



## FOCAL CEMENTO-OSSEOUS DYSPLASIA: AN UNUSUAL CASE REPORT WITH REVIEW

### Dental Science

<b>Dr. Shah J.S.*</b>	M.D.S in Oral Medicine & Radiology, Professor & Head, Oral Medicine & Radiology, Govt. Dental College & Hospital, Ahmedabad-16, Gujarat, India *Corresponding Author
<b>Dr. Parikh S.J.</b>	M.D.S in Oral Medicine & Radiology, Professor, Oral Medicine & Radiology, Govt. Dental College & Hospital, Ahmedabad-16, Gujarat, India
<b>Dr. Parmar P.R.</b>	Oral Medicine & Radiology, Govt. Dental College & Hospital, Ahmedabad-16, Gujarat, India
<b>Dr. Patel A.J.</b>	Oral Medicine & Radiology, Govt. Dental College & Hospital, Ahmedabad-16, Gujarat, India

### ABSTRACT

Focal cemento-osseous dysplasia (FCOD) is a benign fibro-osseous lesion which changes cancellous bone tissue with each fibrous tissue and cementum-like material, or both together. Typically, the lesion is asymptomatic and is detected on routine radiographic examination. The teeth that are related to the lesion are vital. Radiological appearance of FCOD is radiolucent, radio-opaque or mixed radiolucent- radio-opaque together. In this case report a 37-year-old male patient presented with mixed radiopaque lesion surrounded by radiolucent area between the apical region of 2<sup>nd</sup> premolar and the 1<sup>st</sup> molar on the right side of mandible. The lesion was totally asymptomatic and found accidentally on radiographic examination.

### KEYWORDS

Cone Beam Computed Tomography, Focal Cemento-osseous Dysplasia, Fibro Osseous Lesion

### Introduction & Review

Osseous dysplasia is defined as a reactional and non-neoplastic process developing in periapical tooth area and characterized by normal bone replacement by fibrous tissue and metaplastic bone. When such process occurs in the periapical area of mandible's anterior teeth, is so-called periapical cemento-osseous dysplasia.<sup>1</sup>

Periapical cemento-osseous dysplasia (PCOD) has been described as a reactive or dysplastic Fibro osseous lesion in the tooth-bearing area, presumably of periodontal ligament origin (Waldron 1993) or unknown etiology. The World Health Organization (WHO) in their Histological Typing of Odontogenic Tumors (1992) referred to PCOD as periapical cemental dysplasia (PCD) (Kramer et al. 1992) and classified PCD as a type of cement-osseous dysplasia under non-neoplastic bone lesions. Other names for PCOD found in the literature are: cementoma, periradicular cemental dysplasia, periradicular fibrous dysplasia, periradicular osteofibrosis (Eleazer et al. 2012), periapical fibrous dysplasia (Kramer et al. 1992) and periapical osteofibrosis (Falace & Cunningham 1984). In 1956, Hamilton B.G. Robinson described similar lesions as periapical osseous dysplasia (Robinson 1956).<sup>2</sup>

In 1934, Stafne found a prevalence of 0.24%. Neville *et al.* observed a prevalence of 5.9% of periapical cemento-osseous dysplasia in Black women. Vicci & Capelozza determined prevalence of periapical cemento-osseous dysplasia was 1.8%. In 2008, Pereira *et al* found a prevalence of 1% for this disease.<sup>1</sup>

The etiopathogenesis of COD is still not clear. Some authors agree on the concept that the proliferation of fibroblastic mesenchymal stem cells in the apical periodontal ligament which are considered as cementoblastic precursor stem cells leads to the formation of agglomerated mass whereas others agreed on the view that COD may arise from the remnants of the cementum left after tooth extraction.<sup>3</sup>

On the basis of radiographic site of presentation cemento-osseous dysplasia is classified into three types:<sup>4</sup>

1. Periapical type - typically surrounds the periapical region of the teeth and is generally bilateral (usually seen in mandibular anterior region);
2. Florid type - characterized by sclerotic symmetrical masses involving multiple areas or quadrants;
3. Focal type - generally found as a single lesion.

