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Odontogenic Myxoma - A Rare Case Report

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Abstract: ***Odontogenic myxoma (OM) is an intraosseous neoplasm that comprises 3–20% of all odontogenic tumors and is characterised by its slow growth and bony invasions, resulting in painless facial deformity. Incidence of such a tumour is approximately 0.05 new cases per million populations per year. The majority of cases are asymptomatic and diagnosed in a routine dental radiograph, but there are few patients reported to have increasing pain due to invasion of surrounding structures. Here is a case report of odontogenic myxoma in a 25 - year - old male patient who presented with pain and swelling for a period of 8 months.***

Keywords: odontogenic myxoma, Odontogenic tumour, case report

1. Introduction

Myxomas are rare tumors of the hard sclerous and soft tissues in the body. Odontogenic myxomas comprise only a small fraction of myxoma [1]. Odontogenic myxoma (OM) is an intraosseous neoplasm that may arise in the maxilla or mandible, which can be locally aggressive. It accounts for 3–6% of all odontogenic tumors [2]. It was first introduced in 1974 by Thoma and Goldman [3]. WHO defined the tumour as “a locally invasive neoplasm that consists of angular and rounded cells in mucoid background” [4]. OM appears to originate from dental papilla, follicle, or periodontal ligament. The evidence for its odontogenic origin arises from its almost exclusive location in the tooth bearing areas of the jaws, its occasional association with missing or unerupted teeth, and the presence of odontogenic epithelium [5]. According to the World Health Organization (WHO), OM is classified as benign tumour of ectomesenchymal origin with or without odontogenic epithelium [6]. It is an asymptomatic lesion that shows an infiltrative growth pattern. It is invasive locally and has a high recurrence rate accounting 1 - 19% of all odontogenic tumors [7] affecting mostly young patients in their second and third decade of life [4], has a slight female predilection, and involves the mandible more commonly than the maxilla. Clinically, it is a slow - growing, expansile, painless tumour, which may cause root resorption, tooth mobility, bone expansion, cortical destruction and facial deformity [8]

Radiographically, the classic presentation is that of a multilocular radiolucency, with well - developed locules, consisting of fine trabeculae, arranged at right angles, known as the ‘Tennis - racquet’ or ‘Step - ladder’ pattern which is diagnostic feature that is contributed only to this odontogenic myxomas. A ‘sun - ray’ or ‘sun - burst’ appearance has also been reported in the literature [9, 10] On gross examination, the surgical specimen is characteristically loose, slippery or gelatinous in nature. Histopathologically, the lesion consists of loosely arranged spindle, stellate - shaped or round cells, in an abundant myxoid stroma [8] Here in this case report, we describe a case of a 25 - year - old male who presented with a painless swelling and with a distinct radiographic feature of odontogenic myxoma in detail.

2. Case Report

A 25 years old male patient reported to the department with a chief complaint of swelling in the lower left back tooth region for past 8 months. Patient gives history of gradual increase in size of the swelling [figure 1] to the present size, and is associated with intermittent throbbing pain which aggravates during mastication and relieves on analgesic medication. His past medical and dental history was non - contributory. His intraoral examination revealed clinically missing 38 with evidence of a single diffuse swelling in the left buccal vestibule in relation to 37, 38 regions, measuring approximately 3 x 1.5 cm, extending anteroposteriorly from mesial aspect of 37, to 1cm away from the pterygomandibular raphae; and mesiodistally, from the marginal gingival level of 37 up to 1 cm distal to it diffusing into the buccal mucosa, with presence of obliteration of buccal vestibule. The colour of the mucosa over the swelling appears erythematous due to impingement of opposing tooth with no evidence of bleeding, pus discharge or sinus opening [figure 2 &3]. On palpation, the swelling was soft to firm in consistency, tender, non - compressible and non - reducible with no evidence of secondary changes. Based on the history and clinical findings a provisional diagnosis of Benign odontogenic cyst/ tumour in relation to 38 region was considered with a differential diagnosis of dentigerous cyst or odontogenic keratocyst. The patient was subjected to several investigations starting with laboratory investigations which revealed all the parameters [RBC count, WBC count, Hb %, platelet count, bleeding time, clotting time, ESR and RBS] were within normal range. His radiological investigations showed in orthopantomogram [figure 4], evidence of multilocular radiolucency involving the left body, ramus and angle of mandible, extending superioinferiorly from 1cm below the sigmoid notch up to inferior border of mandible, and mesiodistally from the distal aspect of 37 to 1cm anterior to the angle of mandible, circumscribing impacted 38 with presence of multiple septae present at right angles to each other, within the lesion. The lesion caused inferior displacement of inferior nerve canal and the inferior border of mandible appears to be intact. The patient was then subjected to higher investigations; CBCT [figure 5] which revealed Evidence of well - defined hypodense multilocular lesion seen in the left posterior mandible extending from 38 to the ramus of Size42.5 x 13.5mm in axial section and 50.5 x 38mm in sagittal section. There is also presence of Thinning and expansion of the lingual cortex in relation to inferior aspect of the lesion and Borders of the lesion appears to be scalloped. In few areas the septa appear to be straight and intersecting at right angles. the histopathological examination [figure 6] of incisional biopsy specimen showed loosely arranged stellate, oval or spindle shaped cells in a myxoid stroma with few

