Original Article

Cemento-osseous dysplasia: Clinicopathological spectrum of 10 cases analyzed in a tertiary dental institute

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Abstract

Background: Cemento-osseous dysplasia (COD) is a distinct entity, which is confined to the tooth-bearing areas of the jaws or edentulous alveolar processes.

Aims: This study analyzes the demographic, clinical, radiographic and histopathological features of COD.

Materials and Methods: Archival data from 2013 to 2017 present in the Department of Oral Pathology were retrieved. Clinicopathological and radiological features of all the cases which had been previously diagnosed as COD were analyzed in detail.

Results: In the present study, 10 cases of COD were analyzed (1 periapical, 6 florid and 3 focal). Equal sex predilection was observed. The mean age in females and males was 42.6 years (22–64 years) and 28.6 years (17–36 years), respectively. Bony expansion was seen frequently, but pain was not a common feature. Mandible was affected almost twice as frequently as maxilla and posterior region was affected more than anterior region. Most cases showed mixed radiopaque and radiolucent lesion (5/10) and peripheral radiolucent rim (9/10) on radiographic examination. Histopathological features included the presence of curvilinear trabeculae (4/10), sheets of compact bone (3/10) or both (3/10). Foci of mineralization in form of ossicles or cementicles were noted in 5 out of 10 cases.

Conclusion: COD is a nonneoplastic process usually confined to the tooth-bearing areas of the jaws or edentulous alveolar processes. Depending on the location and extent, it can be either focal/periapical/florid variant. Histopathological features of COD can overlap with other fibro-osseous lesions. Hence, the correlation of clinical, radiological and histopathological features is of paramount importance in the accurate diagnosis of COD.

Keywords: Cemento-osseous dysplasia, fibro-osseous lesions, florid, focal cemento-osseous dysplasia

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INTRODUCTION

The benign fibro-osseous lesions of the jaws comprise a heterogeneous group of conditions which are characterized by replacement of normal bone by fibrous tissue. These may contain variable amounts of mineralized material, which may resemble bone or cementum.[1,2] Cemento-osseous dysplasia (COD) is a distinct entity within this group, which is confined to the tooth-bearing areas of the jaws or edentulous alveolar processes.[3] A paucity of calcified material is exhibited by immature lesions, whereas more

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mature lesions exhibit development and coalescence of dense, globular masses of calcified material.[4] Diagnosis of COD cannot be solely based on microscopic features as histopathological appearances of fibro-osseous lesions are quite similar, if not identical.[5-8] The lesions are usually innocuous and often discovered on routine radiographs; however, larger lesions causing significant cortical expansion, displacement of teeth and encroaching the maxillary sinus have also been reported. Although they are frequently asymptomatic, the association with simple bone cysts and complications related to secondary infection or osteomyelitis may further complicate the management.[4] Misdiagnosis of COD may lead to initiation of unnecessary endodontic and surgical treatment. The aim of the present study was to analyze clinical, radiological and histopathological features of ten cases of COD, which may aid in its diagnosis.

MATERIALS AND METHODS

Archival data of the cases which were histopathologically diagnosed as COD were retrieved from 2013 to 2017. Ten cases for which all relevant clinical, radiological and histopathological data were available were selected. Clinical features of age, sex, site and clinical presentation of the lesion were analyzed along with key radiological features including the periphery (well/ill defined, cortication and radiolucent band), internal structure (mixed, radiopaque and radiolucent with radiopaque foci) and effect of lesion on the surrounding structures. The cases were divided as florid, focal and periapical variants depending on the location and number of sites involved.

The following histopathological features were analyzed on hematoxylin and eosin-stained sections and graded by three oral pathologists:

A. Bone pattern
B. Presence of ossicles and cementicles
C. Stromal amount
D. Stromal cellularity
E. Presence of areas of haemorrhage.

The bone pattern included the presence of either curvilinear trabeculae, sheets of compact bone or both. Stromal amount and cellularity were graded as less, moderate and high.

To assess the vascularity of the lesion, immunohistochemistry (IHC) was performed using ready-to-use mouse monoclonal CD34 primary antibody (cell Marque, California, U.S.), which stained the blood vessels. The IHC procedure was followed according to the manufacturer's instructions.

