Case Report

Focal cemento-osseous dysplasia: A case report with a review of literature

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Abstract

Cemento-osseous dysplasia (COD) is the most prevalent lesion and presents as an asymptomatic, mixed radiolucent/radiopaque lesion in the tooth-bearing region of the jawbones. Histological features of COD include a cellular connective tissue stroma interspersed by the islands of woven or lamellar bone and cementum-like calcifications. Radiographically, the early COD lesions appear radiolucent, whereas in the mature lesions, radiopacities are observed surrounded by a thin rim of radiolucency. Early lesions tend to get misdiagnosed as their radiographic appearance mimics periapical cyst or granuloma. In the mixed radiolucent-radiopaque stage, the lesion could be confused with chronic sclerosing osteomyelitis, cemento-ossifying fibroma, odontoma and osteoblastoma. A correct diagnosis is of utmost importance as most of the CODs are self-limiting and nonneoplastic and do not require surgical intervention. However, periodic follow-up is recommended because occasional cases of focal COD are known to progress into florid COD. Here, we present the case of focal COD in a 27-year-old male patient.

Keywords: Benign fibro-osseous lesion, cemento-osseous dysplasia, periapical lesion

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INTRODUCTION

The term "benign fibro-osseous lesion" refers to a group of nonneoplastic conditions in which the normal bone is replaced with a fibrous connective tissue matrix containing abnormal bone or the cementum.[1] Fibro-osseous lesions of the jaw include fibrous dysplasia, ossifying fibroma, and cemento-osseous dysplasia (COD). COD is the most prevalent osseous-fibrous lesion and presents as an asymptomatic, mixed radiolucent/radiopaque lesion in the tooth-bearing areas of the jawbones.[2] As the pathological features of COD are similar to those of fibrous dysplasia and ossifying fibroma, the correct diagnosis can be problematic, but it is critical to the appropriate management.[3]

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In 1994, the term "focal COD" was suggested by Summerlin and Tomich.[4] In 2005, the World Health Organization described the three distinct forms of COD based on the clinic-radiographical appearances, namely focal COD, periapical COD and florid COD.[5] Periapical COD consists of solitary or multiple lesions found in the apical region of vital mandibular anterior teeth. Focal COD includes the asymptomatic lesions occurring commonly in the middle-aged females and involving the posterior mandibular molar region. Florid COD encompasses more extensive and multifocal forms involving both maxilla and mandible or occurring bilaterally in the mandible.[6]

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Radiographically, evident changes occur over time due to the alterations in the internal radiolucency or radiopacity of these entities. On radiographic evaluation, an early COD lesion may appear as a round/oval apical radiolucency with a well-defined radiopaque border. In the second/mixed stage, a radiolucent lesion may include radiopaque sclerotic masses. In the mature stage, the internal mixed area becomes completely radiopaque with a thin radiolucent periphery.[7]

The diagnosis of COD is often based on the typical clinical and radiographical features. In many cases, the diagnosis of COD may be particularly confusing because the radiographic features of early lesions tend to mimic those of a periapical cyst or granuloma.[8] Similarly, in the mixed radiolucent-radiopaque stage, the lesion could be confused with chronic sclerosing osteomyelitis, cemento-ossifying fibroma, odontoma and osteoblastoma.[9] In such a scenario, histopathological diagnosis becomes extremely useful for the appropriate treatment of the patient.

Histological features of COD include a cellular connective tissue stroma interspersed by the islands of woven or lamellar bone and cementum-like calcifications.[2]

Here, we present the case of mandibular focal COD arising in the mandibular left third molar region in a 27-year-old male patient.

CASE REPORT

A 27-year-old male patient reported with pain in the lower left back region of the jaw. On clinical examination, only the cusp tips of partially impacted mandibular left third molar (38) with inflamed pericoronal flap were seen. With a provisional diagnosis of pericoronitis associated with 38, the patient was advised for orthopantomograph (OPG) and was advised for the extraction of 38.

