

# PERIAPICAL, FOCAL AND FLORID CEMENTO-OSSEOUS DYSPLASIA: A SYSTEMATIC REVIEW FROM INDIAN POPULATION

Simi Thankappan<sup>1</sup>, Sherin Nedumpillil<sup>2</sup>, Ravi Seth<sup>3</sup>, Sunita Pathak<sup>3</sup>

<sup>1</sup>Geetanjali Dental and Research Institute, Udaipur, Rajasthan.

E-mail: [tsieolin@gmail.com](mailto:tsieolin@gmail.com)

<sup>2</sup>Government Dental College, Dibrugarh, Assam

<sup>3</sup>Rama Dental College Hospital & Research Centre, Rama University, Mandhana, Kanpur, Uttar Pradesh- India 209217

## ABSTRACT

Cemento-osseous dysplasia (COD) is an uncommon lesion in Indian population. For evaluating this rare lesion, a systematic literature review to provide an up-to-date summary of cemento-osseous dysplasia from the Indian perspective was carried out. The WHO classification for COD like “periapical”, “focal” and “florid” cemento-osseous dysplasia was the main terms used. Electronic databases like Medline, EBSCO, Scopus and "Web of Science" were used for the searches. The search revealed a total of 48 articles, out of which 37 were only included. This included twenty nine case reports and eight case series. Of the total 56 patients, from all the case reports and series, of which 47 were females and only nine, were males. Age ranged from 13-64 years. Florid COD was the commonest type of COD among the articles with 42 cases. Majority of the florid cases involved both maxilla and mandible, and involvement of maxilla was rare in focal and periapical varieties. To our knowledge, this is the first systematic review of cemento-osseous dysplasia in Indian population.

**Key words:** Periapical cementoma, Focal Cemento-Osseous Dysplasia, Florid Cemento-Osseous Dysplasia, Indian, Radiolucent Rim, Mixed Radiolucencies, CBCT, Systematic Review

## Introduction

Cemento-osseous dysplasia (COD) is an uncommon lesion in Indian population, but also the most common fibro-osseous lesion that is reported in Indian patients. COD, a benign bony lesion arising from undifferentiated cells of the periodontal ligament tissues, is characterized by normal bone replaced by fibrous tissue, and later calcification with osseous and cementum-like tissue. COD is classified by WHO into 3 categories according to its location: periapical (associated with periapical region in anterior teeth, focal (associated with a single tooth), and florid lesions appearing in more than one quadrant.<sup>1,2</sup> The principal features of COD from the Indian population have been evaluated in this review, since there are no systematic reviews on COD from the Indian population.

## Materials and methods

A literature review was carried to provide an up-to-date summary of COD in Indian population. Only case reports and series were considered eligible for inclusion in this review, if they were published during the past 21 years (period of publication from January 1, 2000 to October 31, 2021). All included studies were published in English. To ensure study quality, only those published in national or international peer-reviewed journals were considered. Study identification and data extraction were performed by searching the scientific literature databases like Medline, EBSCO, Scopus and "Web of Science". The following search terms were used: "periapical", "focal" and "florid" cemento-osseous dysplasia, "cementoma", periapical cemental dysplasia, periapical osseous dysplasia, focal cemento-osseous dysplasia, periapical cementoma. In addition, relevant cross-references references and bibliographies were manually searched by trained researcher for additional case reports.

From each of the identified studies, the following information was extracted: age and gender of the patient, site of jaw affected, type of COD (periapical COD, focal and florid-WHO criteria), the radiographic stage at the time of diagnosis (first stage/osteolytic stage, second stage/cementoblastic stage, final /mature stage), and the findings were tabulated. Other findings like symptoms of swelling, pain and pus discharge, presence of cortical expansion, radiographic features, presence of secondary infection and co-existing other lesions, investigations, vitality of teeth, CT and CBCT findings in addition to conventional imaging findings, biopsy and treatment were also recorded.

