

A Rare Case of Florid Cemento Osseous Dysplasia with Gingival Manifestations

Vasudevan Sanjay^{1*} Reddy Ajay P² Kumar Rajani G³ Vaishnavi Varanasi⁴

¹Professor and Head, Department of Periodontics, Army College of Dental Sciences, Secunderabad, India.

²Professor, Department of Periodontics, Army College of Dental Sciences, Secunderabad, India.

³Reader, Department of Periodontics, Army College of Dental Sciences, Secunderabad, India.

⁴Senior Lecturer, Department of Periodontics, Army College of Dental Sciences, Secunderabad, India.

ABSTRACT

Background: A 76-year-old female patient came with chief complaint of swelling of gums, dull pain with mild bleeding in the lower right back tooth region since 2 weeks associated with discomfort while eating. Biopsy of the gingival tissue was done and diagnosed as fibrous hyperplasia. However radiographic examination revealed it to be a case of Florid Cemento Osseous Dysplasia which is characterized by there placement of normal bone tissue with fibrous tissue that contains newly formed mineralized tissue, benign and multifocal fibro-osseous dysplastic lesion affecting tooth-bearing areas of the jaw. This article reports and discusses about how a gingival swelling will lead to underestimation of underlying pathology of bone. Hence a small gingival lesion cannot be under estimated and through clinical and radiographic examination has to be done to examine underlying bone pathology.

Keywords: Osteomyelitis, Florid cemento osseous dysplasia, Paget's Disease.

INTRODUCTION

Florid cemento-osseous dysplasia (FCOD) is defined as "Lobulated masses of dense, highly mineralized, almost acellular cemento-osseous tissue typically occurring in several parts of the jaw".^[1] Florid cemento-osseous dysplasia (FCOD) belongs to the group of fibro osseous lesions in which normal bone is replaced by fibrous connective tissue and calcified cementum tissue of the avascular type which appear predominantly radiolucent (osteolytic phase), become mixed over time (cementoblast phase), and ultimately become radiopaque (osteogenic phase) with a thin radiolucent peripheral halo. ^[2] The term "florid osseous dysplasia" was introduced by Melrose et al in 1976. Waldron (1985) proposed the term "FCOD" as the dense and sclerotic masses that form resemble the cementum. When lesions with similar radiologic and microscopic features extend to involve two or more quadrants of the jaw, it is termed as Florid cemento-osseous dysplasia (FCOD). Clinical and

radiographic findings reveal three subtypes: periapical (affecting anterior mandible), focal (single lesion), and florid.^[6]

The aim of this case report was to highlight the importance of thorough examination and investigation of simple gingival swelling so that we don't miss out on underlying major pathology.

CASE REPORT

A 76 year female patient came with chief complain of swelling of gums and mild bleeding in the lower right back tooth region since 2 weeks associated with discomfort while eating. Intraoral examination revealed, a swelling which was sessile, originating from mesial of 47 and extending into attached gingiva, measuring approximately 5x5 mm with smooth shiny surface and fibrotic in consistency (Figure 1 – Clinical picture of the gingival swelling).

Received: Apr. 6, 2019: Accepted: May. 19, 2019

*Correspondence Dr. Vasudevan Sanjay.

Department of Periodontics, Army College of Dental Sciences, Secunderabad, India.

Email: drsanjayvasudevan2003@yahoo.co.in



Fig 1: Clinical case picture.

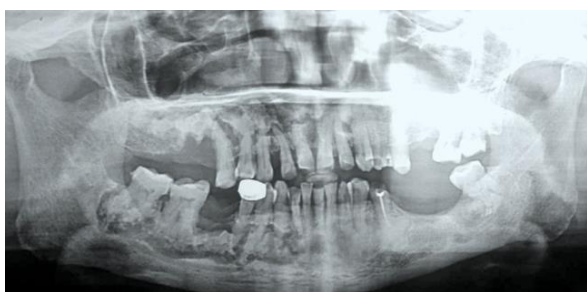


Fig 2: Radiograph showing mixed radiopaque and radiolucent appearance of the alveolar bone.

