Case Report

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"Cemento-ossifying Fibroma: A Comprehensive Exploration Through A Striking Case Report"

Abstract:

Cemento-ossifying fibroma (COsF) is a benign mesenchymal odontogenic tumor. The origin of COsF occurs within the periodontal ligament, housing multipotent stem cells capable of generating cementum, lamellar bone, and/or fibrous tissue. This case report of a 35-year-old male reported swelling in the left lower posterior jaw region over the past year. The objective of this report is to provide clinicians and researchers with insights into the diagnostic challenges, treatment modalities, and the evolving landscape of COF, facilitating improved patient management and furthering the discourse on this intriguing maxillofacial pathology

Key-words: .....

Introduction:

Cemento-ossifying fibroma (COsF) is a benign mesenchymal odontogenic tumor in the W.H.O. 2022 classification[1], affecting the maxillofacial region, characterized by a slow and progressive growth pattern. The controversy surrounding its terminology, origin, and diagnostic criteria has sparked considerable discussion in recent literature[2,3]. This central neoplasm of bone and the periodontium has been a subject of debate, with some studies emphasizing its close relation to other lesions like fibrous dysplasia[4].

Cemento-ossifying fibroma (COF) is a distinctive fibro-osseous lesion categorized within the jaw region. This rare benign tumor is characterized by its slow and progressive growth, commonly presenting in the mandible and occasionally in the maxilla[5]. COF typically manifests as well-circumscribed masses with the potential to expand the underlying bone, leading to its classification as a fibro-osseous lesion of the jaws. While these tumors are usually small, they can attain significant sizes if left untreated. Diagnosis and differentiation from other lesions involve careful consideration of clinical, radiological, and histopathological characteristics, ensuring an accurate understanding of the nature and potential complications associated with COF[6].

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Case Presentation:

A 35-year-old male visited the Oral Medicine and Radiology outpatient department, reporting a swelling in the left lower posterior jaw region over the past year. Approximately a year ago, he was asymptomatic, but subsequently experienced teeth mobility and observed a gradual enlargement of a mild swelling on the left lower jaw. The patient did not report any

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past pain, and both medical and dental histories were unremarkable. Upon general examination, the patient exhibited moderate build, and no signs of pallor, icterus, clubbing,edema, or cyanosis. Vitals were within normal ranges. He was a habitual tobacco chewer and betel smoker for the past 3 years.

Extra orally, a well-defined, dome-shaped swelling was observed extending from the Superoinferiorly alveolus tothe lower border of the mandible and mesiodistally from the commissure to the angle of the mandible (Figure 1). Palpation revealed a bony-hard, afebrile, non-fluctuant, non-mobile, and non-compressible swelling with no translucency and no palpable pulsation.

Intraorally, a swelling extending from 34,35,36,37 and 38. There was expansion of both buccal and lingual cortical plates which was accompanied by the loss of vestibular space. The swelling was bony hard in consistency, and non-tender on palpation. The swelling was associated with grade II mobility of the teeth from the premolar to the third molar of the left side. A provisional diagnosis of benign neoplasm of bone with odontogenic/non-odontogenic was made. A differential diagnosis of ameloblastoma, odontogenic myxoma, CEOT, cemento-ossifying fibroma, and fibrous dysplasia was considered.

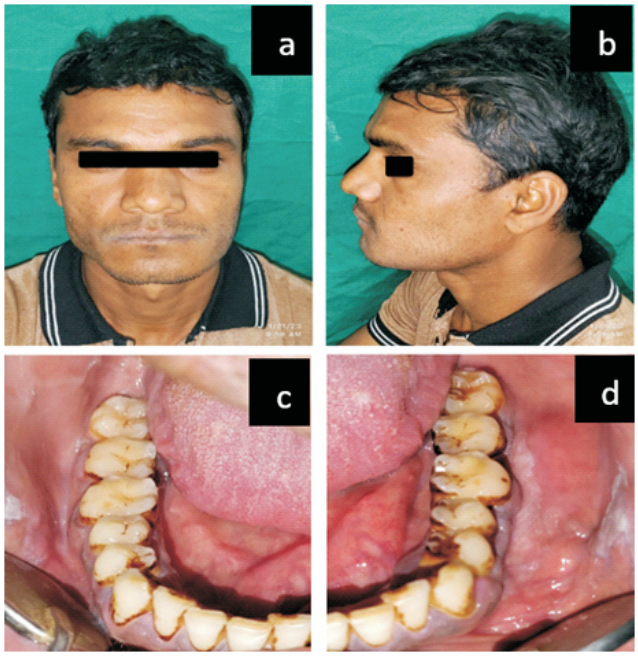

Figure 1: **a,b** Showing extraoral photographs with mild facial swelling over the body of the left mandible. **c,d** image showing the intraoral image of the right and left side of the mandibular arch respectively. **d** showing vestibular obliteration from 34,35,36,37 and 38 regions.