collagen fibrils and also stromal areas exhibiting prominent amount of collagen, numerous capillaries with areas of haemorrhage are also seen giving a Histopathological diagnosis of odontogenic myxofibroma. Based on history, clinical and investigative findings a Final diagnosis of odontogenic myxofibroma of left mandible was given. The patient was managed with segmental resection of left mandible and reconstruction with transport distraction osteogenesis [figute7] and kept under periodic follow - up and no recurrence had been noted till date.

3. Discussion

Myxomas are very rare benign tumors of mesenchymal origin. These tumors are locally invasive (McFarland et al., 1996; Simon et al., 2004; Martinez - Mata et al., 2008; Zarbo, 2010; Speight, 2013) and occur in various tissues, such as the heart, bones, skin, subcutaneous tissue, aponeuroses, genitourinary tract, and skeletal muscle (Kyriakos, 1990). 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) [1].

Odontogenic myxoma, also termed as odontogenic fibromyxoma or myxofibroma, is a subtype of myxoma occurring mainly in the hard, bony tissues of the face [11]. OM is frequently reported to be the fourth or third most frequent odontogenic tumour [12]. This neoplasm originates from tooth bearing areas of the jaw and is known for invading bone marrow spaces, showing a tendency to relapse after incomplete removal, as cellular neoplastic material can persist within small bone marrow spaces [13]. Zimmerman et al. reported that the average age for the odontogenic myxoma is 26.5 years, although majority of the investigators found that this lesion occurs in second or third decade of life [14]. Most of the reports suggest that there is a slight female preponderance, mandibular predilection, and the lesion has a silent locally destructive nature [8]. All these features were evident in our case with the difference in gender predilection making it a rare occurrence. This case, though a benign tumour, was a highly aggressive lesion, involving almost half of the mandible within a short span of 8 months. Another interesting finding was that it did not cause much of a cortical expansion or facial deformity and appeared to be invading the bone antero - posteriorly, as well as displayed scalloping between the roots of the involved teeth, in much the same manner as an odontogenic keratocyst. The radiographic features of the OM are variable, ranging from small unilocular lesions to large multilocular neoplasms, which often displace teeth or less frequently resorb roots [6]. The multilocular trabecular pattern has been described as “honey comb, ” “soap bubble, ” “wispy, ” “spiderweb, ” and “tennis racket [15]. The typical tennis racket appearance in radiological investigations is an implicit characteristic feature of odontogenic myxoma, which is evident in present case in orthopantomogram as well as in cone beam CT images making a clear - cut diagnosis of odontogenic myxoma without the need for additional investigations. Histologically, the myxoma is bland in appearance and is composed of loosely arranged, evenly dispersed spindle - shaped, rounded, and stellate cells with a lightly eosinophilic cytoplasm in a mucoid rich (myxoid), intercellular matrix [16, 17]. Many stellate tumour cells have anastomosing, long tapering, and cytoplasmic processes. Although some degree of mild nuclear pleomorphism may exist, including an occasional mitosis or binucleate cell [6]. The choice of treatment is surgery and decision should be made regarding with the type of surgical treatment modality that should be applied to each case due to its high recurrence rate [18]. The lack of capsule and infiltrative growth pattern is responsible for high rate of recurrence when conservative treatments like enucleation, curettage, and cryotherapy are performed. Radical treatment of block resection is advised by most authors over conservative treatment due to its invasive nature, large size, and recurrence history. The reason for high recurrence is due to the local invasion into cancellous bone beyond radiographically visible margins, absent encapsulation unlike benign neoplasm and presence of mucoid ground substance [19].

4. Conclusion

Myxomas are very rare and locally aggressive tumors and they tend to recur if not treated properly. Most cases of odontogenic myxomas are asymptomatic and are identified on a routine radiograph making an early diagnosis questionable but a thorough knowledge on a clinical presentation as well as radiological features will lead to a prompt diagnosis without delay. An accurate histopathological diagnosis, sound surgical planning and its implementation are required to improve the prognosis.

