

# Concurrence of complex odontoma and cemento ossifying fibroma: A rare case report

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## ABSTRACT

Odontomas are the foremost common sort of odontogenic tumors containing enamel, dentin, and cementum and pulp tissue. They are included under the benign calcified odontogenic tumors. Odontomas are basically classified into two types, complex and compound odontomes. Various theories or etiological factors are been quoted for the occurrence of odontomes. Generally, they're asymptomatic.

Cemento-ossifying fibroma defines as a relative rare osteogenic neoplasm of the jaw. This tumor includes fibrous and osseous components. During this paper we report a rare case of ossifying fibroma related to complex odontoma within the mandible.

**Keywords:** Cemento ossifying fibroma, Complex odontoma, Mandible

## INTRODUCTION

The concept of fibro-osseous lesions of bone has evolved over the last several decades and now includes two major entities: fibrous dysplasia and ossifying fibroma, also because the other less common lesions like florid osseous dysplasia, periapical dysplasia and focal sclerosing osteomyelitis. Fibroosseous lesions of the jaws are classified by Waldron (Waldron CA 1993 and Kramer et al. 1992). The cement-ossifying fibromas or ossifying and cementifying fibromas are described also demarcated or rarely encapsulated neoplasms, consisting of animal tissue containing varying amounts of mineralized material resembling bone and/or cementum. (Kramer et al. 1992) These benign fibro-osseous lesions can arise from any a part of the facial skeleton and skull with over 70 per cent of cases arising within the head and therefore the neck region and principally within the jaws. (Cakir B et al. 1991 and Mitrani M et al. 1988) Radio-graphically, the neoplasm may be a well-delineated radiolucent or mixed radiolucent and radiopaque lesion counting on the

contributions of sentimental and hard tissue components.

The term odontoma refers to any tumor of odontogenic origin. An odontome may be a growth during which both epithelial and mesenchymal cells exhibit complete differentiation with the result that functional ameloblasts and odontoblasts form enamel and dentin (Shafer WG et al. 1983). These enamel and dentin are usually laid down in an abnormal pattern because the organization of the odontogenic cells fails to succeed in a traditional state of morph differentiation. Odontoma represents a hamartomatous malformation instead of a neoplasm. It's the foremost common odontogenic tumor, representing 67% of all odontogenic tumors (Wood NK et al. 1997).

A few articles have reported simultaneous occurrence of two or more odontogenic tumors or central lesions within the jaws (Gamoh S et al. 2015 and Pushpanshu K et al. 2013). There is one report of subsequent occurrence of COF at the location of odontoma (Matsuo K et al. 2013) and another

article describing complex odontoma related to ossifying fibroma (Hosseini et al. 2011). Herein, we report uncommon occurrence of complex odontoma and COF within the same region.

## CASE REPORT

A 65 years old female patient reported to the department of oral medicine and radiology with the chief complaint of swelling within the anterior part of mandible over 2 months. No evidence of facial asymmetry or lymphadenopathy was seen.

A bony hard swelling was seen within the mandibular anterior teeth region measuring approximately 2 X 1.5 cm in size. Figure 1 Patient gives the history of extraction of teeth with same region 2 months back. Pus discharge was seen with an equivalent region. No relevant medical record was reported by the patient.



Figure 1: Preoperative view.

There was an expansion of buccal and lingual cortical plates of anterior region of mandible, extending from the region of right first premolar to right central incisor. All the teeth from right first premolar to left lateral incisor were missing. There was intact overlying mucosa, no mobility on palpation, tenderness or paresthesia. Vitality test was performed on the proper second premolar and left canine as they were tender on palpation. Both the teeth were vital.

On the idea of history and clinical findings, a provisional diagnosis of infected residual cyst was given and patient was referred for the radiographic investigations. Bird's eye view revealed a mixed radiolucent-radiopaque lesion located within the anterior region of mandible. (Figure 2)



Figure 2: Panoramic view.

In cone beam computerized tomography (CBCT), a well-defined mixed radiopaque and radiolucent lesion was seen in lower anterior region extending from right first premolar to left lateral incisor region crossing the midline (Figure 3). Borders were well demarcated and corticated. The size of the lesion in coronal section, axial section and sagittal section were 18.27 X 18.60 mm, 14.28 X 20.49 mm and 13.76 X 14.72 mm respectively. Thinning and expansion of buccal and lingual cortical plates were seen. Within the substance of the lesion, multiple radio-pacities of variable sizes seen resembling denticles.