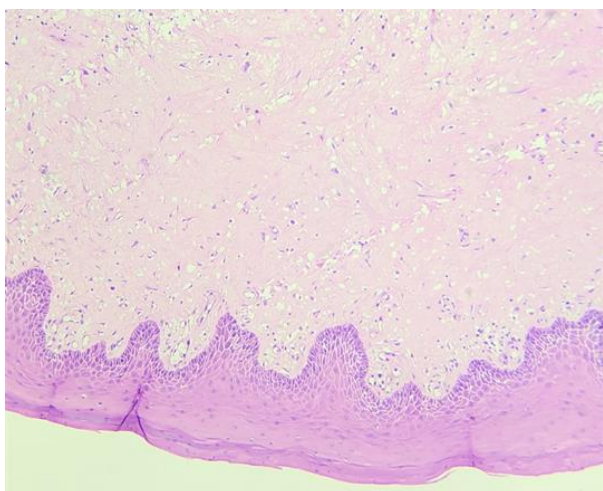


Fig 3: Histological section in scanner view with H&E staining showing Stratified squamous parakeratinised epithelium overlying a fibromyxoid stroma.

Panoramic radiographic examination revealed generalized horizontal bone loss and edentulous area in the third and fourth quadrant. On site specific evaluation around 46, revealed mixed radiographic changes, radio opacity round the

apical regions of 47 and 48 and radiolucency bordered above by the root apices and below by upper border of inferior alveolar canal. It also revealed diffuse, multinodular, irregularly shaped radiopacities or "cotton wool" appearance throughout the alveolar process of both quadrants, in both the maxilla and the mandible. Initial therapy was done with antibiotic coverage (Figure 2 – Radiograph showing mixed radiopaque and radiolucent appearance of the alveolar bone). Case was reviewed after a week and decision to take a biopsy was made since there was no change in the healing of the lesion. Medical history revealed patient is a known hypertensive and is on medication since 5years and is well under control. Patient elicits a dental history of traumatic extraction of 36, 37 and 46 elsewhere 5 years back associated with delayed healing.

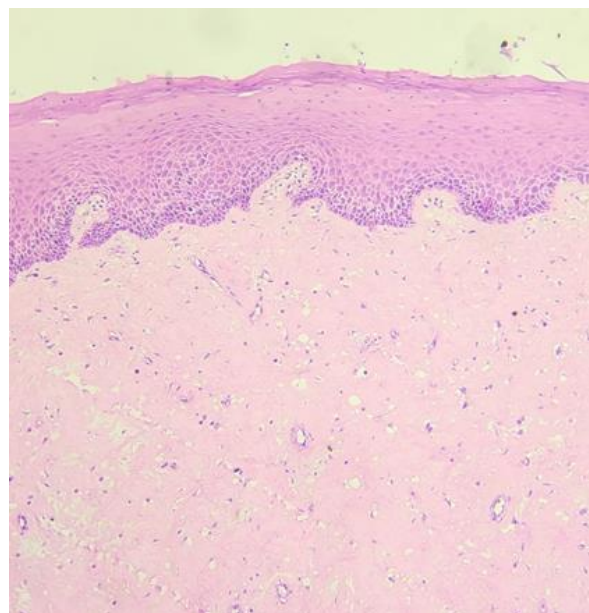


Fig 4: Histological section in scanner view with H&E staining showing mature collagen bundles arranged haphazardly with myxoid stroma.

INVESTIGATIONS

Complete blood picture revealed normal values. Biochemical analysis of serum alkaline phosphatase, calcium and phosphorus were 45 U/L; 9.4 mg/dl and 3.5 mg/dl respectively, being within the normal range, which are 39-117 U/L; 8.8-10.6 mg/dl and 2.5-4.8 mg/dl respectively, according to the protocols used. These tests were performed to make the differential diagnosis with Paget's disease, which shows results abnormally elevated. The

clinical diagnosis was florid cemento-osseous dysplasia with an associated region of osteomyelitis.