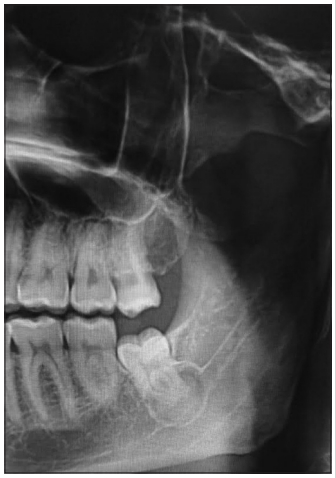

Figure 1: Orthopantomograph of the present case

The radiographic picture revealed an impacted (mesioangular) 38 with a round, well-defined lesion of approximately 2 cm in diameter, with radiopaque central mass surrounded by the thin radiolucent rim with sclerotic border [Figure 1]. The lesion was extending periapically from the middle third of the roots and was overlapping the inferior alveolar nerve canal. There was no evidence of root resorption. Radiographical diagnosis of odontome, cementoblastoma, hypercementosis and focal COD was considered.

Surgical extraction of 38 along with the complete excision of the lesion was performed under local anesthesia. During the extraction, it became evident that there was a calcified mass fused to the roots of 38.

The microscopic examination of hematoxylin and eosin-stained tissue sections revealed the presence of numerous trabeculae of calcified tissue resembling osteoid with intertwining fibrous connective tissue [Figure 2]. Trabeculae had osteocytes in lacunae. A few globular areas of cementum-like tissue were noted along with few blood vessels. Based on the microscopic findings, the final diagnosis of focal COD was given. The patient was kept under follow-up for 3 months, and postoperative OPG was taken to ensure the healing of bone.

DISCUSSION

COD constitutes a group of fibro-osseous lesions occurring in tooth-bearing region of the jawbones. The etiology and pathogenesis are unknown, but it is thought to be arising from the periodontal ligament because of the proximity of the lesions to the teeth and the formation of cementum-like calcifications.[2] Trauma from occlusion, caries, periodontal disease, infection, hormonal imbalance, or systemic diseases is also considered as causative factors. Based on the clinical and radiographical features, COD is divided into three groups: focal, periapical and florid.[3] All three patterns of COD demonstrate similar histopathological features. Microscopic examination shows a mixture of woven bone, lamellar bone and cementum-like tissue against a fibrous connective tissue background.[2,3]

The concept of focal osseous dysplasia was not clarified until the mid-1990s. Before that time, most cases were misdiagnosed as a variant of ossifying fibroma.[3] In 1994, Summerlin and Tomich studied 221 osseous dysplasia cases and suggested the term "focal COD."[4] Focal COD presents as asymptomatic lesion associated with the vital

tooth. Focal COD may occur in any area of the jaws, but the mandibular posterior region is the most common site of occurrence.[2,3] The mandibular first molar tooth is most commonly affected. However, in the present case, the lesion was found in association with mandibular impacted third molar. The African–American population followed by the Asian population is considered markedly predisposed to focal COD.[10] Most cases are diagnosed in patients in their fourth or fifth decades with female predilection.[11-14] The findings in the present case were in unique in a way that occurred in a 27-year-old male patient.

Although the previous reports have generally suggested that COD is usually asymptomatic and discovered on routine dental radiographs, Kawai *et al*. demonstrated that of 54 patients, 59% presented at least one symptom of inflammation, including pain, swelling, purulent discharge and tenderness on palpation.[12] In the present case, the patient complained of pain with respect to partially impacted third molar.