Follow-up data were available for all ten cases. The data were tabulated and analyzed in detail.

RESULTS

Demographic data and clinical features

Of the ten cases studied, five females and five males were affected. Age of the patients ranged from 17 to 64 years (mean age, 35.6 years). Sixty-one percent of the total lesions involved the mandible (16/26), especially the posterior region. Swelling was observed in majority of the cases (9/10) and was associated with pus discharge in two cases. Pain was not a frequent symptom. One case each showed tooth displacement and extension of the lesion into the maxillary sinus.

Radiographic features

Radiographic analysis revealed that the lesions were well defined with most of the cases (8/10) having irregular boundary. A characteristic radiolucent rim or band was observed (9/10) with absence of cortication (6/10). COD presented as either radiolucent lesion with radiopaque foci (2/10), radiopaque (3/10) or mixed lesion (5/10) on the radiographs. Other features such as root scalloping (3/10), absence of lamina dura (8/10), hypercementosis (5/10) and absence of root resorption (10/10) were also observed [Table 1 and Figure 1].

Gross and histological features

The gross specimens which were received consisted of irregular gritty hemorrhagic fragments of tissue appearing grayish brown in color.

On microscopic examination, cases showed the presence of short curvilinear trabeculae (4/10), solid sheets of compact bone with minimal stroma (3/10) or admixture of both (3/10). The presence of calcified material as ossicles

Table 1: Radiological features of cemento-osseous dysplasia

Features	*n*=10(%)
1. Periphery	
Well defined	8 (80)
Irregular shape	8 (80)
Radiolucent rim/band	9 (90)
No cortication	6 (60)
2. Internal structure	
Mixed	5 (50)
RO	3 (30)
RL with foci of opacification	2 (20)
3. Effects on surrounding structures	
Root scalloping	3 (30)
No lamina dura	8 (80)
Normal PDL space	4 (40)
No root resorption	10 (100)
Hypercementosis	5 (50)
Cortical expansion	9 (90)
Displacement of anatomic structures	2 (20)

RO: Radiopaque, RL: Radiolucent, PDL: Periodontal Ligament
n=10 (%)

and cementicles (5/10) and areas of free hemorrhage were also observed (6/10) [Table 2 and Figure 2].

Moderate-to-high vascularity with blood vessels having larger perimeter was noted upon analyzing CD34-stained sections which had radiolucent lesions. The five cases which had mixed lesions having sclerosis exhibited moderate number of vascular channels within the areas having trabecular arrangement or droplets of cementum; however, areas which exhibited sheet-like arrangement showed relatively less number of blood vessels having constricted lumen. In three radiopaque lesions, the vascularity was least having narrow and constricted vessels [Figures 3 and 4].

Follow-up and recurrence

All the cases have been under routine follow-up since the

Table 2: Histopathological features of cemento-osseous dysplasia

Features	*n* = 10 (%)
1. Bone pattern	
Curvilinear trabeculae	4 (40)
Sheets	3 (30)
Mixed	3 (30)
2. Ossicles or cementicles	
Present	5 (50)
Absent	5 (50)
3. Stromal amount	
Less	3 (30)
Moderate-high	7 (70)
4. Stromal cellularity	
Moderate	3 (30)
High	7 (70)
5. Vascularity	
Moderate-high	2 (20)
Moderate-low	5 (50)
Low	3 (30)
6. Areas of hemorrhage	
Present	6 (60)

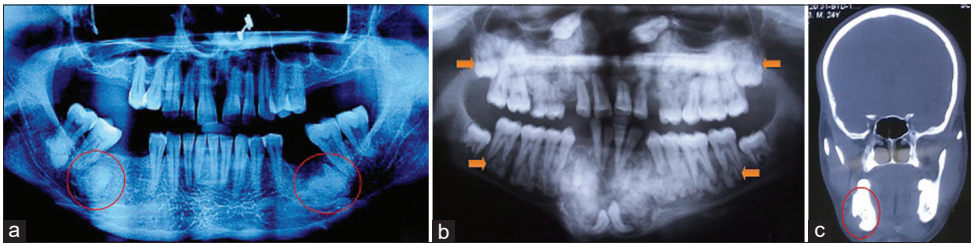

Figure 1: Radiographic features in cemento-osseous dysplasia. (a) Orthopantomography reveals well-defined, irregular radiopaque lesion surrounded by radiolucent rim in bilateral mandibular body region in a case of florid cemento-osseous dysplasia. (b) Orthopantomography of florid cemento-osseous dysplasia case reveals multiple mixed radiolucent-radiopaque lesions showing bilateral symmetry in the mandible and maxilla leading to displacement of all impacted canine teeth. (c) Computed tomography reveals radiopaque lesion in right body of mandible with disruption of buccal cortical plate

