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CASE REPORT/ OLGU SUNUMU

Seven-year Follow-up of A Focal Cemento-Osseous Dysplasia Associated with Mandibular First Molar: A Case Report

Mandibular Birinci Molar ile İlişkili Fokal Semento-osseöz Displazinin 7 Yıllık Takibi: Bir Olgu Sunumu

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ABSTRACT

Cemento-osseous dysplasia (COD) is a benign fibro-osseous lesion with replacement of normal bone tissue by fibrous connective tissue. This case report presents an asymptomatic mixed radiolucent-radiopaque lesion in the posterior mandible of a 37-year-old female patient's panoramic radiography. Clinical and radiographic evaluation led to the diagnosis of focal cemento-ossifying dysplasia (FCOD) and regular radiographic follow-up was recommended as focal COD may progress to florid COD. The lesion was followed up for seven years with three examinations. During this time, the lesion remained asymptomatic and the associated tooth remained vital. An increase in the size of the internal hyperdense content was observed in cone-beam computed tomography evaluation during the follow-up visits. The maturation process was well observed. A minimally increased expansion was also noted.

Keywords: cemento-osseous dysplasia, cone-beam computed tomography, fibro-osseous dysplasia

ÖZ

Semento-osseöz displazi (SOD), normal kemik dokusunun fibröz bağ doku ile yer değiştirdiği, iyi huylu bir fibro-osseöz lezyondur. Bu olgu raporunda 37 yaşındaki kadın hastanın panoramik radyografisinde posterior mandibulasında asemptomatik, mikst radyolüsent-radyoopak görünümlü lezyon sunulmuştur. Klinik ve radyografik muayene sonucunda fokal semento-osseöz displazi (FSOD) olduğu düşünülen lezyonun düzenli radyolojik takibi önerilmiştir. Lezyon yedi sene boyunca takip edilmiş ve radyografik olarak değerlendirilmiştir. Takip süreci boyunca asemptomatik olan lezyon ile ilişkili dişin de vital olduğu gözlenmiştir. Ayrıca, lezyonun içeriğindeki hiperdens alanın boyutunun arttığı ve ekspansiyonun minimal olarak arttığı konik-ışınlı bilgisayarlı tomografi görüntülerinde izlenmiştir.

Anahtar Kelimeler: fibroosseöz displazi, konik-ışınlı bilgisayarlı tomografi, sementoosseöz displazi

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INTRODUCTION

Cemento-osseous dysplasia (COD) is the most common benign fibro-osseous lesion of bone, in which normal bone is replaced by fibrous connective tissue, followed by calcification of the fibrous tissue with the formation of osseous and cementum-like tissue. Normal bone formation is replaced by abnormal bone or cementum (MacDonald, 2021). COD is usually seen as an incidental finding on x-ray examination. It is more likely to be observed in women (Nam et al., 2022).

Regarding lesion location in the jaw, COD can be categorised into three types: periapical, focal, and florid. Periapical COD is a mandibular dysplastic lesion occurring in the anterior region (Nelson and Phillips, 2019). Focal COD (FCOD) refers to lesions confined to one quadrant of the posterior mandible. In a more extensive form of COD, lesions can be found in more than one quadrant of the jaw and this is referred to as florid COD (Macdonald-Jankowski, 2008).

In many cases, FCOD is diagnosed incidentally during a routine dental examination. The adjacent teeth are vital. The options for treatment are assessed according to the presence or absence of symptoms. Primary FCOD is usually asymptomatic and does not require treatment. Secondarily infected COD may become symptomatic, and intervention is required if the condition becomes symptomatic (Bulut et al., 2012; Ravikumar et al., 2020).

In the osteolytic stage, it first appears as a radiolucent area. Patients may be misdiagnosed as having periapical lesions and undergo unnecessary root canal treatment or extraction. Due to bone repair through the defect, the radiographic pattern changes to a mixed pattern as the disease progresses. The lesion eventually develops into a radiopaque image with a rim of radiolucency in the mature stage. A well-defined border is present, but slight irregularity

may also be observed (Bulut et al., 2012; Cavalcanti et al., 2018; Min et al., 2018; Seifi et al., 2022).

