

Cystic process during the follow-up of a focal cemento-osseous dysplasia

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ABSTRACT

Introduction: Focal cemento-osseous dysplasia (FCOD) is a benign and asymptomatic fibro-osseous malformation of the jaws. Etiology of FCOD is not clear. It is incidentally detected on radiographic examination. **Case Report:** A 23-year-old female case of FCOD together with a residual cyst and a simple bone cyst occurred in the same region within a five-year follow-up period is presented. **Conclusion:** The FCOD is a lesion that certainly needs long-term follow-up. The clinicians should be alert for different clinical outcomes ranging from simple bone cyst to florid cemento-osseous dysplasia during follow-up.

Keywords: Focal cemento-osseous dysplasia, Long-term follow-up, Residual cyst, Simple bone cyst

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INTRODUCTION

Focal cemento-osseous dysplasia (FCOD), despite its indefinite etiology, appeared in literature as a reactive lesion considered arising from the periodontal ligament [1]. However, replacement of the connective tissue by the cemento-osseous tissue instead of normal bone tissue due to a bone metabolism disorder is considered to be the pathophysiology of benign fibro-osseous lesions [2, 3]. FCOD is a benign condition and appears in the apical region of the teeth with vital pulp or in the area of extracted teeth [1, 4, 5]. Clinically, FCOD is observed mostly in the posterior mandibular region, can reach 1–2 cm in size, and may lead to local bone enlargement. In addition, FCOD may be infected secondarily and thereby becomes symptomatic [1, 6]. The lesion is more common in females in the 4th–5th decades of life [1, 4, 6, 7].

Focal cemento-osseous dysplasia can be detected only by radiographic examinations. Radiographic image shows variations and consists of the combination of radiopaque image and radiolucent image with sclerotic boundary. The lesion may display a wide range of appearance from intense radiopacity to complete radiolucency [1, 4, 5].

Histopathological appearance of FCOD varies in each stage. While vascular structures are observed within the fibrous connective tissue in early stages, irregular cement-like structures attract attention in later stages. In general, FCOD consists of fusiform cells and cement-like structures [8].

Herein, we present a case of a lesion diagnosed as focal cemento-osseous dysplasia and discuss its diagnostic, therapeutic, and follow-up periods in the light of above-mentioned information and histopathological evaluation.

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CASE REPORT

A 23-year-old female was admitted to the Department of Maxillofacial Surgery at Yüzüncü Yıl University Faculty of Dentistry in April 2011 for the removal of deeply decayed tooth (number 38). Her anamnesis revealed that she had no systemic disease. The patient also had a slow growing swelling in the left mandibular molar region in the last one year with no numbness in the relevant side. Intraoral examination demonstrated bone expansion with pain on palpation in the buccal sulcus in the region of previously extracted mandibular left first molar. Extraoral examination revealed a mild asymmetry in the facial region and lymphadenopathy. Radiographically, a lesion with in size 2x2.5 cm displaying mixed radiopaque and radiolucent appearance was detected between the apexes of the mandibular left second premolar and second molar teeth below the toothless crest and 3 mm inferior to the top of the crest (Figure 1).

After obtaining the consent of the patient, the third molar was extracted firstly; thereafter, the area was accessed by elevating a mucoperiosteal flap under local anesthesia and the lesion was curetted. Bony boundaries were shaved by a drill. The well-demarcated sequestered material was sent for histopathological examination (Figure 2). On histopathological examination, the material was dense, tuberous, and bone-cement in appearance, as well as proliferation containing hard tissue components displaying irregular lamellation and cellular connective tissue components consisted of focally remarkable fusiform fibroblastic cells were observed; the diagnosis was “FCOD”.

At the fifth month of follow-up, the patient was readmitted to our clinic with pain in the relevant area. No problem was observed with regard to oral tissue healing and surrounding tissues were healthy. On her extraoral examination, the left submandibular lymph nodes were tender. Radiographic examination demonstrated a radiolucent, well-demarcated lesion suggestive of a cystic formation (1.5x1 cm) in the relevant region (Figure 3). Although the lesion was adjacent to the inferior alveolar nerve, the patient declared no numbness. After one-week of antibiotherapy, the patient underwent surgery under local anesthesia. The lesion containing cystic fluid was enucleated and the removed material was sent for histopathological examination together with the clinical information attached. As the result of histopathological evaluation, the diagnosis was established as “residual cyst”.

At the end of the third year of her ongoing follow-up visits, radiological examination of the relevant area demonstrated a large, drop-shaped radiolucency (2x2 cm) (Figure 4). The patient was re-operated; the area was accessed under local anesthesia and no cystic fluid or cyst epithelium was observed. The area was decompressed, the inner side of the lesion was curetted, and the soft tissue cover was primarily sutured. The lesion was diagnosed as “simple bone cyst” considering the lesion history of the

patient together with radiological and clinical findings. No complaint regarding the surgical site or no healing problem was encountered in the patient 1 year after the last surgery and the patient is still under follow-up (Figure 5).

DISCUSSION

Recurrence of FCOD is not a common issue or it is not a lesion followed for a long time. Many cases do not become symptomatic clinically thereby do not require treatment [7].



Figure 1: Preoperative panoramic image.



Figure 2: Removed materials.