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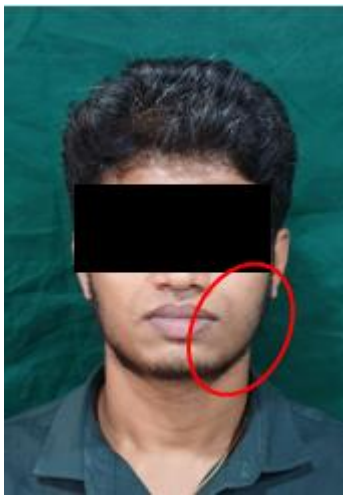

Figure 1

Profile

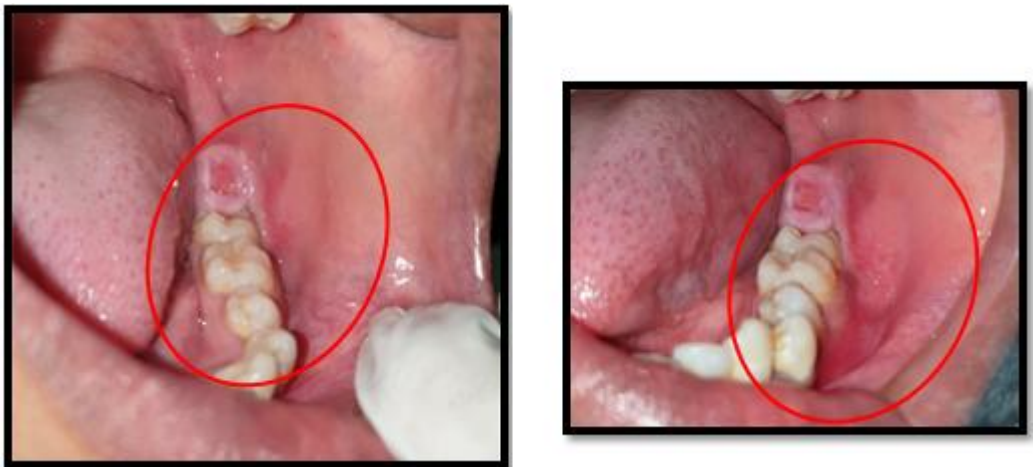

Figure 2 & 3: Left Buccal Vestibule in Relation To 37, 38

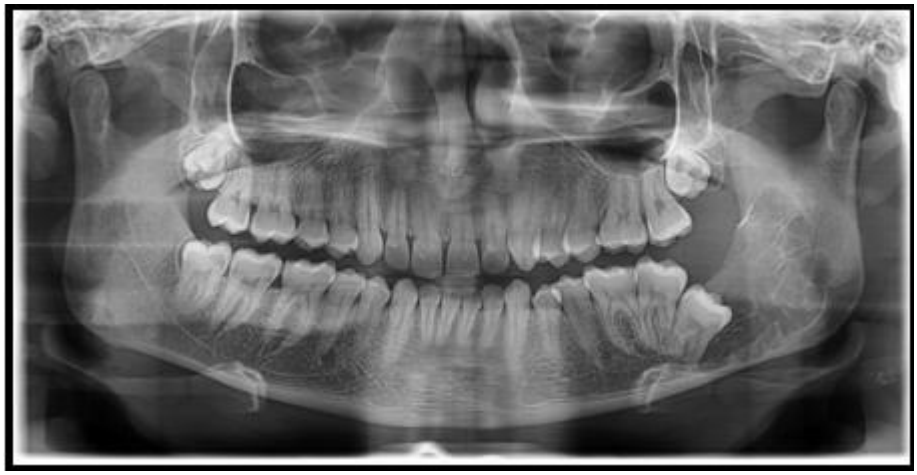

Figure 4: OPG

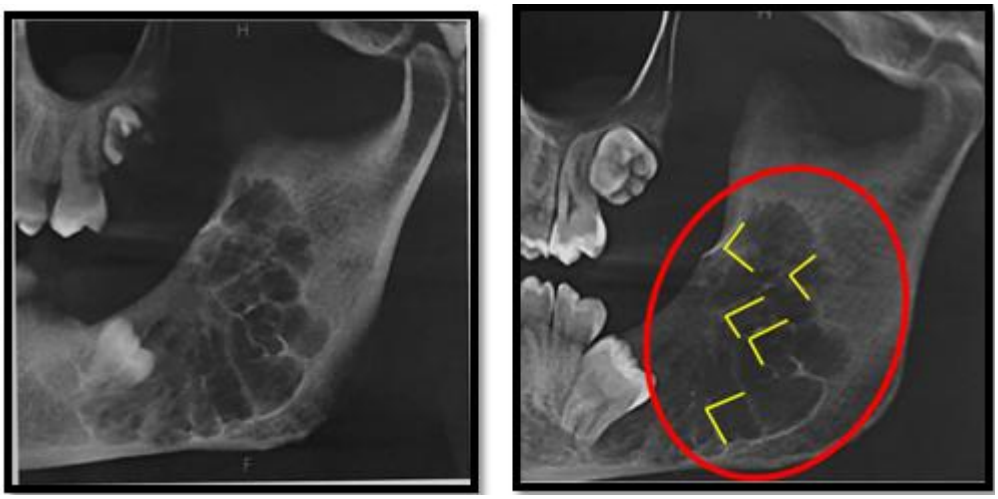

Figure 5: CBCT

