Treating Beyond Pockets- an unusual Case of Central Ossifying Fibroma and its Management

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ABSTRACT

Periodontal pocket and teeth mobility are common symptoms of patients having periodontitis. Although there are several reasons for teeth mobility, one of the reason is an odontogenic and non-odontogenic tumor. An ossifying fibroma is a rare benign fibro-osseous neoplasm of the jaw characterized by the substitution of normal bone by fibrous tissues and newly formed calcified products such as bone, cementum or both. This case report describes an unusual case of tooth mobility and swelling in mandibular anterior teeth region with respect to 31, 32 and 33 teeth of a 28 year-old female patient.Initially, the clinical symptoms were suggestive of the periodontal pocket. However, the radiographical, surgical and histological findings confirmed the diagnosis of Central ossifying fibroma. Controversies regarding the terminology and classification along with the differential diagnosis are discussed and a review is provided of the literature on the subject.

Keywords: *Fibro-osseous lesion, Ossifying fibroma, Tooth mobility.*

The term fibro-osseous lesion (FOL) is a generic designation of a poorly defined group of lesions affecting the jaws and craniofacial bones. They comprise a diverse group of pathologic conditions including developmental lesions, reactive or dysplastic lesions, and neoplasms. All are characterized by the replacement of bone by a benign connective tissue matrix [1]. Clinically, the cemento-ossifying fibroma presents as a painless, slowlygrowing mass in the jaw where the displacement of teeth may be the only early clinical feature. Tumor shows female preponderance in the range of 20-40 years. In the mandible, its occurrence is 70-90%, where it occurs more frequently in the premolar-molar region followed by the involvement of maxilla, ethmoidal and orbital regions also. Also, a few cases showed bilateral involvements [2]. Mobility of teeth can be because of several factors like periodontal disease, trauma, pregnancy,odontogenic or non- odontogenic tumor or parafunctional habits. Correct diagnosis is very much important in every case. Tooth mobility is one of the most important symptoms of Central ossifying fibroma (COF).

An ossifying fibroma is a neoplasm that, if left untreated, may exhibit considerable growth and bone destruction [3]. The following is one such case, where the patient complaint of tooth mobility in the mandibular teeth region. However, the radiological and histological investigations led to the diagnosis of COF.

CASE REPORT

A 27-year-old woman was referred for the evaluation of mobility of teeth in the left mandibular anterior region of the jaw. Also, there was a painless firm swelling expanding on the lingual side of the same region. The patient stated that the mobility and swelling had been gradually increasing in size for 5 months. The past medical and dental histories were non-contributory. Also, the family history was not contributory.

On clinical examination, there was a 5 mm deep pocket and grade 2 mobility with the mandibular anterior teeth specifically 31, 32 and 33. A localized swelling measuring approximately 8 x 5 cm in size was present in the left lingual side of the mandible in the lateral incisor-canine region (31, 32 and 33) (Fig. 1). The swelling was bony-hard in consistency and asymptomatic completely. An obliteration of lingual vestibule in the lateral incisor-canine region (31,32,33) was also noted. No abnormality

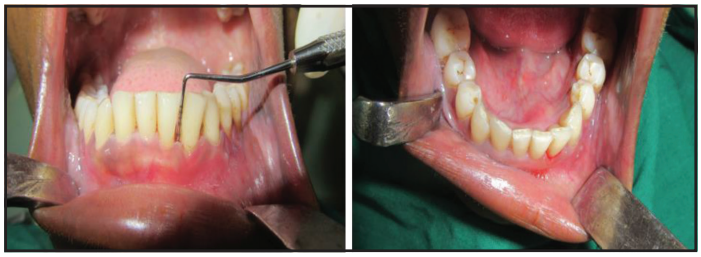

Figure 1: Preoperative view of the patient showing 5mm deep pockets and swelling in the anterior region of the mandible.

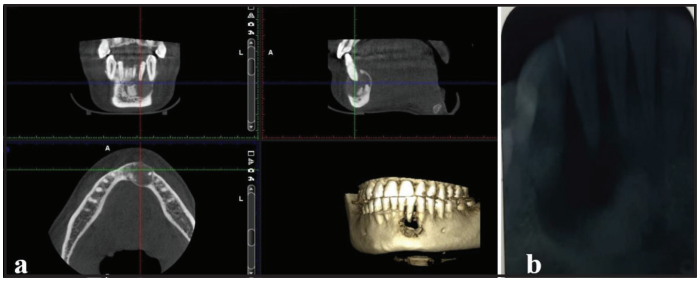

Figure 2: (a) CBCT and (b) IOPA of the patient.

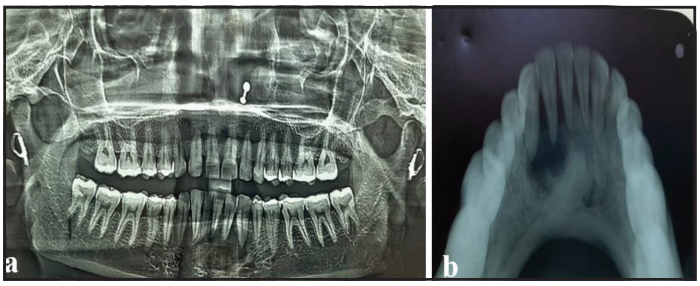

Figure 3: (a) OPG and (b) occlusal radiograph of the patient.