THOMA classified lesions of this type into 3 stages of development (an osteolytic stage, a cementoblastic stage and a mature, inactive stage) based on radiographic and histological findings. Radiographically, lesions are located at the apices of teeth and exhibit radiolucency in the osteolytic stage that resembles periapical inflammatory diseases. In the

cementoblastic stage, formation of hard tissue usually occurs in the center of radiolucent areas. These lesions then become radiopaque in a mature, inactive stage in accordance with increase of hard tissue. There appears to be a radiolucent border around radiopaque masses with no direct continuity between roots of the affected teeth. This may become a means for the differential diagnosis of PCD from a true cementoma.<sup>5</sup> On the basis of clinical findings COD is divided into sporadic and familial types. The sporadic cases have single site involvement; however, the familial forms appears to be inherited as an autosomal dominant trait with variable phenotypic expression which is characterized by more expansile lesions and tend to occur in the younger subjects.<sup>5</sup>

Irrespective of Variety, this lesion is usually asymptomatic and is often found accidentally during routine radiographic examination. The affected teeth are often vital in electrical and thermal pulp testing.<sup>5</sup>

The key points for this disease diagnosis, according to Brannon & Fowler are:

- Predilection for mid-age Black women;
- One or more circumscribed lesions (0.5 cm or shorter) at the periapical area of vital teeth;
- Painless non-expansive lesion located usually at mandible's anterior area;
- Radiographic characteristics can be radiolucency of mixed density (radiolucent with opacities), or opaque with a narrow radiolucent margin;
- Cellular fibrous stroma with lamellar osseous tissue and/or oval calcifications.<sup>1</sup>

Histopathology picture of an early lesion shows cellular fibrous tissue containing trabeculae of woven bone with cementum like calcification. With maturation, the ratio of fibrous tissue to mineralized materials are decreased and trabeculae become more curvilinear structures that progress to the radiopaque stage in which the cementum-like tissue coalesce to form large basophilic calcification with resting and reversal line. Lesions are composed of anastomosing bone trabeculae and layers of cementum like calcification embedded in fibroblastic background.<sup>3</sup>

The differential diagnosis of COD includes Paget's disease, chronic diffuse osteomyelitis, and Gardner's syndrome, enostosis or exostosis, odontome and odontogenic tumours, especially cemento ossifying fibroma.<sup>6</sup>

Due to the nature and evolution of this lesion, no treatment is necessary. Because the teeth remain vital, tooth extraction or endodontic treatment should not be performed. On the other hand,

regular following-up examinations are recommended comprising dental prophylaxis and oral hygiene instruction reinforcement to prevent periodontal disease and caries lesions which can lead to tooth loss.<sup>1</sup>

### Case report

A 37 year old male patient came to the oral medicine and radiology department of Govt. Dental College and Hospital, Ahmedabad with the Chief complaint of pain in mandibular right posterior tooth region since 2-3 months. Pain was dull, gnawing and continuous in nature. Past medical and dental history was not contributory except habit. Patient had habit of mawa chewing 5-7 pkts/day for about 10 years, he quit the habit since 3 years and presented with OSMF (Moderate stage).

The examination revealed reduced mouth opening (Approx 25mm) with blanching and fibrosis of mucosa. Vertical fibrous bands were palpated over bilateral buccal mucosa extending antero-posteriorly from pterygomandibular raphe to corner of mouth, supero-inferiorly from occlusal level to depth of vestibule. Mild Attrition was present in mandibular anterior teeth with generalized mild stain and plaque. Localized supragingival plaque and mild calculus were also present over mandibular 45, 46 and 47. Periodontal pockets were present distal to mandibular 45 and 46 interdentally. All teeth were vital (Cold, thermal and electrical pulp testing). Bilateral submandibular lymph nodes were not enlarged, not palpable and not tender. Based on these finding clinical diagnoses localized periodontitis with OSMF (Moderate stage) was made.

