

# Idiosyncratic Presentation of Cemento-Osseous Dysplasia – An in Depth Analysis Using Cone Beam Computed Tomography

SAI KIRAN CHENNOJU<sup>1</sup>, RAMASWAMY PACHIGOLLA<sup>2</sup>, VANYA MAHITHA GOVADA<sup>3</sup>, SATISH ALAPATI<sup>4</sup>, SMITHA BALLA<sup>5</sup>

## ABSTRACT

Bone dysplasias comprise of a condition where the normal bone is replaced with fibrous tissue. Periapical Cemento-Osseous Dysplasia (PCOD) is a benign fibro-osseous condition where bone tissue is supplanted with fibrous tissue and cementum-like material. This condition affects mostly mandibular anterior region and rarely occurs in the maxilla. PCOD is seen above 30 years of age and has slight female predilection. Generally the teeth related to such lesions appear to be vital and are usually asymptomatic. These lesions are mostly seen during routine radiographic examination whose presentation may vary from complete radiolucency to dense radiopacity. The advent of Cone Beam Computed Tomography (CBCT) has brought a massive change in the field of dentistry which has become an important tool for diagnosis. Hence we hereby present an unusual case of cemento-osseous dysplasia in an unfamiliar location with an atypical presentation. The shape of the pathology was completely idiosyncratic and different from an orthodox lesion of COD, as the lesion was observed to grow out of the palatal surface with a prominent palatal expansion. This case highlights the importance of CBCT in radiographic diagnosis and in evaluating the characteristics of such lesion, which present with high diagnostic dilemma.

**Keywords:** Cementum, Fibro-osseous lesions, Maxilla, Palate, Periapical

## CASE REPORT

A 19-year-old female patient reported to the Department of Oral Medicine & Radiology with a chief complaint of swelling associated with pain in the upper right back tooth region from past six months. Swelling was initially of a peanut size, but gradually increased to the present size. There was no history of any discharge from the swelling. Her past medical history and dental history were noncontributory. Extra oral examination revealed no abnormality. Intra oral examination revealed a diffuse ovoid swelling of size approximately 2cm x 3cm in relation to 14, 15 palatally [Table/Fig-1a]. On inspection swelling was extending antero-posteriorly from 12 to distal aspect of 16, palatally and 0.5 cm away from the midline to the occlusal aspect of 14 and 15, medio-laterally [Table/Fig-1b]. No prominent buccal cortical expansion was evident. The lesion was confined completely in the palatal surface of maxilla. The mucosa over the swelling appeared normal without any secondary changes.

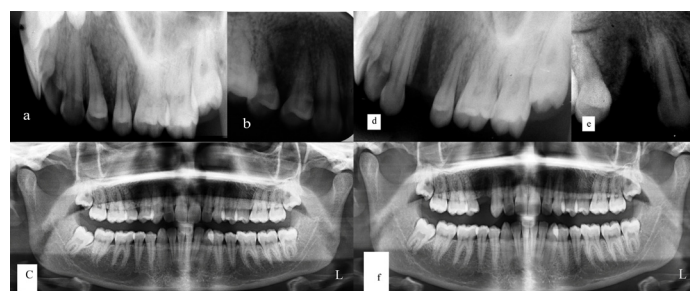


**[Table/Fig-1a-d]:** (a) A diffuse ovoid swelling of size approximately 2cmx3cm in relation to 14,15 palatally. (b) Swelling was extending antero-posteriorly from 12 to distal aspect of 16 palatally. (c) Histopathological picture showing a mixture of matured bone with osteocytes, fragments of cellular connective tissue containing cementoid and bone surrounded by hemorrhagic areas are evident. (d) The extracted premolar and tissue bits collected after excisional biopsy.