This case report describes a 37-year-old female patient with FCOD. The lesion was followed up for seven years. The importance of follow-up and asymptomatic lesion characteristics are also reviewed.

CASE REPORT

The patient's informed consent was obtained for all clinical and radiographic examinations and for the presentation of this lesion.

A 37-year-old female patient was referred to the Outpatient Clinic of Marmara University, Faculty of Dentistry, Department of Oral and Maxillofacial Radiology, without any systemic diseases or daily medication use.

Intraoral examination revealed minimal expansion of the buccal aspect of the left mandibular first molar. There was no discolouration of the oral mucosa.

A radiolucent area with thickening of the molar root was noted as an incidental finding on panoramic radiography. The lesion was located at the apex of the left mandibular first molar and was surrounded by a radiolucent halo (Fig 1a). A detailed cone beam computed tomography (CBCT) scan revealed that the lesion involving left mandibular first molar was not completely hypodense, but was mixed and fused to the root (Fig. 1b). CBCT reconstruction showed destruction of the buccal cortical bone plate with minimal expansion. No root resorption or tooth displacement was observed. The dimensions of the lesion were determined to be as follows: 7.03 mm x 7.62 mm x 8.36 mm (Fig. 1c, d). The associated teeth were vital.

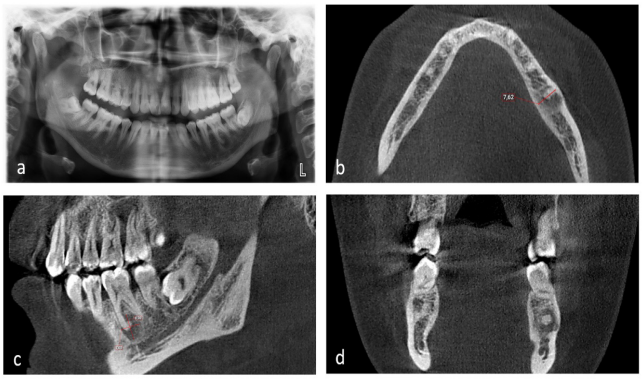

Figure 1: a. Panoramic radiography of the patient (year 2015) with a well-defined radiolucent lesion in the apex of the left mandibular first molar, **b**. CBCT showed minimal expansion in the axial plane and thinning of the buccal cortical plate, **c.** Hyperdense internal content adjacent to the molar root in sagittal plane, **d.** Thinning of the buccal cortical plate can be observed in coronal plane.

The preliminary diagnosis was hypercementosis associated with FCOD (MacDonald, 2021). Lesion was subjectively asymptomatic and the vitality test results, it was decided that no surgical or endodontic treatment was required. The patient was advised to be followed up radiologically.

The patient did not appear at the recommended time. The patient's second visit was five years after the first visit and the clinical and radiological examinations revealed a thicker mass at the apex of the left mandibular first molar. (Figure 2a). A hypodense area with radiolucent contents was observed in CBCT sections, centrally within the lesion (Fig 2b). The dimensions of the lesion were measured as follows: 7.35 mm x 8.79 mm x 8.88 mm and were compared with the initial CBCT measurements. A slight increase in size was noted (Fig. 2c, d). The lesion was still asymptomatic and the tooth was still vital. The patient was recommended for further follow-up.

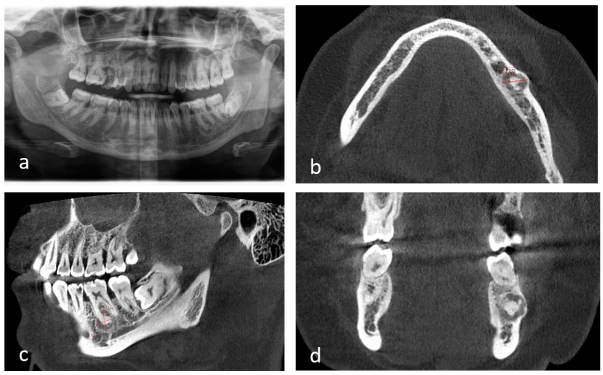

Figure 2: a. Panoramic radiography of the second follow-up session (year 2020), **b**. A well-defined mixed lesion was observed. The internal opaque contents were observed to be thicker compared to the initial radiograph, **c**. Minimally increased dimensions of the lesion, **d**. CBCT showed minimally increased expansion in the axial plane (Fig. 2b) and coronal plane and thinning of the buccal cortical plate in the axial and coronal planes.