OPG and Intraoral periapical radiograph of mandibular right side 44, 45, and 46 revealed horizontal bone loss present interdentally between 45 and 46 with an accidental finding of Well-defined radiopaque mass size of 2\*1 cm approx. surrounded by radiolucency with corticated border present on apical region of 45, extending from distal root surface of 44 to mesial root of 46. There was mesial displacement of root portion of 45 was present. Multiple ill-defined radiolucency with corticated border was present over mandibular anterior teeth extending from mesial aspect of 35 to distal aspect of 44. Cross Sectional Mandibular occlusal view revealed slight expansion of buccal cortical plate in relation with 45, 46. (Figure 1,2,3) Based on these finding radiographic diagnosis was Focal cemento-osseous dysplasia i.r.t 45, 46 was considered. (Figure 2) Complex odontome was considered in differential diagnosis.

To rule out extension and attachment of capsule patient was advised CT scan of maxillofacial region. With such type of peculiar presentation of this lesion a diagnostic dilemma prevailed in the mind of diagnosticians and radiologists, hence patient was subjected to CBCT. The lesion was easily and clearly distinguishable in CBCT images.

CBCT of maxillofacial region revealed evidence of well-defined lytic lesion involving body of mandible on right side measuring 18mm\*10 mm in size, with internal foci of calcification-mineralization with few septation which lies in relation to root of 2<sup>nd</sup> premolar. There was expansion of overlying cortex without cortical bone loss or irregularity present. The axial slice with slice thickness of 1.98mm revealed an oval mixed hypo and hyperdense area present in the 2<sup>nd</sup> premolar region, the border was sclerotic. (Figure 4) This finding was in favour of focal cement osseous dysplasia.

Considering all the above possibilities the lesion was subjected to biopsy along with extraction of 45. Histopathological evaluation revealed connective tissue mass with loose collagen fiber bundles and stellate shaped fibrocytes. Foci of osteoid and lamellar bone surrounded by extravasated RBCs. (Figure 5) With these findings a final diagnosis of focal cemento-ossifying fibroma was confirmed.

Patient is under observation for every 2 months and for further clinical and radiographic evaluation for Focal cemento-osseous dysplasia. As patient was diagnosed case of OSMF he was under the treatment of OSMF with topical application of Betamethasone [0.05%] and mouth opening exercise, systemic antioxidant and multivitamins therapy.

### Discussion

FCOD is predominantly seen in women, with a mean age in the mid-30s and a slightly higher incidence (64%) among African Americans. Most (62%) of these lesions are asymptomatic, and they can reach up to 1.5 cm in size.<sup>8</sup> The mandible is the most frequent site of occurrence

(86%). In the presented case FCOD was found in 37 year old male patient involving mandibular right premolar-molar area and was ~2 cm in diameter, which is consistent with the literature.

The etiology and pathogenesis of FCOD remain unknown, and this lesion is considered to be a reactive or dysplastic process in periapical tissues. Although the lesion is not considered to be odontogenic, it is undoubtedly related to the presence of teeth.<sup>9</sup>

Focal cemento osseous dysplasia typically presents as a focal, asymptomatic lesion with vital teeth diagnosed incidentally on routine radiography, predominantly in a single quadrant in the mandible, although multiple lesions can occur within the same quadrant.<sup>6,7</sup> Similar finding was noted in present case, However, MacDonald Jankowski has reported that pain and swelling was seen with rates of 25% and 28%, respectively. Su et al. have reported that some swelling and slight discomfort can be seen.<sup>7</sup>

Focal cemento-osseous dysplasia and periapical cemento-osseous dysplasia are two terms that state same pathologic processes in different localizations. Florid cemento-osseous dysplasia however, means the multifocal wide involvement of chins by the lesions with the same histological appearance of PCOD and FCOD.<sup>7</sup>

FCOD is usually found during routine radiographic examination. It is seen most frequently in the anterior and in the premolar areas of the mandible. Seventy percent of the FCOD cases display an intimate relationship to the periapex. The remaining 21% is found in the sites of previous extraction and may mimic a residual cyst.<sup>9</sup> Presented case shows relationship to the periapex of 2<sup>nd</sup> premolar. Almost all FCODs appear above the mandibular canal and are thus confined to the alveolar process, suggesting at least some Odontogenic influence on their genesis.<sup>9</sup> Presented case shows similar findings.