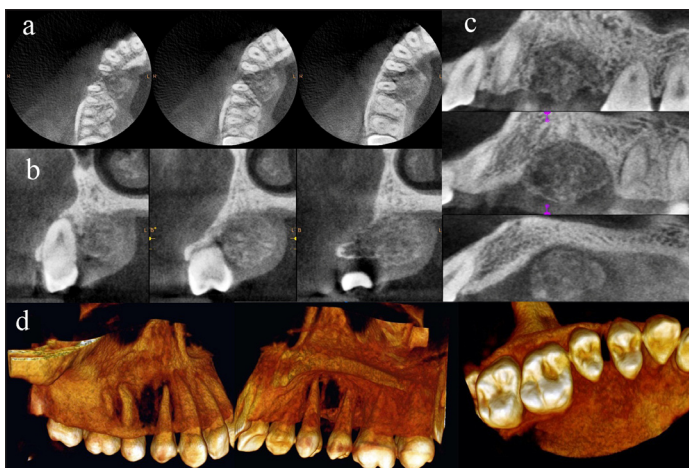
On palpation all the observed findings were confirmed. The swelling was firm in consistency and non-fluctuant. Mobility of teeth was observed in relation to 14 & 15. Aspiration was found to be negative. Pulp vitality test revealed vital pulps in relation to 14 & 15. Considering all the above features a provisional diagnosis of benign tumor in relation to 14 & 15 was given.

Patient was subjected to radiographic investigations which included the following: Intra oral periapical radiograph and lateral occlusal radiograph revealed mixed radio-opaque radiolucent lesion with ill-defined borders in the periapical region of 14 extending from the distal aspect of 13 to the mesial aspect of 15 [Table/Fig-2a, 2b]. There was slight distal displacement of the root portion of 14. But the lesion was observed to be very indistinct with ill-defined borders even in the panoramic radiograph [Table/Fig-2c]. With such type of peculiar presentation of this lesion, a diagnostic dilemma prevailed in the minds of diagnosticians and radiologists. Hence patient was subjected to cone beam volumetric imaging.

Surprisingly the lesion was easily and clearly distinguishable in CBCT images. The axial slices with slice thickness of 76 microns revealed an oval mixed hypo and hyper dense area of size 10.2mm X 10.7mm in the first and second premolar region with prominent



**[Table/Fig-2]:** (a-b) Intra oral periapical radiograph and lateral occlusal radiograph revealed mixed radio-opaque radiolucent lesion with ill-defined borders in the periapical region of 14 extending from the distal aspect of 13 to the mesial aspect of 15. (c) Lesion observed to be very indistinct with ill-defined borders even in the panoramic radiograph. (d-f) Post-operative radiographs after one month, showing healing socket.



**[Table/Fig-3a]:** The axial images showing an oval mixed hypo and hyper dense area in the first and second premolar region with prominent palatal expansion.

**[Table/Fig-3b,c]:** The trans-axial and custom slicing sections (curved planar images) showing well-defined hypodense border encircling the lesion and differentiating it from the normal bone. **[Table/Fig-3d]:** Three dimensional picture of the lesion on the buccal, palatal and occlusal aspect showing prominent bone loss.

palatal expansion. The borders were irregular and sclerotic. Loss of buccal cortical plate was evident in the interdental region of 14 & 15 [Table/Fig-3a]. Trans-axial sections with slice thickness of 76 microns revealed a prominent oval mixed hypo and hyperdense mass of size 10.6mm X 11.2mm, on the palatal surface of 14 & 15. The lesion was observed to be encroaching into the buccal cortical bone in the inter-radicular region of 14 & 15 [Table/Fig-3b]. The images obtained from custom slicing tool (curved planar images) were more interesting. The lesion in these images was more prominent with a well-defined hypodense border encircling it and differentiating it from the normal bone [Table/Fig-3c]. Three dimensional evaluation of the lesion were presented in [Table/Fig-3d]. These radiographic findings revealed the exhaustive involvement of the lesion within the bone and confirmed the aggressiveness of the lesion.

Considering all the above possibilities the lesion was subjected to excisional biopsy along with extraction of 14. Histopathological evaluation revealed stratified squamous non keratinized epithelium. The under lying connective tissue was fibrous with few blood capillaries and chronic inflammatory cell infiltrate. At the periphery immature bone with osteocytes, fragments of cellular connective tissue containing globular calcifications resembling cementoid and bone surrounded by hemorrhagic areas were evident [Table/Fig-1c,d]. With these findings a final diagnosis of cemento-ossifying fibroma was confirmed. Patient was recalled after one month, where we found postoperative healing to be satisfactory [Table/Fig-2d-f].