## Results

Forty eight articles were obtained from the literature review, published over a period of 20 years. (Table 1) Majority of the reports were from peer-reviewed indexed journals like Medline. The search term "Osseous Dysplasia" was the most effective for all databases. From 48 articles, 37 articles were included of which 29 were case reports and eight were case series. Eleven case reports were omitted due to publication in non-indexed journals. Articles appearing only in Google scholar or index Copernicus, journals with e-mail submissions, and predatory suspicious sites were excluded. Seven cases were florid Cods or focal CODs, and not PCODs. One case of florid COD was retracted by a journal, so was not included in the review. All the COD cases are summarized in Table no.3. <sup>3-39</sup>

There were 56 patients in total from all the case reports and series, published during the period of 2000-2021. Of these 56 patients, 47 were females (84%) and only 9 were males (16%). The female to male ratio was 1:5. The age ranged from 13 to 65 years, the average age being 39 years. The majority in the 20-29 age group (22%) and rarely occurring under 20 years (6.7%). (Table no.2)

Florid COD was the commonest type of COD among the articles with 42 cases (75%). Twelve patients had focal COD (21%) and only two 2 cases of PCOD was reported (3.6%). Majority of the florid cases involved both maxilla and mandible (all quadrants; 24%). Involvement of bilateral maxilla only, was nonexistent in these reports, whereas, involvement of mandible

bilaterally was 28%. Focal cases in mandible (15%) were also more common than in maxilla (11%), especially focal and periapical COD.

Many patients came with caries and toothache for which radiographs were taken, but majority of the patients had symptoms (82%). Most of the focal COD lesions were asymptomatic, unlike florid COD cases that reported with swelling because of the involvement of multiple arches. Pain or discomfort was the most frequent symptom for many patients, with or without swelling. Some of the patients with pain and swelling had pain and pus discharge and were diagnosed with osteomyelitis (12.5%).

Second stage of COD was noticed commonly than the first stage (Stage 1, 7%; Stage 2, 28%). Third stage was the majority with 50% of patients reporting in late stages of the pathology. Some cases were in different stages of maturation and couldn't be included in any stages (14%).

### Discussion

Cemento-Osseous Dysplasia (COD) is an uncommon lesion unique to the jaw bones. Depending on the site of involvement, they are classified into three types, periapical, focal and florid COD (Fig. no. 1) The cause for the pathology was largely unknown, and was thought to be a fibro-osseous lesion arising from the periodontal tissues, and recent studies have shown some light regarding the molecular pathogenesis of the diseases, which can involve a combination of dysregulation of Wnt and *NOTCH* pathways, overactive Wnt pathway with enhanced CTNNB1 expression, spontaneous growth arrest, recurrent signaling alterations and mutations in RAS-MAPK pathway genes, endocrine stimulation that can cause the diseases more commonly in females such as potential hormonal stimulus, possibly estrogen.(Fig.no.2)

Of these 56 patients reported, the majority were females, which has been reported as the gender commonly affected.<sup>5,13,22,33,34</sup> Interestingly, five of the nine males were reported in a single case series, and age ranged from 13-64 years. Even though most of the case series reported clear female predilection, only one case reported an equal sex predilection. In the same series, the individual case details were not elaborated.<sup>34</sup>

Florid COD was the commonest type of COD in our review,<sup>3-8,10,11,13,14,16,18-20,22-25,29-32, 34,35,37-39</sup> followed by 12 patients of focal COD,<sup>9,12,15,17,21,26,28,33, 34,36</sup> and only two 2 cases of PCOD.<sup>27,34</sup> Two cases with family history was diagnosed as florid familial COD.<sup>13,25</sup> There was a case of focal COD involving mandible 44-46 region, but diagnosis was given as florid.<sup>20</sup>