Radiologic images of FCOD change depending on the increase of mineralization. Well defined radiolucency can be observed in the early stage but it is not frequent. In the intermediate stage, mixed or full radiodensity with sclerotic borders is seen, and sometimes the characteristic radiolucent rim around the radio-opacity is observed. In this stage, semiluculent no rim poorly defined mixed radiodensity is observed in half of the cases. The last stage is more mature. In this stage, a sclerotic radiodensity with poorly defined borders is typical. The radio-opaque appearance of FCOD can be cotton wool-like irregular or diffuse.<sup>7</sup> The presented case was in accordance with the mixed stage; a poorly defined radio-opaque area surrounded by a radiolucent rim was observed. CBCT images may be preferable to observe the progression of the lesion.

During the formulation of the differential diagnosis, the developmental stage of the lesion should be considered, and the possibilities should include periapical granuloma or cyst and chronic osteomyelitis in the osteolytic stage. In the mixed and radiopaque stages, florid or periapical COD, chronic sclerosing osteomyelitis, ossifying/cementifying fibroma, odontome, osteoblastoma and ameloblastoma should all be considered.<sup>12</sup>

The focal type requires biopsy and Histopathological examination of the tissues for definitive diagnosis.<sup>13</sup> In the presented case, similar radiographic features of focal cemental dysplasia were seen. So, we had gone for Histopathological examination.

Histopathologically, FCOD is a heterogeneous lesion consisting of a benign fibrous stroma containing irregular trabeculae of mature and immature bone and cementum/osteoid-like material.<sup>8</sup> similar findings was seen in presented case.

Focal cemento-osseous dysplasia does not require surgical intervention as they are benign and their growth is limited. It is recommended that these lesions should be followed regularly, because they can turn into florid osseous dysplasia, which is the advanced form of dysplasia, or simple bony cysts can developed within the FCOD areas.<sup>7</sup> In present case, since the lesion was asymptomatic, and not causing any destruction of bone or root resorption of tooth, patient kept under regular follow-up. As patient was diagnosed as OSMF, topical application of betamethasone [0.05%] and mouth opening exercise, systemic antioxidant and multivitamins therapy was recommended.

### Conclusion

Periapical pathology presents as a wide spectrum of lesions that may

mimic and masquerade each other. This case highlights the importance of careful clinical examination, having an unbiased view of age, gender, and ethnicity when differentiation of these conditions. In such conditions of severe diagnostic dilemma CBCT can be considered to be more effective in providing the intricate anatomical details of the lesion and providing good information for better diagnosis and prognosis which also helps to avoid risk of failure of osseointegration with dental implant.

#### Legend Of Figure:

**Figure 1:** Intraoral periapical radiograph showing a wellcorticated radioopaque lesion surrounded by radiolucency present in the 45 region

**Figure 2:** Orthopantomograph shows a wellcorticated radioopaque lesion surrounded by radiolucency present in the 45 region.

**Figure 3:** Occlusal view of mandible shows buccal expansion i.r.w 45 and 46.

**Figure 4:** CBCT shows mixed hypo and hyperdense area present in the 45 region.

**Figure 5:** Shows connective tissue mass with loose collagen fiber bundles and stellate shaped fibrocytes. Foci of osteoid and lamellar bone surrounded by extravasated RBCs are also seen. (HE stains 10x).