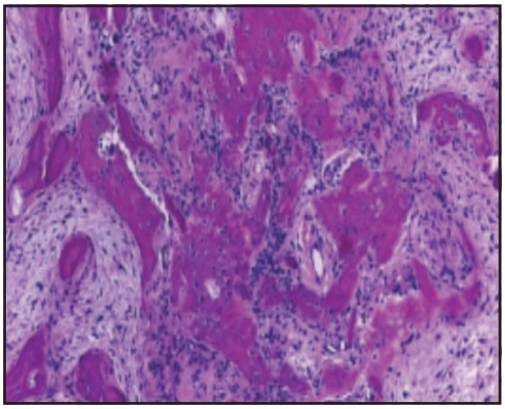

Figure 4: Histopathological investigation of the patient.

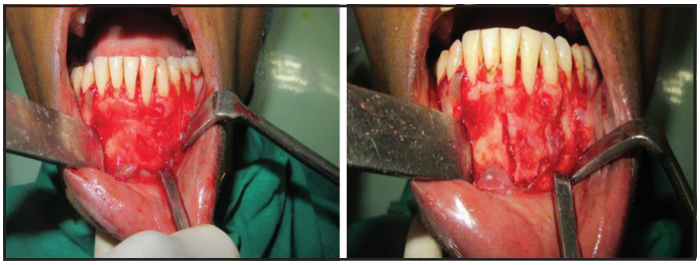

Figure 5: Surgical procedure for removing the lesion.

was detected in relation to the vitality of teeth in the affected area. No lymphadenopathy was detected.

Diagnostic guidelines including complete hemogram, serum calcium, serum phosphorus, serum alkaline phosphatase were suggestive of no abnormalities. Radiographic analysis was also done with the help of intraoral periapical radiograph (IOPA) (Fig. 2b), Orthopantomogram (OPG), occlusal radiograph and Cone-beam computed tomography (CBCT). CBCT scan revealed a heterodense lesion with a regular expansion of the lingual cortical plates and with central foci of calcification (Fig. 2a). OPG revealed a mixed radiolucent-radiopaque lesion of size approximately 8× 5 cm extending from 31 to 33 region. The extension starts superiorly from the apical portion of 31 and inferiorly upto the superior band of the inferior border of the mandible. The inferior border of mandible was intact. The borders of the lesion were well-corticated and well-defined (Fig. 3a).In radiographic evaluation, mandibular occlusal cross-sectional view showed a region of cortical expansion on the lingual surface of the left lateral incisor-canine (31, 32 and 33) (Figure 3b). These findings led to radiographic diagnosis of ossifying fibroma.

In order to investigate further, the patient was advised for an incisional biopsy. The incised specimen was sent for histopathologic evaluation. Histopathological examination confirmed the diagnosis of Ossifying Fibroma by revealing the presence of immature bone trabeculae with entrapped osteocytes and lined by a dense rim of enlarged osteoblasts (Fig. 4).

The patient was scheduled for surgical excision of the lesion. The surgery was performed under local anesthesia 1:200000 Lignocaine and Adrenaline. The full-thickness flap was raised from the central incisor to the premolar region (31-34). The area of lesion was completely exposed. The bony lesion was demarcated with a round bur. Chisel and mallet technique was used to separate the entire lesion (Fig. 5). The entire lesion was excised and was sent for histopathological examination (Fig. 6). The bleeding was controlled with bone wax and the flap was sutured with 4-0 Mersilk suture (Fig. 7a). Antibiotics (Amoxicillin 500 mg), anti-inflammatory and analgesic (Enzoflam) was prescribed for thrice daily for 5 days. The patient was recalled after 1 week to evaluate the surgical site. The swelling had subsided and the pain was reduced. The patient had difficulty in mastication for which she was advised prosthetic rehabilitation. The patient is

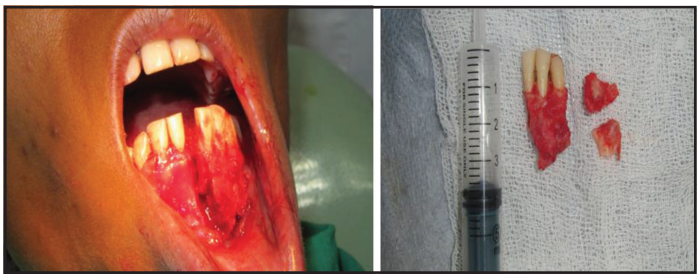

Figure 6: Resection of the lesion.