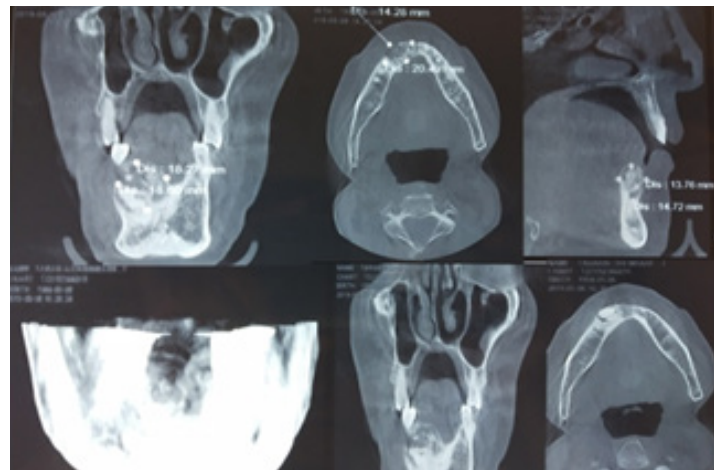


Figure 3: CBCT of the lesion.

Our medical diagnosis, on the idea of clinical and radiographic examination were: calcifying odontogenic cyst (or Gorlin's cyst) which usually develops within the incisor-canine regions and radio-graphically presents as a well-circumscribed, unilocular radiolucency that contains irregular calcifications, additionally it's sometimes related to unerupt-

ed teeth or odontoma (Steinberg MJ et al. 2001) and Cemento-ossifying fibroma (COF) that shows a predilection for females. The mandible is that the most ordinarily involved site. The radiographic borders appear well defined and mostly corticated. The density of the lesion is mixed. The lesion tends to be concentric within the medullary a part of the bone with outward expansion approximately equal altogether directions (Liu Y et al. 2010).

The patient was taken for surgery under local anesthesia in any case the blood investigations being done. The patient was prepped with painting and draping done. Inferior alveolar nerve block and native infiltration within the region was done using 2% Lignocaine with 1:100000 adrenaline. A crestal incision was made with two lateral releasing incisions within the buccal vestibule. The incision was far away from the lesion on sound bone. A full thickness mucoperiosteal flap was raised to show the calcified mass using sharp dissection with a periosteal elevator. Drilling round the periphery of the mass was through with the assistance of round burs and straight handpiece under constant irrigation and suction. The calcified mass was easily demarcated from the encompassing healthy bone (Figure 4). It had been curetted out with a pointy curette and therefore the resulting bony bed was thoroughly irrigated using saline and betadine. It had been later full of gel foam and therefore the wound was closed using 3-0 mersilk sutures (Figure 5). A pressure pack dressing was given to realize complete hemostasis. The patient was given antibiotics and analgesics for five days post-operatively. Patient's tolerance and cooperation was good. The excised specimen was then sent for the histopathological evaluation. (Figure 6)



Figure 5: Post-operative view after suture placement.

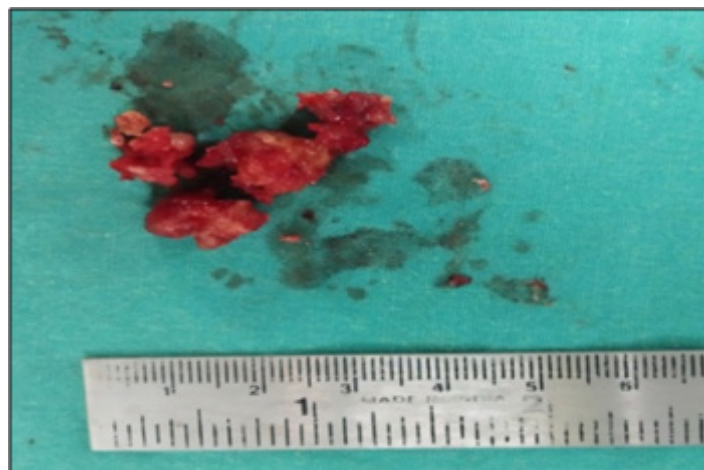


Figure 6: Excised biopsy specimen.



Figure 4: Intraoperative view.

Histopathological picture had shown highly cellular fields with some calcified areas. Cellular component was composed of fibroblasts arranged in different patterns. Calcified areas appeared to be composed of cementum like material. The other part of the lesion was composed of tooth like structures with tubular dentin around the pulp-like tissue. (Figure 7) Excisional biopsy of the lesion confirms a diagnosis of cemento-ossifying fibroma associated with complex odontomas.