HISTOPATHOLOGY

The biopsy report of the gingival tissue specimen, viewed under H&E stained sections, show epithelium overlying a fibromyxoid stroma. Under higher magnification, the epithelium is stratified squamous type with most flattened stromal junctions. No apparent abnormality in epithelium is observed. The underlying stroma mostly shows mature collagen bundles arranged haphazardly with few areas showing myxoid changes with stellate fibroblasts and extensive ground substance. Histological examination revealed diagnosis of fibrous hyperplasia. Bony changes could not be ascertained histologically as patient was not giving consent for bone biopsy. So based on the clinical and radiographic findings and present histopathology report, the final diagnosis was made (Figure 3- Histological section in scanner view with H&E staining showing stratified squamous parakeratinised epithelium overlying a fibromyxoid stroma), (Figure 4 - Histological section in scanner view with H&E staining showing mature collagen bundles arranged haphazardly with myxoid stroma).

TREATMENT

Excisional biopsy of the gingival swelling was done under local anesthesia. Antibiotics (AMOXICILLIN 500mg, TID, 5 days) and analgesic (IBUPROFEN 400mg & PARACETOMOL 325mg, BD, 5 days) was prescribed. However, FCOD is a condition in which the diagnosis relies on radiology and clinical findings alone. This condition being asymptomatic requires no treatment, but regular follow-up is needed.

DISCUSSION

The term florid cemento-osseous dysplasia (FCOD) has been proposed in the third edition of World Health Organization's (WHO) "World Health Organization Classification of Tumours / Pathology and Genetics of Head and Neck Tumours". This condition has been interpreted as a dysplastic lesion or developmental anomaly arising in tooth-bearing areas. It is strictly located in tooth-bearing areas and is not associated with any other skeletal

disease. It may emerge as the result of progressive alveolar atrophy under a denture or after extraction of teeth in an affected area or dysplastic changes in the periodontal ligament might be a cause for this disease. Another disease that may resemble the FCOD is chronic diffuse sclerosing osteomyelitis (CDSO). It appears as a single, poorly delineated opaque segment of the mandible whereas FCOD is seen as multiple round or lobulated opaque masses.^[3]

Over time, as it became clear that conditions labelled "Chronic Sclerosing Osteomyelitis" were not associated with infection, leading to the development of "Cementoma" and "Benign Periapical Fibroma", there were those such as Robinson, who still proposed a reactive cause for these lesions, calling this lesion "Osseous Dysplasia". Waldron et al advocated a periodontal ligament origin for this group of lesions because this class of lesion was generally confined to the alveolus and contained a histopathological appearance similar to that seen in the normal periodontal ligament. This concept is still generally applied today.

Although Waldron considered that the majority of cases called "Chronic Sclerosing Osteomyelitis" were "Florid Osseous Dysplasia" (FOD), Panders and Hadders used "Chronic sclerosing Osteomyelitis" in relation to cases that were clearly similar to Groot et al, "Diffuse Sclerosing Osteomyelitis", differing markedly from "Florid Osseous Dysplasia". The former affected the basal process of the mandible in addition to the alveolar process, to which the latter was largely confined.^[1] Paget's disease of the bone may also have a cotton-wool appearance. However, this disease affects the bone of the entire mandible and shows loss of lamina dura, whereas FCOD is centered above the inferior alveolar canal.

Differential diagnosis of FCOD should also include sclerosing osteomyelitis, which can be a complication of the disease. It appears as a single, poorly delineated opaque segment of the mandible, whereas FCOD is seen as multiple round or lobulated opaque masses. Chronic diffuse sclerosing osteomyelitis involves the body of the mandible from the alveolus to the inferior border and may extend into the ramus.^[5]

Florid cemento-osseous dysplasia(FCOD), describes an extensive variant of the reactive fibro-osseous process known as cementoma, which has been alternately referred to as sclerosing osteitis, multiple enostosis, gigantiform cementoma, multiple cemento-ossifying fibromas, sclerotic cemental masses of the jaws, multiple periapical osteofibromatosis, periapical cementoblastoma, and gigantiform cementoma. Despite the plethora of appellations, FCOD is a well-defined clinicoradiologic entity involving the apices of vital teeth and distinguished by the presence of lesions in multiple quadrants of the jaw.