## DISCUSSION

Bone dysplasias encompass a group of conditions where normal bone is replaced with fibrous tissue [1]. Benign fibro-osseous lesions are instabilities in bone metabolism where normal bone is supplanted by a connective tissue matrix that gradually develops cemento-osseous tissue [2]. Cemento-Osseous Dysplasia (COD) is a non-neoplastic process which is decently confined to the tooth-bearing area of the jaws or edentulous alveolar processes [3]. The term cemento-osseous dysplasia was espoused by World Health Organization (WHO) in 1992 [4]. In 2005 WHO sub classified these CODs into periapical, florid and other CODs [5]. These lesions are again sub-classified as Periapical Cemento-Osseous Dysplasia (PCOD) and Focal Cemento-Osseous Dysplasia (FCOD) based on their site of presentation. Su et al., considered PCOD and FCOD as two different entities of the same condition, at different locations [6]. Waldron suggested that both PCOD and FCOD are inter-related to each other and may develop into florid cemento-osseous dysplasia on long standing [7]. Summerlin et al., proposed

that the histopathological appearance of all fibro-osseous lesions would be similar, making them difficult to differentiate from each other [8].

PCOD most commonly occurs in the anterior region of mandible, in patients older than 30 years of age with significant female predilection [9]. Maxillary involvement of these lesions is a rare presentation [10]. In the present case, the lesion was observed to be in maxillary premolar region which was found to be a rare site. Su et al., reported that these lesions may also present with swelling associated with slight discomfort [6]. Similar presentation was observed in the present case, where a prominent swelling with mild pain was present.

PCODs are mostly asymptomatic unless they are secondarily infected. Generally the involved tooth remains vital and asymptomatic [11]. They can be described in three developmental stages with specific radiographic features. The early osteolytic stage will present with well-defined radiolucent area associated with loss of periodontal ligament and lamina dura [12]. The intermediate cementoblastic stage will present with small radiopacities within radiolucent area due to deposition of cementum like droplets in fibrous connective tissue [13]. The matured osteosclerotic stage is characterized by complete radiopacity [13]. The diagnosis is confirmed by evaluating the stage of development using radiographic examination. Waldron proposed that "in the absence of good clinical and radiographic information, a pathologist can only state that a given biopsy is consistent with a fibro-osseous lesion" [7]. He insisted upon adequate clinical and radiographic information for prompt diagnosis of these fibro-osseous lesions. Otherwise this diagnostic confusion may lead to misinterpretation and mismanagement of the lesion leading to poor prognosis.

But our case had a unique presentation of the lesion. It was mostly observed to be in the intermediate or cementoblastic stage with mixed radiopaque and radiolucent regions, but the typical characteristics were lost. The borders were indistinct and the exact extensions of the lesion were not evident. But the palatal bone expansion and the exhaustive involvement of the buccal bone were evident. This could be attributed to the patient's age. As they are usually reactive lesions and are found in people above 30 years, they usually tend to be more aggressive in younger age groups. These features made the clinicians and surgeons to opt for extraction of the offending teeth in the present case.

In such extreme conditions CBCT can help in determining the radio-density of the lesion consisting of bone and cementum [11]. The advent of CBCT has been a colossal impact on dental imaging. CBCT was initially developed for angiography but later, has found its long way into other medical applications, such as radiotherapy guidance and mammography [14]. This state of art imaging modality is useful in evaluating the intricate anatomy of many unusual presentations, measuring their exact dimensions and their association with surrounding structures.

The advantage of CBCT lies in clear demarcation of minute calcifications and multiplanar reformatting of images for viewing the pathology from all directions. Interestingly the lesion was observed to be more precise and clear in CBCT. In the present case, the shape of pathology was completely idiosyncratic and different from an orthodox lesion of COD, as the lesion was observed to grow out of the palatal surface with a prominent palatal expansion. The lesion presented with mixed hyper and hypodense areas with a prominent hypodense area encapsulating the lesion. The exhaustive involvement of the buccal bone clearly depicts the aggressive nature of the lesion.