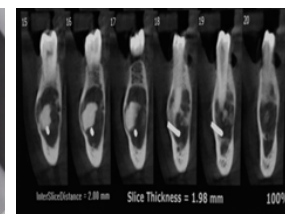
**Figure 1:** Intraoral periapical radiograph showing a wellcorticated radioopaque lesion surrounded by radiolucency present in the 45 region



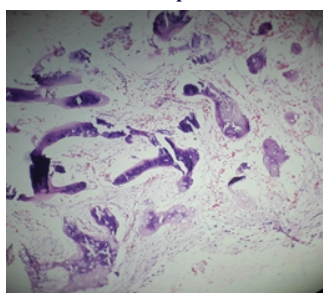
**Figure 2:** Orthopantomograph shows a wellcorticated radioopaque lesion surrounded by radiolucency present in the 45 region



**Figure 3:** Occlusal view of mandible shows buccal expansion i.r.w 45 and 46.



**Figure 4:** CBCT shows mixed hypo and hyperdense area present in the 45 region.



**Figure 5:** Shows connective tissue mass with loose collagen fiber bundles and stellate shaped fibrocytes. Foci of osteoid and lamellar bone surrounded by extravasated RBCs are also seen. (HE stains 10x).

#### REFERENCES

- Sumie Morikawa, F., Onuki, L. Y., Lima Chaiben, C., Martins Tommasi, M. H., Vieira, I., & Soares de Lima, A. A. (2012). Periapical cemento-osseous dysplasia: case report. RSBO Revista Sul-Brasileira de Odontologia, 9(1).
- Senia, E. S., & Sarao, M. S. (2015). Periapical cemento-osseous dysplasia: a case report with twelve-year follow-up and review of literature. International endodontic journal, 48(11), 1086-1099.
- Pandya, D. P., Varshney, A., Pandya, P. D., & Shirke, P. D. Fibrous Dysplasia of the Mandible: A Case Report. Fibrous Dysplasia, 6.
- Chennoju, S. K., Pachigolla, R., Govada, V. M., ALAPATI, S., & Balla, S. (2016). Idiosyncratic Presentation of Cemento-Osseous Dysplasia-An in Depth Analysis Using Cone Beam Computed Tomography. Journal of clinical and diagnostic research: JCDR, 10(5), ZD08.
- Tanaka, H., Yoshimoto, A., Toyama, Y., Iwase, T., Hayasaka, N., & Moro, I. (1987).

Periapical cemento-osseous dysplasia with multiple lesions. International journal of oral and maxillofacial surgery, 16(6), 757-763.

- Potochny, E. M., & Huber, A. R. (2011). Focal osseous dysplasia. Head and neck pathology, 5(3), 265-267.
- Yoganandha, R. (2016). Multifocal Polyostotic Craniofacial Fibrous Dysplasia. IJHRMLP, 2(01), 127.
- Cankaya, A. B., Erdem, M. A., Olgac, V., & Firat, D. R. (2012). Focal cemento-osseous dysplasia of mandible. BMJ case reports, 2012, bcr2012006432.
- Bhandari, R., Sandhu, S. V., Bansal, H., Behl, R., & Bhullar, R. K. (2012). Focal cemento-osseous dysplasia masquerading as a residual cyst. Contemporary clinical dentistry, 3(Suppl1), S60.
- Eskandarloo, A., & Yousefi, F. (2013). CBCT findings of periapical cemento-osseous dysplasia: a case report. Imaging science in dentistry, 43(3), 215-218.
- Nascimento, R. N. D. M., Cavalcante, L. H. D. A., Felix, A. M. L. F., De Melo, D. P., Pereira, J. V., Diniz, C. N., & Diniz, D. N. (2015). Periapical Cemental Dysplasia: Case Report. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology, 120(2), e38.
- Alsufyani, N. A., & Lam, E. W. (2011). Osseous (cemento-osseous) dysplasia of the jaws: clinical and radiographic analysis. J Can Dent Assoc, 77, b70.
- Burket, L. W., Greenberg, M. S., & Glick, M. (2003). Burket's oral medicine: diagnosis & treatment. PMPH-USA.