Given the patient's history and clinical observations, the following procedures were conducted:

- Radiographic examinations, including Orthopantomogram (OPG) and Computed Tomography (CT) scan.
- Hematological investigations.
- An incisional biopsy.

Orthopantomograph (OPG):

The Orthopantomograph OPG revealed a well-defined lesion extending from the mesial surface of the mandibular first premolar to the distal part of the mandibular third molar. There was a corticated outline, delineating the interior of the lesion from the surrounding adjacent normal bone. Areas of mixed radiopacity and radiolucency with sparse septae represented the internal architecture of the lesion. there was evidence of multiplanar root resorption of 34, 35, 36, 37, and 38. Conspicuous thinning and bowing of the inferior border of the mandible were also observed. (Figure 2)

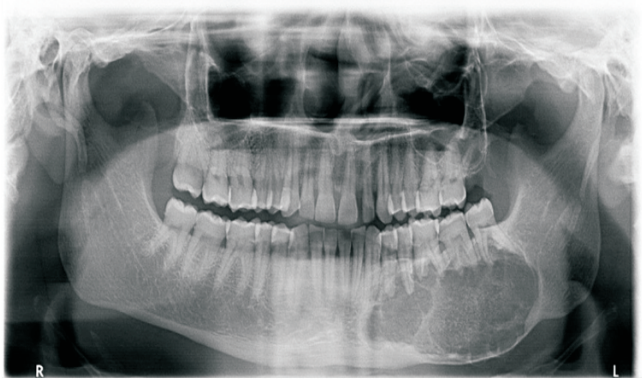

Figure 2- Orthopantograph (OPG) reveals a mixed radiolucent-radiopaque lesion (yellow arrow) extending from the mandibular first premolar to the molar region. There is evidence multiplanar of root resorption in the mandibular right second molar (white arrow)

Computed tomography scan (CT):

The large well-defined expansile lesion in the left mandible with equidirectional expansion measuring 5.6 x 3.4 x 3.7 cm in the left of the mandible and involved the ipsilateral alveolar ridge. Marked Multiplanar root resorption is seen with 34,35,36,37 and 38. There was remodeling of the adjacent cortex and e, with focal erosions and thinning. The lesion showsnon-coalescing hyperdense calcific foci within the lesion. (Figure 3 and 4)

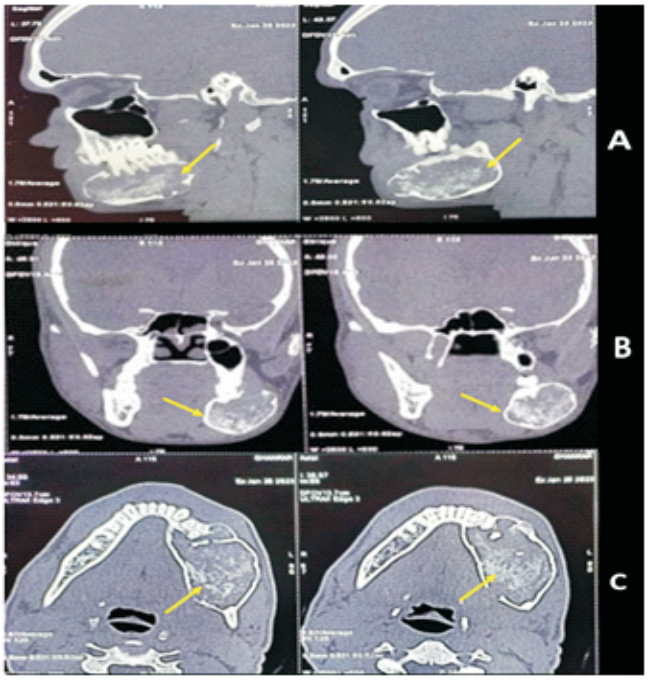

Figure 3 Computed tomography scan of Sagittal section (A), coronal section (B),and Axial Sections showing the expansile lytic lesion with non-coalescing hyperdense calcific foci within the lesion.