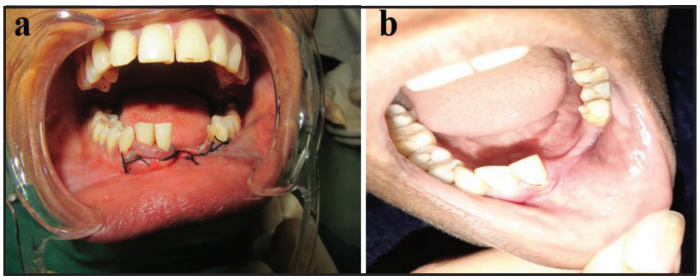

Figure 7: (a) Suturing done; (b) Follow-up of the patient after 6 months.

under follow-up for last 6 months, and no recurrence has been reported yet (Fig. 7b).

DISCUSSION

There has been a lot of controversies regarding the terminologies and classification of ossifying fibroma. In 1971 the World Health Organization (WHO) classified four types of cementum-containing lesions: fibrous dysplasia, Ossifying fibroma, Cementifying fibroma and COF [4]. According to the second WHO Classification, benign fibro-osseous lesions in the oral and maxillofacial regions were divided into two categories, osteogenic neoplasm and non-neoplastic bone lesions; Cementifying ossifying fibroma belonged to the former category. However, the term "Cementifying Ossifying Fibroma" was reduced to ossifying fibroma in the new WHO classification in 2005 [5]. In 2008, Eversole et al. gave a comprehensive classification by including developmental lesions, neoplastic lesions and inflammatory/ reactive processes [6].

An ossifying fibroma is a true neoplasm with significant growth potential. The neoplasm is composed of fibrous tissue that contains a variable mixture of bony trabeculae, cementum like spherules or both [7]. COF is the most common benign fibro-osseous neoplasm of the oral and maxillofacial region. It was described by Menzel in 1872 but was appointed by Montgomery in 1927. This lesion tends to occur in the second and third decades of life, commonly in women, and in the mandibular premolar and molar areas [8-11].

The etiology of ossifying fibroma is unknown but odontogenic, developmental and traumatic origins have been suggested.There are several theories regarding its origin, one of which theory suggests that COF is of periodontal origin. This theory is based on the concept that periodontal ligament has the capacity to produce cementum and osteoid material [12]. Ossifying fibroma develops from the multipotential mesenchymal cells of periodontal ligament origin which are able to form both bone and cementum [13,14]. In 1968, Hamner et al did clinical, radiographic, histologic analysis and follow up of 249 cases of fibro-osseous jaw lesions of periodontal membrane origin and classified them. Benign fibro-osseous lesions of periodontal membrane origin were found to be much more prevalent in the jaws (249 cases) than fibro-osseous lesions of medullary bone origin (154 cases) [15].

In 1973, Waldron and Giansanti reported 65 cases (of which 43 cases had adequate clinical histories and radiographs) and concluded that this group of lesions is best considered as a spectrum of processes arising from cells in the periodontal ligament and have the potential to form bone, cementum, and fibrous tissue in varying combinations. Some of these lesions were reactive in nature, while others appear to be neoplastic [16]. In 1985, Eversole et al described the radiographic characteristics of COF, and two major patterns were noted, expansile unilocular radiolucencies and multilocular configuration [17].

The triggering mechanism for its derivation from aberrant periodontal membrane growth or development from endosteal fibrous tissue remains controversial. Wenig et al (1984) described the case of COF in a 26-year-old Hispanic male that has suggested that trauma-induced stimulation may play a role in its origin [18]. Inactivation of the HRPT2 tumor suppressor gene is associated with the pathogenesis of the hereditary hyperparathyroidism-jaw tumor syndrome and malignancy in sporadic parathyroid tumors. Recent genetic studies have revealed a mutation in tumor suppressor gene HRPT2, a protein product known as Parafibronin which leads to tumor formation [19].

The differential diagnosis of COF is fibrous dysplasia, cemento-osseous dysplasia, condensing osteitis, Pindborg's tumor, retained root, and odontome. Fibrous dysplasia generally has ground glass appearance not seen in COF. Pindborg's tumor has a high association with impacted teeth. Odontome can be differentiated by the presence of tooth-like structure. Vitality test helps to differentiate it from condensing osteitis. Multiple COF occurs rarely [20,21].

The clinical management of COF remains uncertain. In some reports, the authors favor conservative surgery rather than en bloc resection. Some cases treated by conservative surgical excision have shown no recurrence over a 17-year follow-up period [22]. In contrast, Zama et al. (2004) reported an immediate recurrence 15 days after conservative surgery to treat COF of the mandible, which required a second operation for hemi-mandibulectomy and reconstruction [23]. To avoid or minimize the chance of recurrence, a partial or en bloc resection of the jaw is preferred for larger lesions [24,25]. Our reported case had a large lesion, the surgical protocol applied was resection because the lesion was well-circumscribed and could be separated from normal bone during surgery, and the current follow-up has not shown any clinical signs of recurrence.

CONCLUSION

Correct diagnosis is very much important in every case. For ossifying fibromas, surgical excision is the treatment of choice, and the well-circumscribed nature of this lesion allows relatively easy removal. The prognosis is excellent after complete excision. Recurrence of the lesion is a rare phenomenon.

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