All subtypes of COD progress through three stages of evolution, each with characteristic radiographic and histologic features. In the early, or lucent phase, resorption of normal bone and deposition of fibrous tissue along the periodontal ligament leads to the development of discrete radiolucencies around the root apices of vital teeth or previously tooth-bearing areas. These lucent lesions can have a round, ovoid, or lobular configuration and often feature a sclerotic margin of varying width. As lesions evolve, progressive intralesional sclerosis leads to a mixed lucent and sclerotic appearance. The intermediate stage, also referred to as the mixed stage, denotes the development of radiopaque density within these radiolucent structures. Older lesions may appear entirely radiopaque; these lobulated, confluent, densely sclerotic masses often have a thin, radiolucent rim which can help differentiate them from late-stage Paget's disease.

Uncomplicated FCOD may persist for an indefinite period of time without causing symptoms. Infection may result in dull pain, purulent mucosal discharge, or jaw expansion. Diminished vascularity in the affected tissue predisposes to chronic infection, pathologic fracture, and poor healing. Inadequate diffusion of antibiotics into the densely sclerotic tissue generally renders them ineffective. Slow sequestration of the cementum-like masses is usually followed by healing in that area. Asymptomatic lesions require no invasive treatment. Although histologically benign, FCOD may rarely undergo dysplastic hyper proliferation, which can result in cosmetic deformity and functional disturbances. [4]

CONCLUSION

It is a primary inflammatory condition of the mandible with cyclic episodes of unilateral pain and swelling. The affected region of the mandible exhibits a diffuse opacity with poorly defined borders. [6] In asymptomatic patients line of treatment is they should be kept under observation, and regular radiological follow-up should be made. [7] Biopsy for histopathological examination may not be required to confirm the diagnosis due to their characteristic radiological features. On the contrary, such attempt may increase the risk of infection or fracture of the jaw and hence will adversely affect the patient's health. [8]

So a case giving a misconception of fibrous hyperplasia with underlying bony changes leading to formation of FCOD should be properly examined and cannot just be concluded with a simple soft tissue excision. Thus a simple case of gingival swelling should never be underestimated and thorough and detailed examination is necessary for every case to rule out major underlying pathology.

ACKNOWLEDGEMENT

This research was supported by Dr. Harshavardhan Jois, HOD of Oral Pathology.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

REFERENCES

1. References: Mac Donald-Jankowski . Florid cemento osseous dysplasia: a systematic review. Dento Maxillofacial Radiology 2003;141-149.
2. Debora Delai, Anarela Bernardi, Gabriela Santos Felipe,Cleonice da Silveira Teixeira, Wilson Tadeu Felipe, Mara Cristina Santos Felipe . Florid Cemento-osseous Dysplasia: A Case of Misdiagnosis, J Endod 2015, 132-135.
3. Cyntia Helena Pereira de Carvalho , Emeline das Neves de Araújo Lima a Joabe dos Santos Pereira a Ana Miryam Costa de Medeiros Ericka Janine Dantas da Silveira. Florid cemento-osseous dysplasia and osteomyelitis: A case report of a simultaneous presentation. Rev Odonto Cienc 2012;166-169.

4. Sarah Fenerty , Wei Shaw , Rahul Verma , Ali B. Syed, Riya Kuklani , Jie Yang et al. Florid cemento-osseous dysplasia: review of an uncommon fibro-osseous lesion of the jaw with important clinical implications. Skeletal Radiol DOI 10.1007/s00256-017-2590-0.
5. Harika Kutluay Koklu , Dilek Aynur Çankal , Suleyman Bozkaya , Gulfem Ergun , Emre Barış . Florid cemento-osseous dysplasia: Report of a case documented with clinical, radiographic, biochemical and histological findings. J Clin Exp Dent. 2013.
6. Riccardo Aiuto , Federico Gucciardino , Roberta Rapetti , Sandro Siervo , Andrea-Edoardo Bianchi .Management of symptomatic florid cemento-osseous dysplasia: Literature review and a case report. J Clin Exp Dent. 2018.
7. Preeti Arwind Jha, Ashwini Ashok Pai, Nikhil Diwan, Rashmi Sapkal. Florid Cemento-osseous Dysplasia: A Series of Case Report. Journal of Dental and Allied Sciences ;2017.
8. Bijay Kumar Das, Surya Narayan Das, Aprna Gupta, Suryakanti Nayak .Florid cemento-osseous dysplasia. Journal of Oral and Maxillofacial Pathology; 2013.