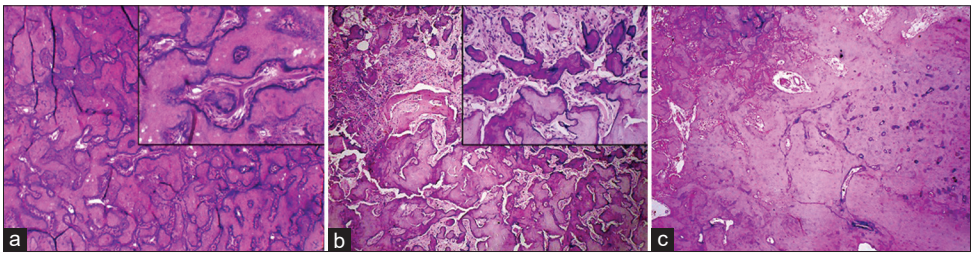

Figure 2: Histopathological patterns in cemento-osseous dysplasia (a) H&E-stained sections show intermingling short curvilinear trabeculae (×10: inset ×40), (b) H&E-stained sections show curved trabeculae and foci of mineralized cementum-like masses (×10: inset ×40), (c) H&E-stained section shows sheet-like arrangement of mineralized material (×10)

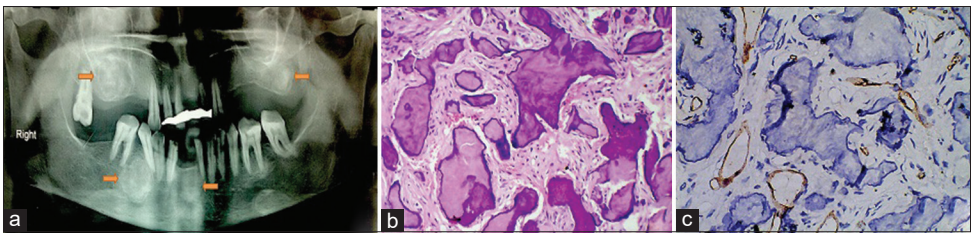

Figure 3: (a) A case of florid cemento-osseous dysplasia, presenting as well-defined mixed density lesion in right and left maxillary posterior region and more radiopaque lesions in anterior and right posterior mandible. (b) H&E section (×10) shows presence of mineralized material in form of small irregular trabeculae and ossicles/cementicles. (c) Immunohistochemistry with CD34 shows numerous blood capillaries within the stroma (×40)

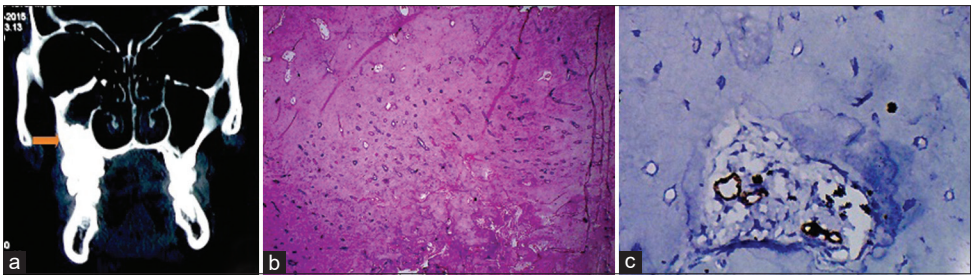

Figure 4: (a) A case of focal cemento-osseous dysplasia in advanced stage, presenting as well-defined sclerotic lesion in right maxillary posterior region with obliteration of right maxillary sinus. (b) H&E section (×10) shows sheets of bone with minimal marrow spaces. (c) Immunohistochemistry with CD34 shows few constricted blood capillaries (×40)

time of diagnosis till date. The follow-up period ranges from minimum of 2 years to maximum of 6.5 years. None of the cases have shown any signs of recurrence till date.