Most of the *focal* COD lesions were asymptomatic, since they were restricted to the periapical region of a single tooth, and were discovered as an incidental finding when patient had other conditions seeking dental consultation.<sup>3, 4,5, 18,19,20, 21,29,30,33</sup> Most of the *florid* COD cases reported with swelling because of the involvement of multiple arches, or because of the mature stage they are usually shows manifestations.<sup>5,6,7,8,9,11,12,13,15,20,22,23,24,25,28,29,32,34,37</sup> A swelling of 10 years duration with significant facial asymmetry was reported in a patient who was diagnosed with florid COD.<sup>8</sup> Periapical cemento-osseous dysplasia (PCOD) is a characteristic lesion occurring usually in anterior mandible of patients older than 30 years of age, female patients, with involved teeth vital, and mostly having a racial predilection, affecting blacks. Our review from Indian population had only two cases of PCOD.

Pain or discomfort was a frequent symptom for many patients with or without swelling.<sup>5,9,10,11,12,14,15,22,25,26,29,30,36,37,39</sup> Some of the patients with pain had pus discharge<sup>7,16,17,23,24,25,29,35</sup> and many were diagnosed with osteomyelitis.<sup>7,13,16,17,23,31</sup> Dull, continuous pain in healed extraction sites or non healing extraction socket were symptoms for focal and florid CODs.<sup>10,16,31,36</sup> Other complaints reported were spacing and mal-positioned teeth<sup>13</sup> and numbness.<sup>38</sup> Vitality of adjacent teeth in area of lesion was mentioned in majority of reports.<sup>3,5,6,8,12,17,19,20,21,23,27,29,33,37</sup> In cases where adjacent teeth were non-vital, it was due to a carious lesion near the COD and not due to the direct involvement by COD.<sup>5</sup> Majority of the florid cases involved both maxilla and mandible, whereas involvement of maxilla alone in cases like focal and periapical were very few.<sup>9,15,25,27,28</sup> When in maxilla, the lesions were almost in posterior regions, and encroached the maxillary sinus in some cases,<sup>15,24</sup> but not all.<sup>28</sup> Most of the lesions that were involved multiple areas, and diagnosed as florid COD and presented as swelling clinically, had bucco-lingual cortical expansion.<sup>10,13,14,15,20,22,24,25, 28,30,35</sup> Even though radiographic stages for COD are mentioned along with their clinical types, most of the cases didn't name their stages. In those cases, we evaluated the radiographs and included the lesion accordingly, confirmed by a third researcher. Lesions when diagnosed in the 1<sup>st</sup> stage were typical periapical radiolucencies that mimicked periapical granulomas or cysts, but mostly the teeth were normal and vital.<sup>3,12</sup> Second stage of COD was noticed commonly than the first stage.<sup>4,7,8,9,11,15,18,20,29,33,37</sup> The third stage or maturation stage can have different radiographic appearances despite being completely radio-opaque. Cases described lesions as 'sclerotic' and 'radiopaque masses', 'patchy', 'globular', 'discrete', 'confluent', 'dense' 'lobulated' and 'lobular' radiopacities.<sup>6,7,14,15,16,19,21,22,23,24,26,28,30,35,39</sup> In florid COD, the various lesions were in different stages of radiolucencies and radio-opacities, so it was easier to categorize them under 'various stages of maturation.'<sup>10,13,17,18,19,23,24,25,34,35</sup> Almost all the lesions were having radiolucent rim, and most lesions were having well defined borders even though irregular in shape.<sup>3,5,10,14,18- 22,25, 30,32-36,39</sup> Some stage three lesions were described as cotton-wool appearance,<sup>14,30</sup> ground glass appearance,<sup>23</sup> and scalloped borders around radiolucent lesion.<sup>10</sup> Some lesions were with ill-defined borders<sup>14,17,20,24,25,29</sup> and moth eaten appearance.<sup>16,29,31,35</sup> Some authors have reported an 'atypical' PCOD in maxillary premolar region, presented entirely as a palatal swelling. Other radiographic findings in other case reports were hypercementosis,<sup>29,34</sup> no direct attachment of pathologic tissue to root cementum,<sup>7,19,28</sup> attached to the root apex,<sup>37,33,21</sup> displaced and impacted teeth,<sup>15</sup> and associated with other pathologies like dentigerous cyst<sup>8</sup> and simple bone cyst.<sup>10</sup> CT and CBCT scans described the COD lesions as 'patchy', well defined 'hyperdense' or 'hypodense' lesions with a prominent hypodense area encapsulating the lesion.<sup>10,13,14,23,27-30,35, 38, 39</sup> Routine haematological investigations and biochemical analysis of serum calcium, phosphorus and alkaline phosphatase showed values within normal limits in almost all case reports.<sup>6,8- 10,13,14,16,19,20,22,23,28-31,35,37</sup> Increased alkaline phosphatase levels was reported by Choudhary SH et al.<sup>32</sup> Majority of the lesions were diagnosed by clinical and radiographic features alone,