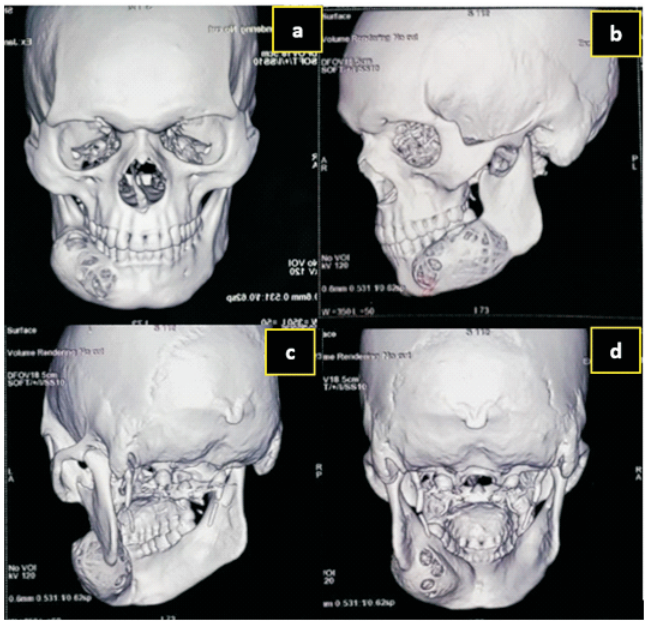

Figure 4: Computed tomography three-dimensional surface shaded display images from frontal (a) and lateral (b) views showing the expansile lesion with smooth outlines without any cortical break.

Hematology report:

Hemoglobin-14.4mg%, total count-2500 cell/mm^3, Bleeding time (Dukes method)-1 minute 20 seconds, Clotting time (Lee and White method): 5 minutes 40 seconds, differential count: Neutrophils-65%, lymphocytes-30, eosinophils-03, monocytes-02%. PTT 15sec, INR 1.2, Serum Sodium 141.0mEq/l, Serum Potassium 4.68mEq/l. HIV, HBsAg was found to be negative by ELISA (tri dot and rapid kit respectively).

Management:

Preoperative antibiotics (amoxicillin 250 mg, one tablet three times a day) were prescribed for three days, and a planned excisional biopsy was executed. The surgical excision, performed under local anesthesia, included primary closure of the lesion using 3-0 silk sutures. The excised mass, along with teeth 35, 36, 37, and 38, was submitted to the Department of Pathology for histopathological examination. Antibiotic and anti-inflammatory drug regimens were maintained for the following week. Complete suture removal occurred on the seventh postoperative day, during which the lesion exhibited signs of regression. A follow-up examination 15 days later revealed regression of the lesion (Figure 5), and the normal edentulous ridge was restored.

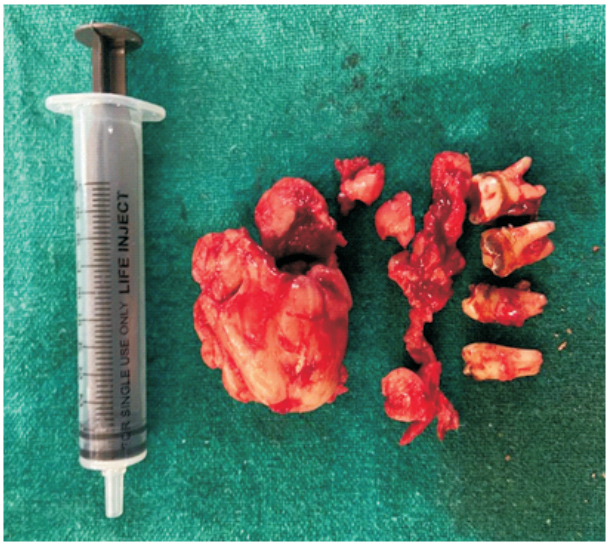

Figure 4:Gross specimen after surgical enucleation

Histopathology:

Using a sub-labial incision, a biopsy was performed and sent to our department for examination. The given H &E-stained section under microscopic examination showed highly cellular connective tissue stroma interspersed with plump to fine fibroblast. Stroma was richly vascular with multiple proliferating blood vessels. Numerous areas of mineralization are seen. The mineralizing portion is basophilic and poorly cellular spherules with a brush border appearance that resembles cementum. Suggestive of Cemento Ossifying Fibroma. (Figure 5)

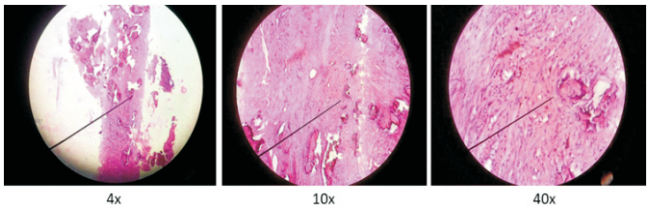

Figure 6: Sowing Histopathological image at 4x,10x and 40x Hence, the Final Diagnosis of Cemento Ossifying fibroma was confirmed. Six-month follow-up showed no signs of recurrence.

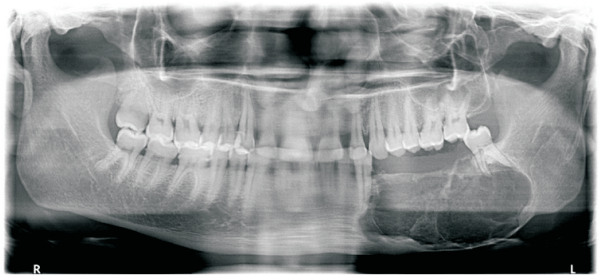

Figure 7: Showing OPG after 6 months follow-up.