Usually no treatment is required for PCOD, but followup is essential due to the possibility of secondary osteomyelitis and progression into florid cemento-osseous dysplasia. However in the present case, surgical excision was observed to be mandatory, as the lesion was symptomatic and was observed to be aggressive.

## CONCLUSION

Fibro-osseous lesions present with wide spectrum of characteristics that may mimic each other. The present case has an unusual occurrence at an early age, with atypical clinical and radiographic characteristics. In such conditions of severe diagnostic dilemma CBCT can be considered to be more effective in providing the intricate anatomical details of the lesion, thus providing good information for better diagnosis and prognosis.

## REFERENCES

- [1] White S, Pharoah M. Oral Radiology. Principles and Interpretation. 5<sup>th</sup> edition. St.Louis, MO: Mosby Inc. 2004.
- [2] MacDonald-Jankowski DS. Fibro-osseous lesions of the face and jaws. *ClinRadiol*. 2004; 59:11–25.
- [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] Kramer IR, Pindborg JJ, Shear M. The WHO histological typing of odontogenic tumours. A commentary on the second edition. *Cancer*. 1992;70:2988-94.
- [5] Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization Classification of Tumours. Pathology and Genetics of Head and Neck Tumours. Lyon: IARC Press. 2005.
- [6] 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 RadiolEndod*. 1997; 84:540-49.
- [7] Waldron CA. Fibro-osseous lesions of the jaws. *J Oral Maxillofac Surg*. 1993;51:828-35.
- [8] Summerlin DJ, Tomich CE. Focal cemento-osseous dysplasia: a clinicopathologic study of 221 cases. *Oral Surg Oral Med Oral Pathol*. 1994; 78:611-20.
- [9] Falace D, Cunningham C. Periapical cemental dysplasia: simultaneous occurrence in multiple maxillary and mandibular teeth. *J Endod*. 1984; 10:455-56.
- [10] Bulut EU, Acikgoz A, Ozan B, et al. Expansive focal cemento-osseous dysplasia. *JContemp Dent Pract*. 2012; 13:115-18.
- [11] MacDonald D. Oral and maxillofacial radiology, a diagnostic approach. New Jersey: Wiley-Blackwell, 2011; 176–78.
- [12] Melrose RJ. The clinico-pathologic spectrum of cementoosseous dysplasia. *Oral MaxillofacClin Nor Am*. 1997; 9:643-53.
- [13] Bhandari R, Sandhu SV, Bansal H, Behl R, Bhullar RK. Focal cemento-osseous dysplasia masquerading as a residual cyst. *ContempClin Dent*. 2012; 3:S60-62.
- [14] Scarfe WC, Farman AG. What is cone-beam CT and how does it work? *Dent Clin N Am*. 2008; 52:707–30.

### PARTICULARS OF CONTRIBUTORS:

1. Senior Lecturer, Department of Oral Medicine and Radiology, G.S.L Dental College and Hospital, Rajahmundry, Andhra Pradesh, India.
2. Professor and Head of the Department, Department of Oral Medicine and Radiology, St.Joseph Dental College, Eluru, Andhra Pradesh, India.
3. Post Graduate Student, Department of Oral Medicine and Radiology, St.Joseph Dental College, Eluru, Andhra Pradesh, India.
4. Senior Lecturer, Department of Oral Medicine and Radiology, St.Joseph Dental College, Eluru, Andhra Pradesh, India.
5. Reader, Department of Oral Medicine and Radiology, St.Joseph Dental College, Eluru, Andhra Pradesh, India.

### NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sai Kiran Chennouju,  
Senior Lecturer, Department of Oral Medicine and Radiology, G.S.L Dental College and Hospital,  
Rajahmundry-533296, Andhra Pradesh, India.  
E-mail: iamchennoujukiran@gmail.com

Date of Submission: **Sep 15, 2015**

Date of Peer Review: **Nov 07, 2015**

Date of Acceptance: **Mar 09, 2016**

Date of Publishing: **May 01, 2016**

FINANCIAL OR OTHER COMPETING INTERESTS: None.