especially the stage of lesion, and symmetrical lesions bilaterally in jaws in FCOD.<sup>6,7,29,13,19,30</sup> In some cases authors went for biopsy and histopathological confirmation.<sup>6,7,9,12,22,25,27,35,37</sup> Some lesions which were discovered during radiographic examination and were asymptomatic, were not treated.<sup>3,22,25,29,30,39</sup> Some lesions didn't change the size even after two years, stressing on the fact that all cases of COD doesn't need to be treated and needs only follow-up.<sup>5</sup> Rest of the lesions were surgically excised with sequestrectomy and debridement under local anesthesia<sup>6,8,10,13,15,16, 26-29, 32,37,38</sup>

MacDonald-Jankowski DS was the first one to do an exhaustive systematic review in florid and focal CODs. They observed 158 cases of FCOD in 17 case series, in which the racial distribution was majority in Blacks (59%), Orientals (37%) and Caucasians (3%). Most of the literature has identified prevalence among blacks and in Asian races. Like in our review, 97% patients were females. Pain was most frequent in those with presenting symptoms, and in our review, it was swelling followed by pain..<sup>40</sup>

Of the 20 series considered for focal CODs by the same author, Focal COD predominantly affected females and the common site was mandible, like our review.<sup>41</sup>

To our knowledge, this is the first systematic review of cemento-osseous dysplasia in Indian population. While PCOD and focal COD are asymptomatic and discovered as an incidental finding while taking radiographs, florid CODs present with swelling in most patients due to the duration and maturity of the lesions.

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**Table 1: Demographic and Clinical features of COD**

Variable	No. of patients (%)	Variable	No. of patients (%)
<b>Gender</b>	<b>n=56</b>	<b>Symptoms</b>	<b>n=56</b>
Female	47 (84%)	Asymptomatic	10 (17.9%)
Male	9 (16%)	Symptomatic	46 (82%)
		Symptomatic with OML	7 (12.5%)
<b>Age (year)</b>	<b>n=45*</b>	<b>Location</b>	<b>n=46*</b>
<20	3 (6.7%)	Maxilla,Focal	5 (10.9%)
20-29	10 (22%)	Maxilla, Bilateral	0
30-39	8 (17.8%)	Mandible,Bilateral	13(28.3%)
40-49	9 (20%)	Mandible,Focal	7 (15.2%)
50-59	8 (17.8%)	All Quadrants	21 (24%)
60-69	7 (15.6%)		
>70	0		
<b>Stage of diagnosis</b>	<b>n=56</b>	<b>Type</b>	<b>n=56</b>
Stage 1	4 (7.1%)	PCOD	2 (3.6%)
Stage 2	16 (28.6%)	Focal COD	12 (21%)
Stage 3	28 (50%)	Florid COD	42 (75%)
Various stages of maturation	8 (14.3%)		

Over expression of Beta catenin Parafunctional habits  
 Some cases excluded for estimation, as there was no description of same in case studies