DISCUSSION

COD is a nonneoplastic fibro-osseous lesion which occurs exclusively in the tooth-bearing areas of the jaws or edentulous alveolar processes.[3] The term COD was adopted by the World Health Organization (WHO) in 1992. CODs were subclassified into periapical, florid and other CODs.[9] In 2005, the WHO renamed the group as osseous dysplasias. This classification recognized four lesions: periapical osseous dysplasia, focal osseous dysplasia, florid osseous dysplasia and familial gigantiform cementoma.[10] Eversole *et al*.[11] suggested that periapical COD [PCOD] and focal COD [FCOD] were two different terms for the same reactive lesion based on the anatomic location. PCOD involves the periapical areas of one or more vital teeth, predominantly mandibular incisors, while FCOD is found in tooth-bearing areas of the posterior jaws. Florid COD (FlCOD) was termed by Melrose *et al*.,[12] and it is characterized by extensive involvement of jaws seen in two or more quadrants.[11] The most common presentation is bilateral involvement of mandible with or without collateral involvement of maxilla. More than 90% of the cases have been reported in middle-aged Black women. However, these may exhibit a wide age range (27–79 years) with a peak incidence in the 4th–5th decade.[4,13,14]

The current study also showed similar age distribution, but no sex predilection was observed. Mandibular involvement was predominant, with posterior mandible being the most frequent site as reported by Pereira *et al*.[13] According to previous reports, CODs are usually asymptomatic and incidentally diagnosed on routine radiographic examination.[1,15] However, in our study, well-defined cortical expansion was noticed in almost all cases, although pain was not a common finding. Two patients complained of intraoral pus discharge, which has been previously reported in some cases.[12] Associated tooth in cases of COD is usually vital.

The radiological appearances of COD may vary from completely radiolucent in the early lesions to radiopaque in advanced lesions. Intermediate stage of COD usually presents as mixed radiolucent–radiopaque lesions on radiographs.[13,15] Majority of the cases in our study showed mixed radiolucent–radiopaque lesions as reported by Alsufyani and Lam.[4] COD lesions are usually well delineated from the surrounding bone. The presence of a characteristic surrounding radiolucent rim has been reported by many authors as a distinguishing feature in COD[1,4,6] Interestingly, we also found the radiolucent band or rim to be present in almost all cases except one. Narrowing of periodontal ligament space was uncommon, but intact lamina dura could not be appreciated in most of the cases. Root resorption and displacement of surrounding anatomical structures is not a common feature of COD.[4] These lesions usually have a limited growth potential. However, few of our cases showed extensive involvement of jaws on radiograph and one case showed disruption of cortex as well [Figure 1c]. One case was of FCOD associated with a history of trauma, which showed involvement of the right posterior maxilla showing extension of lesion into maxillary sinus and obliteration of sinus space on computed tomography [Figure 4a]. Another case was of FlCOD that showed displacement of impacted canines in all the four quadrants of the jaws [Figure 1b].

Histologically, CODs exhibit mesenchymal tissue of variable cellularity consisting of spindle shaped fibroblasts, collagen fibers and blood vessels. There are intermixed areas of bone and cementum-like material present as irregular coalescing trabeculae and ossicles/cementicles[1,6] [Figure 2]. Hemorrhage is commonly seen especially near the periphery.[6] This may be attributed to the vascular fragility in the altered connective tissue present in COD. Melrose *et al*.[12] described solitary bone cyst to be commonly associated with COD. Similar association was reported by Mahomed *et al*.[3] and Chadwick *et al*.[16] Furthermore, osteomyelitis frequently develops in association with COD, especially if following trauma or periapical pathology is present. In our series, none of the

cases were found to be histologically associated with these lesions. The radiographic appearance of the lesions correlated with the histological features. The cases which were showing either radiolucent lesions with foci of opacification or mixed lesions on radiograph exhibited abundant cellular stromal tissue, high vascularity and less to moderate amount of mineralized material in the form of short curvilinear trabeculae and ossicles/cementicles [Figure 3]. While the lesions which were radiopaque exhibited less amount of stromal tissue with decreased cellularity, minimal vascularity and acellular sheets of mineralized material Figure 4 (b) and (c).