Discussion:

In 1872, Menzel provided the initial account of a variation of ossifying fibroma, naming it cement-ossifying fibroma[6]. Subsequently, Montgomery detailed it further in 1927. Makek proposed the term 'benign periodontoma' based on its etiology. By 1971, the World Health Organization (WHO) categorized COF within cementum-forming lesions, encompassing fibrous dysplasia, ossifying fibroma, and cementifying fibroma. Nevertheless, in 2005, the terminology was streamlined to simply ossifying fibroma [7]. COsF, previously categorized as a benign mesenchymal odontogenic tumor in the 2017 classification but later elaborated under Fibro-osseous and osteochondromatous lesions[8], is now distinctly recognized as an integral component of benign mesenchymal odontogenic tumors in the 2022 classification[3]. This classification separates COsF from the non-odontogenic juvenile trabecular and psammomatoid types. The pathogenesis of COsF in a minority of cases is associated with inactivating mutations in the tumor suppressor gene CDC73 (HRPT2), typically observed in instances linked to hyperparathyroidism-jaw tumor syndrome[9]. COsF may also be associated with gnathodiaphyseal dysplasia, identified by GDD1 gene mutations[10].

Clinically it presents as a slow-growing tumor of the jaw most often seen between the third and fourth decades of life, which is consistent with the present case. They occur more frequently in women than in men and the predilection for the site is mostly in the mandibular premolar region[11]. Our case shows the lesion in the posterior region of the mandible in concordance with the previous reports.

The growth is asymptomatic, although the patient may present with a swelling resulting in asymmetry[6]. In our cases, the patients presented with asymptomatic swelling that persisted for a long duration.

The radiographic appearance is influenced by the degree of mineralization. Initially, Cemento-Ossifying Fibroma (COF) may manifest as a radiolucent lesion, either unilocular or multilocular. As the lesion progresses, the lucent presentation evolves into a radiopaque form, resulting in a lesion with mixed density. The lesion's margin is relatively well-defined, featuring a sclerotic rim in the host bone due to peripheral osteocondensation[7].

Various authors have detailed the radiographic characteristics of the cemento-ossifying fibroma (COF). According to Waldron and Giansanti (1973)[12], COF exhibits lytic lesions in 26% of cases, 63% show a combination of lytic and radiopaque foci, and 12% display a diffuse and homogeneous appearance. In contrast, Barberi et al. (2003)[13] categorized the radiographic patterns into three groups: lesions without a sclerotic rim (40%), lesions with a sclerotic rim (45%), and lesions with an ill-defined border (15%). Our observed case aligns with the findings of Waldron et al.[12]and Barberi et al.[13].

The crucial diagnostic characteristic of Cemento-Ossifying Fibroma (COF) lies in its centrifugal growth pattern, exhibiting equal expansion in all directions, resulting in a rounded tumor mass[14]. In our case, the CT images consistently reveal an enlargement of both buccal and lingual cortical plates with thinned yet intact margins, affirming the benign nature of the lesion. Additionally, the most prevalent observation in mandibular lesions is the inferior bowing of the lower mandibular border also manifested[14].

Root resorption may be observable, and the displacement of neighboringteeth[13] is multiplanar root resorption is consistent with our case, but there was no tooth displacement of teeth indicating an active proliferating stage with continuous enlargement of the tumor. The potential differential diagnoses include fibrous dysplasia, cement-osseous dysplasia, condensing osteitis, pindborgs tumor, and odontoma.

The primary approach to managing COF involves enucleation for smaller lesions and mono-bloc resection with bone reconstruction for larger cement-ossifying fibromas.

Radiotherapy is a challenging option due to the radio-resistance of COF and the potential for post-radiation complications, making it a complex treatment avenue. The prognosis for this condition is generally favorable; however, the likelihood of relapse is higher in maxillary COF cases than in mandibular ones. This is attributed to the increased challenge of surgically removing maxillary lesions and their larger size at the time of initial presentation[14].

Conclusion:

Fibro-osseous lesions hold significant importance for oral and maxillofacial radiologists. The pathology of all fibro-osseous lesions is similar, necessitating clinical and radiological correlation. COsF is now distinctly recognized as an integral component of benign mesenchymal odontogenic tumors in the WHO 2022 classification[1]. This case report contributes to the evolving understanding of COsF by presenting an unusual case of this jaw lesion, considering the varied patterns of bone formation in a fibroblastic stroma, and adding to the existing body of knowledge on its clinical manifestations. The objective of this report is to provide clinicians and researchers with insights into the diagnostic challenges, treatment modalities, and the evolving landscape of COF, facilitating improved patient management and furthering the discourse on this intriguing maxillofacial pathology.

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