No recurrences or post-operative complications were observed during the 2 months follow-up period and panoramic and intraoral periapical radiographs showed healing of lesion. (Figures 8 and 9)



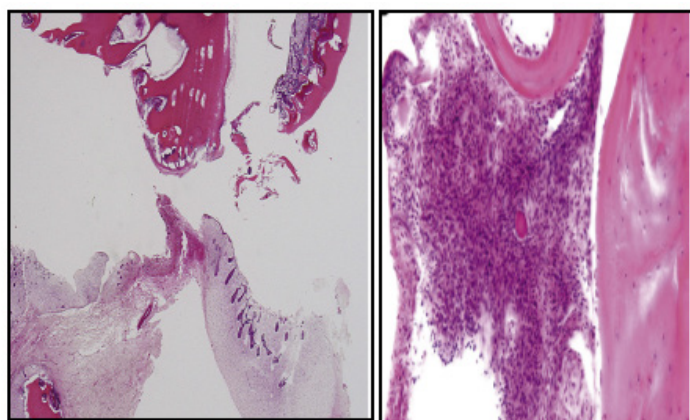


Figure 7: Histopathological view.

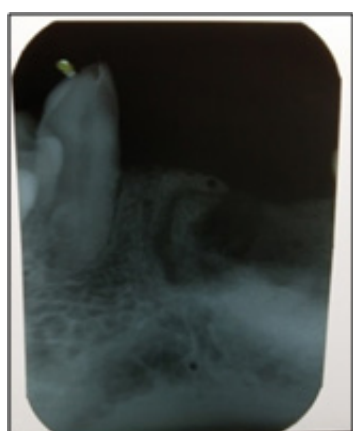


Figure 8: Post op IOPA.



Figure 9: Post-operative OPG.

## DISCUSSION

Cemento-ossifying fibroma is a benign fibro-osseous tumor (Galdeano M et al. 2004) these tumors are thought to arise from the periodontal ligament, and are composed of varying amounts of cementum, bone, and fibrous tissue. Eversole et al. (1985) reported that the production of these cementum like structures may be associated with membranous bone, and may not only be related to cementogenesis (Eversole LR et al. 1985) The WHO classifies cementoossifying fi-

broma as a fibro-osseous neoplasm, included among the non-odontogenic tumors, derived from the mesenchymal blast cells of the periodontal ligament, and with a potential to form fibrous tissue, cement and bone, or a combination of such elements (Wang H et al. 2010). The hybrid name central cement-ossifying fibroma is also used, as these tumors can display a spectrum of Fibroosseous lesions, ranging from those with only deposition of cementum to those with only deposition of bone, and arising from the periodontal ligament (Tamiolakis D et al. 2005). This tumor is more frequent in the premolar and molar regions of the mandible in the third and fourth decades. Female are more commonly affected. Clinical presentation is usually a spherical or ovoid expansion of the jaw. Small lesions are rarely symptomatic and usually are discovered during radiographic examinations. Larger lesions cause painless enlargement of the involved bone and Considerable facial asymmetry. Pain and paresthesia are rarely associated with COF (Sarwar HG et al. 2008).

Complex odontomas are the malformation in which all dental tissues are represented but not in an organized form or disorderly pattern. It is an odontogenic tumor characterized by the formation of calcified enamel and dentin in an abnormal arrangement because of lack of morph differentiation.

In this case, patient reports a history of long term growing of the lesion on her jaw, with bony hard consistency which suggests a nonaggressive intrabony lesion. Occurrence of simultaneous odontogenic lesions or simultaneous odontogenic and non-odontogenic lesions, described combined lesions, sometimes called hybrid lesions which is extremely uncommon and have been reported in a few papers (Yamamoto N et al. 2013 and Hosseini FA et al. 2011). This patient had a complex odontoma and cemento-ossifying fibroma. The origin of COF in this case is not identified, although earlier age of development of odontoma could suggest that COF is derived from odontomas. There is only three reports similar to our case until now; (Matsuo K. et al. 2013) reported multiple complex odontomas and subsequent occurrence of an ossifying fibroma in a 3 year-old boy; In this report the patient was followed for years and OF occurred after odontoma.

There are some papers which reports simultaneous occurrence or occurrence in different time of two or more central odontogenic lesions in the jaw (Matsuo K et al. 2013 and Ohtake K et al. 1993). It suggests the existence of a local environmental condition such as trauma, growth factor that permits the growth of multiple odontogenic lesions. Also occurrence of two lesion developed close to each other in the fairly limited area of alveolar bone suggests the possibility of a local environment permissive for the growth of multiple odontogenic tumors. It would be proposed to research the gene expression in this local environment and the genetic background of patients with like lesions (Hosseini FA et al. 2011).

## CONCLUSION

The relationship between the occurrences of these two lesions is not clear and more studies are needed to establish the relationship between them.

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