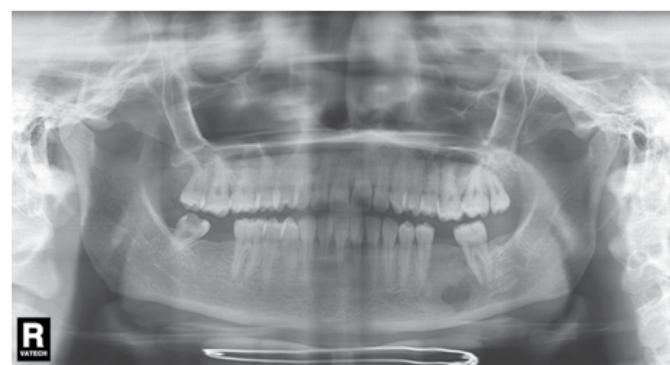


Figure 3: Panoramic image of the residual cyst.



Figure 4: Panoramic image of the simple bone cyst.



Figure 5: Panoramic image of the completely improved region at the end of fifth year.

The present case was followed-up for five years; during this period, a residual cyst and a simple bone cyst were observed in the enucleation site of FCOD. Researchers highlighting the importance of monitoring of these lesions suggest that the possibility of transformation into florid cemento-osseous dysplasia, which exhibits a much more malignant structure than simple bone cyst, should be considered and that follow-up period is a process not to be ignored [1].

Suei et al. reviewed 132 simple bone cysts of the jaws and reported the recurrence rate for cases with cemento-osseous lesions to be 75% [9]. They also reported that improvement or recurrence in majority of their cases was observed within three years and five months of the surgery. The present case was readmitted to our clinic with cystic formation in the same region within 5 months following FCOD enucleation. The fact that this lesion appeared as a simple bone cyst in three years after the initial surgery performed following the diagnosis of residual cyst supports the above-mentioned study. However, in the majority of cases follow-up period is limited with six months [10, 11].

Focal cemento-osseous dysplasia should be certainly considered in the differential diagnosis of odontogenic

pathologies that are considered suspicious for residual cyst and periapical granuloma since it has been reported that FCOD can display similar symptoms with periapical granulomas and may mimic residual cyst although FCOD is seen in toothless jaws [12, 13].

Although they are rare, cases of simple bone cyst together with fibro-osseous lesions can also be encountered [4, 14]. However, to the best of our knowledge, there are only two cases of coexistence of simple bone cyst with FCOD reported to date [15, 16]. In the present case, FCOD and simple bone cyst appeared to be two different lesions of the same region as the initial and final phases of successive processes. In a case reported by Mupparapu et al. in 2008, histopathological evaluations of three separate lesions localized in the mandible, which were suggestive of focal or periapical cemento-osseous dysplasia according to differential diagnosis and appeared as mixed radiographic image, and their recurrences revealed simple bone cyst [16]. Accordingly, in the light of these reported findings, in case of clinical or radiological findings that are suggestive of cemento-osseous dysplasia, it is necessary to investigate the presence of simple bone cysts clinically at the same time or different times and to make a histopathological differentiation. Thereafter, the most important issue to be paid attention is the necessity of monitoring of healing with six-month interval at most in the first three years of follow-up period. In the present case, the first two-year follow-up was performed in the 1st, 3rd, 6th, 12th, and 24th months. Thereafter, the patient was monitored with longer follow-up intervals, which excluded the chance of detecting the lesion when it was much smaller.

Etiological process for the coexistence of benign fibro-osseous lesions such as FCOD and simple bone cyst can be explained by bone metabolism disorders resulting in different outcomes in different ways and in different regions in the jaw [17]. In the etiology of successive processes, it can be thought that bone metabolism disorder continues but results in an empty bone cavity instead of different cemento-osseous infiltration due to different body response in time. On the other hand, enucleation and local curettage performed in the initial surgery for FCOD may cause a traumatic effect in the region and thereby may provide a basis for simple bone cyst with impairment in feeding-drainage cycle.

Differential diagnosis of cemento-osseous lesions differs depending on the developmental stage. While periapical granuloma and chronic osteomyelitis are the pathologies that should be considered in the differential diagnosis in early phase, chronic sclerosing osteomyelitis, ossifying fibroma, odontoma, and osteoblastoma are the pathologies that should be considered in the differential diagnosis in the intermediate and late phases [18].

Although FCOD is usually encountered in the middle-age-group, the pathology affected a young adult individual in the present case report. The patient being a female and the lesion occurring in a toothless area appeared to be consistent with literature.

Definite indications for surgery include the lesion being symptomatic, expanded, and leading to functional and esthetic losses in the patient. The cases that are asymptomatic and do not expand may require not only surgery but also recurrent surgical procedures since they may become more aggressive being involved in a reactive process, as was in the present case.

CONCLUSION

In conclusion, Focal cemento-osseous dysplasia (FCOD) is a lesion that certainly needs long-term follow-up at frequent intervals whether it is asymptomatic and does not require treatment or becomes symptomatic and requires surgical excision. The clinicians should be alert for different clinical outcomes ranging from simple bone cyst to florid cemento-osseous dysplasia during follow-up.

Author Contributions

Cennet Neslihan Eroglu – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for intellectual content, Final approval of the version to be published

Volkan Kaplan – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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