The accurate diagnosis of COD is of paramount importance as it is a less aggressive lesion with self-limiting growth and is usually asymptomatic as opposed to other neoplastic lesions such as central ossifying fibroma and inflammatory lesions such as chronic osteomyelitis. FlCODs can be misdiagnosed as Paget's disease due to wide extent of the lesion, although COD is not usually associated with any biochemical abnormalities. Unlike other lesions, these usually do not require any intervention if the lesions are asymptomatic. Unnecessary interventions may cause complications such as pus discharge, fistula formation and sequestration of bone as the altered tissue in COD is usually avascular, especially in mature lesions.[2,5] Hence, it is best to keep patients under long term follow-up, even when the prognosis for these lesions is excellent.[2,6]

CONCLUSION

Through this study, we have attempted to add ten more cases to the literature with few cases showing extensive involvement of the jaws, which is not a common feature of COD. Hence, it can be concluded that the diagnosis of COD should be derived from analysis of the cumulative clinical, radiological and pathological data and should not be based on only one feature.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Waldron CA. Fibro-osseous lesions of the jaws. J Oral Maxillofac Surg 1985;43:249-62.
2. Waldron CA. Fibro-osseous lesions of the jaws. J Oral Maxillofac Surg 1993;51:828-35.
3. Mahomed F, Altini M, Meer S, Coleman H. Cemento-osseous dysplasia with associated simple bone cysts. J Oral Maxillofac Surg 2005;63:1549-54.
4. Alsufyani NA, Lam EW. Osseous (cemento-osseous) dysplasia of the jaws: Clinical and radiographic analysis. J Can Dent Assoc 2011;77:b70.
5. Brannon RB, Fowler CB. Benign fibro-osseous lesions: A review of current concepts. Adv Anat Pathol 2001;8:126-43.
6. Summerlin DJ, Tomich CE. Focal cemento-osseous dysplasia: A clinicopathologic study of 221 cases. Oral Surg Oral Med Oral Pathol 1994;78:611-20.
7. MacDonald-Jankowski DS. Florid cemento-osseous dysplasia: A systematic review. Dentomaxillofac Radiol 2003;32:141-9.
8. Su L, Weathers DR, Waldron CA. Distinguishing features of focal cemento-osseous dysplasia and cemento-ossifying fibromas. II. A clinical and radiologic spectrum of 316 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84:540-9.
9. Kramer IR, Pindborg JJ, Shear M. The WHO Histological Typing of Odontogenic Tumours. A commentary on the Second Edition. Cancer 1992;70:2988-94.
10. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization classification of tumours. In: Pathology and Genetics of Head and Neck Tumours. Lyon: IARC Press; 2005.
11. Eversole R, Su L, ElMofty S. Benign fibro-osseous lesions of the craniofacial complex. A review. Head Neck Pathol 2008;2:177-202.
12. Melrose RJ, Abrams AM, Mills BG. Florid osseous dysplasia. A clinical-pathologic study of thirty-four cases. Oral Surg Oral Med Oral Pathol 1976;41:62-82.
13. Pereira DL, Pires FR, Lopes MA, Carlos R, Wright JM, Patel P, *et al*. Clinical, demographic, and radiographic analysis of 82 patients affected by florid osseous dysplasia: An international collaborative study. Oral Surg Oral Med Oral Pathol Oral Radiol 2016;122:250-7.
14. Fenerty S, Shaw W, Verma R, Syed AB, Kuklani R, Yang J, *et al*. Florid cemento-osseous dysplasia: Review of an uncommon fibro-osseous lesion of the jaw with important clinical implications. Skeletal Radiol 2017;46:581-90.
15. Alsufyani NA, Lam EW. Cemento-osseous dysplasia of the Jaw Bones: Key Radiographic Features. Dentomaxillofac Radiol 2011;40:141-6.
16. Chadwick JW, Alsufyani NA, Lam EW. Clinical and radiographic features of solitary and cemento-osseous dysplasia-associated simple bone cysts. Dentomaxillofac Radiol 2011;40:230-5.