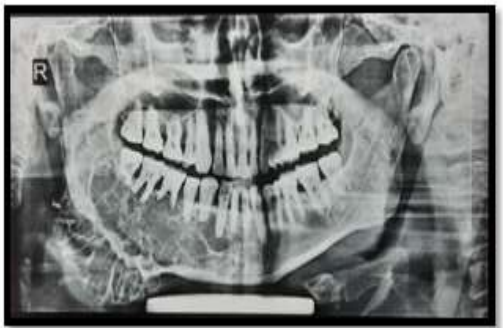

Figure 3(A): Orthopantomography revealed well-defined mixed radiolucency and radio-opacity involving from right mandible to the ramus region along with the extension to submandibular region.

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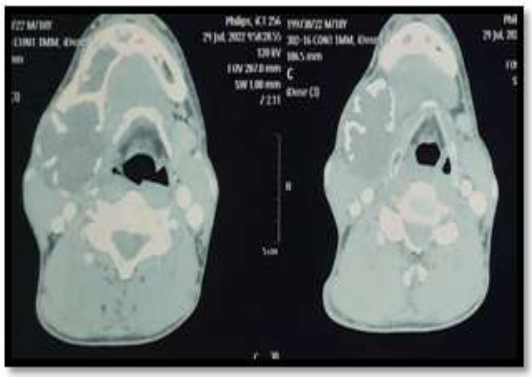

Figure 3(B): CECT revealed well‑defined mixed radiolucency and radio-opacity involving from right mandible to the ramus region.

CECT of the faciomaxillary region reveals a well-defined osteolytic lesion shown in the axial view with the destruction of the buccal and lingual cortices along with the expansion ofthe right hemimandible buccal and lingual cortices.

Based on the clinical diagnosis of Ameloblastoma, a biopsy was performed. The microscopic examination of the hematoxylin and eosin‑stained section showed lesional tissue fragments composed of fibro-collagenous stroma with hypocellular myxoid areas and scattered bands appearing as spindle-shaped cells. No frank pleomorphism or mitosis or necrosis was noted and the section was negative for granulomas or malignancy. No nuclear atypia or mitotic activity was seen and no Ameloblastic epithelium is identified Subsequently, the lesion was diagnosed as Odontogenic Myxoma and surgical resection followed by reconstruction was proposed. [Figure 4]

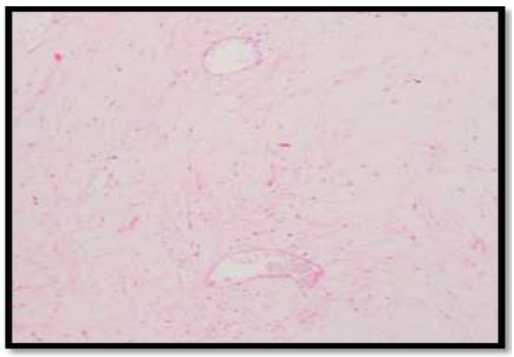

Figure 4: The microscopic examination of hematoxylin and eosin‑stained section showed lesional tissue fragments composed of fibrocollagenous stroma with hypocellular myxoid areas and scattered band appearing spindle‑shaped cells.

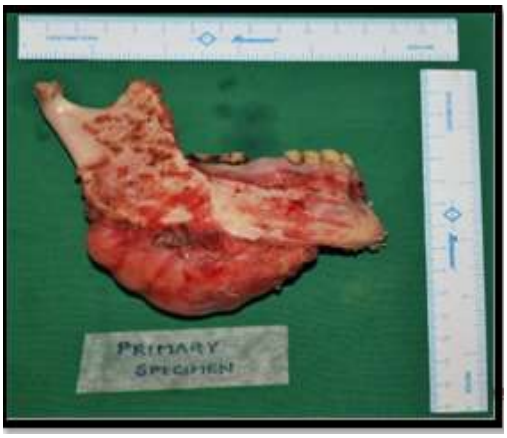

Figure 5: Excised specimen

III. DISCUSSION

We report a rare case of gnathic Odontogenic myxoma. We will review the clinical, radiographic, and histologic features. Odontogenic myxoma is a benign, aggressive neoplasm and it is important to distinguish it from less aggressive gnathic lesions and mesenchymal malignancies to ensure appropriate patient management.

Odontogenic myxoma is a benign, slowly proliferative and locally aggressive tumour. It usually occurs in the 2nd and 3rd decades of life, rarely in children or adults over 50 years of age.[6] Several studies show female predilection and the posterior mandible is more frequently affected than the maxilla.[7] Our patient was a 42 years old male presenting with an OM in the right mandible extending from the body to an angle along with the submandibular region. Unusual cases of rapidly expanding OM of the jaw have been reported. The rapid growth is believed to be due to the production of a mucoid ground substance by stellate tumour cells. The lesion is benign, but it is locally aggressive causing bone perforation, root resorption, tooth displacement, and mobility.[8]According to Reichert and Philipsen, mandibular myxomas accounted for 66.4%, with 33.6% in the maxilla. Whereas 73.8% and 65.1% of the cases were located in the molar and premolar areas in the maxilla and mandible respectively[9]. On gross examination of the specimen, the gelatinous, loose structure of the myxoma was obvious. Microscopically, the myxoma is made up of loosely arranged spindle-shaped and stellate cells, many of which have long fibrillar processes that tend to intermesh. The loose tissue is not highly cellular, and these cells do not show evidence of significant activity (pleomorphism, prominent nucleoli or mitotic figures). The intercellular substance is mucoid. The tumour is usually interspersed with a variable

number of tiny capillaries and occasionally strands of collagen.[10]

The tumour is not radiosensitive, and surgery is the treatment of choice. Treatment of Odontogenic myxoma varies from local excision, curettage, or enucleation to radical resection depending on tumour size. The aggressive nature of odontogenic myxoma is well documented in the literature.[4] While generally considered a slow-growing neoplasm, odontogenic myxoma may be infiltrative and aggressive, with high recurrence rates.In the present case, the tumour was completely removed by right hemi-mandibulectomy and no recurrence was reported even after 6 months of the surgery.

IV. CONCLUSION

There is a wide variety in clinical and radiological appearance of odontogenic myxomas, the most common form of presentation being an asymptomatic expansion in the jaw and a multilocular radiolucent image. Hence correlation of clinical, radiological and histopathological features is essential when trying to diagnose lesions which lack the characteristic appearance. Early detection and complete surgical excision of these lesions followed by long-term follow-up bear importance in clinical management due to their aggressive nature and high recurrence rate. The presented case showed no clinical or radiological evidence of recurrence after 6 months of post-operative follow-up. Since the time elapsed from surgery is still short, continued clinical and radiological monitoring is required.

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