Most of the times, the lesion is accidently detected on routine dental radiographic examination. It arises in close proximity to the teeth, above the inferior alveolar nerve canal and usually does not cause cortical expansion. According to the literature review, a radiopaque mass with poorly defined radiolucent rim is noted in most of the cases, whereas a sclerotic border has been less frequently reported.[4,8,9,12] In the present case, the lesion had a well-defined sclerotic border. In 2011, in an institutional review done by Alsufyani and Lam, it was observed that most cases of COD were in the mixed radiolucent-radiopaque stage (72.0%), and the internal radiopacities were described as being dense and cementum-like (61.4%).[6] The radiographic findings in the current study were in agreement with the literature.[6,13,14]

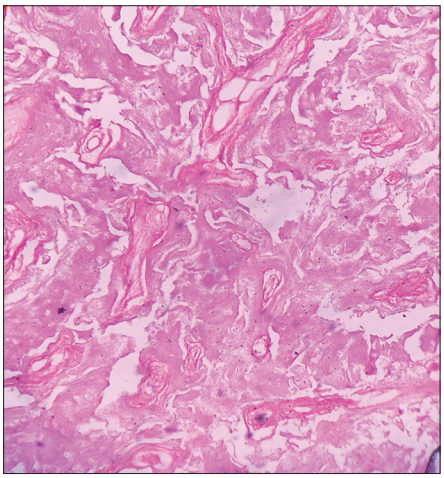

Figure 2: Photomicrograph of H&E-stained slide of focal cemento-osseous dysplasia

The radiographic appearance of focal COD may change with the extent of mineralization. Well-defined radiolucency can be seen in the early or osteolytic stage. In the intermediate or cementoblastic stage, radiopacity with sclerotic borders is seen with the characteristic radiolucent rim around the radiopacity. The last stage is more mature and osteosclerotic. A sclerotic radiopacity with poorly-defined borders is typically seen in this stage.[7,8,14,15] As the lesion matures, the ratio of fibrous connective tissue to mineralized tissue decreases. With maturation, the bony trabeculae become thick curvilinear structures. In the final radiopaque stage, individual trabeculae fuse and form lobular masses composed of sheets or fused globules of relatively acellular and disorganized cemento-osseous material.[3]

Although focal COD is generally self-limiting and nonneoplastic, sometimes it can reach large sizes. Gumusok *et al*. reported a focal COD lesion that had reached a large size and caused the destruction of the alveolar bone.[15] Such cases require surgical excision of the lesion followed by the regular follow-up.

A biopsy is the only definitive method for distinguishing COD from other bony lesions. Particularly, the intraoperative gross findings aid in distinguishing between COD and ossifying fibroma. COD consists of easily fragmented and gritty tissue that can be curetted easily but does not separate cleanly from the adjacent normal bone. In contrast, ossifying fibromas tend to separate cleanly from the bone.[3] The biopsied tissue in our case was fused to the roots of 38 and to the alveolar bone and on histopathological examination revealed multiple bony and cementum-like material and fibrous connective tissue,

Table 1: Summary of focal cemento-osseous dysplasia documented in the literature (2009-2019)

Author	Age of patient (years)	Sex of patient	Site	Clinical finding
Gumusok *et al*.[15]	63	Female	Mandibular second premolar	Incidental radiographic finding
Bhandari *et al*.[18]	25	Female	Mandibular first molar	Painful firm well-defined swelling
Rao *et al*.[19]	47	Female	Mandibular first molar	Incidental radiographic finding
Bulut *et al*.[20]	24	Female	Edentulous mandibular premolar-molar region	Painless, firm swelling causing discomfort on wearing denture
Cankaya *et al*.[21]	69	Female	Edentulous mandibular premolar-first molar region	Firm, painless and stable mass

which was the typical appearance of COD from previous findings.[16,17]

A summary of focal COD documented in the literature during the period from 2009 onward is presented in Table 1.

CONCLUSION

COD represents a group of lesions with variable radiographic appearance. The present case is unique in terms of the gender of the patient, site of occurrence of the lesion and associated symptoms. This case highlights the importance of radiographic evaluation before performing the surgical procedure.

Declaration of patient consent

The authors ensure that we have obtained appropriate patient consent forms. The patient was ensured that his identity will remain confidential and he has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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