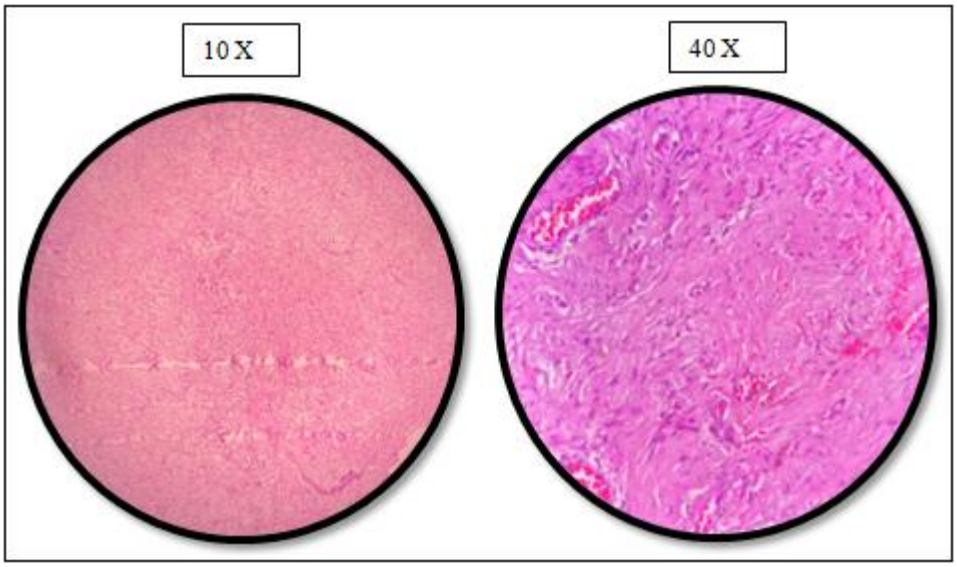

Figure 6: Histology

Courtesy: Department of Oral and Maxillofacial Pathology, Meenakshi ammal dental college and hospitals.

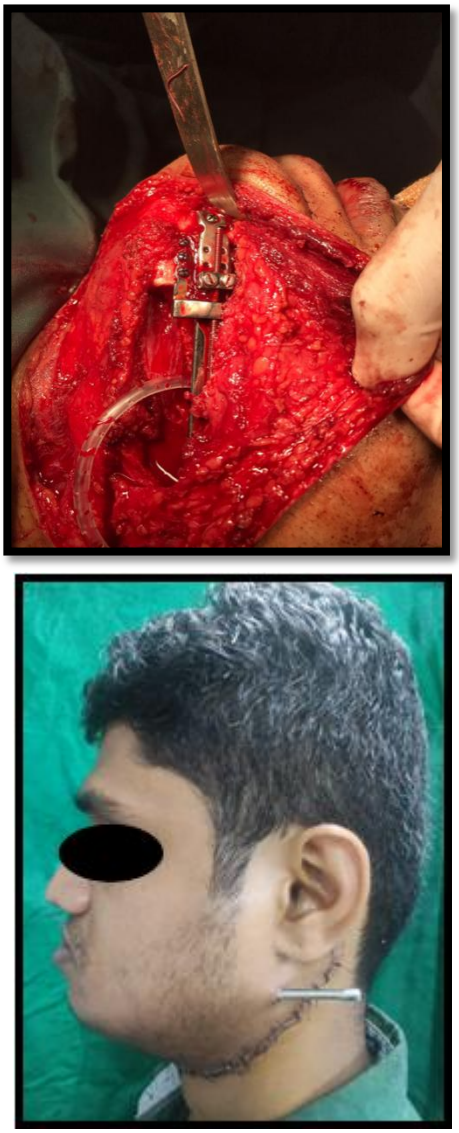

Figure 7: Treatment and Follow Up

Courtesy: Department of Oral and Maxillofacial surgery, Meenakshi ammal dental college and hospitals.