Two years after the second visit, clinical and radiological evaluation showed a still asymptomatic, mixed (Fig. 3a), hypodense-hyperdense (Fig. 3b, c) lesion with minimally increased extent (Fig. 3d). Hyperdense enhancement was observed (Fig. 3c, d). The tooth was still vital. The dimensions of the lesion were as: 8.68 mm x 9.88 mm x 9.70 mm (Fig. 3b, c). Follow-up of the patient will be continued.

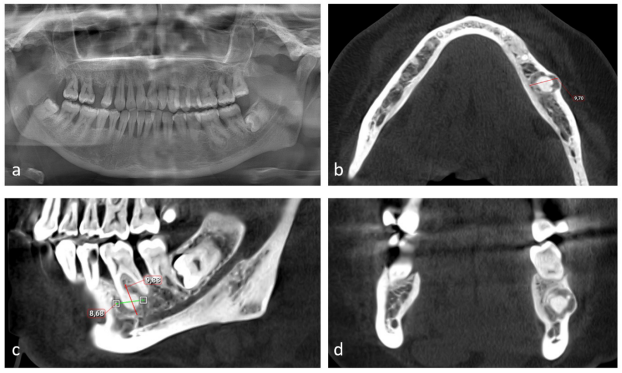

Figure 3: a. Panoramic radiography of the third follow-up session (year 2022), **b.** Increased hyperdense content and expansion in the buccal plate, **c.** Minimally enlarged dimensions of the lesion. No signs of infection. Lesion is not in close relation with mandibular canal. **D.** Coronal plane shows hyperdense lesion surrounded by hypodense area in the left mandibular first molar region with thinning of the buccal cortical plate.

DISCUSSION

COD is a non-neoplastic, mostly asymptomatic, reactive group of fibro-osseous lesions that occur in the tooth-bearing region of the jaw bone (Kato et al., 2020; Thakur et al., 2021). It is not clear what causes or contributes to COD, but the periodontal ligament is thought to play a role. A variety of factors may also be causative, including occlusion, caries, periodontal disease, infection, hormonal imbalances, and systemic diseases (Salvi et al., 2020). Women in their fourth or fifth decade have a predilection for this lesion (Min et al., 2018; Ravikumar et al., 2020).

This case presents a painless lesion of the root of the mandibular first molar in a female patient. As previously reported by Macdonald-Jankowski, FCOD is an incidental finding in the majority of cases (Macdonald-Jankowski, 2008). This case was also an incidental finding. In a previous report, Kawai et al. showed that of 54 patients, 59% had at least one symptom of inflammation, including pain, swelling, purulent discharge and tenderness on palpation (Kawai et al., 1999). Prevention of secondary infection requires good oral hygiene during follow-up. This is essential to maintain the asymptomatic state (Nam et al., 2022). If secondary infections occur as a result of inadequate endodontic treatment or tooth extraction, or for any other reason, these lesions may become symptomatic. When symptomatic, surgical excision is required (Ravikumar et al., 2020). Furthermore, poor healing, bone infection, or osteomyelitis have been reported to occur with any surgery that involves surgical excision of cemento-osseous dysplasia or extraction of the adjacent tooth. Because of the difficulty in managing these lesions, clinicians generally recommend observation rather than intervention (Kato et al., 2020; Thakur et al., 2021). In this case, the patient has had no complaints for seven years and no infection has been detected. The patient was followed up from 2015 to 2022 to ensure that the tooth remained asymptomatic.

Several reports have presented FCODs as well-defined mixed lesions (Macdonald-Jankowski, 2008; Cavalcanti et al., 2018; Min et al., 2018; Seifi et al., 2022). However, the radiological appearance of the lesion varies according to the stage of lesion maturation. The early or osteolytic stages are characterised by a well-defined radiolucency. Proliferative, immature FCOD lesions have a hypodense appearance, whereas later stages have a hyperdense content (Thakur et al., 2021). Accordingly, early lesions may be misdiagnosed as periapical cysts or rarefying osteitis (Salvi et al., 2020; Thakur et al., 2021). In the intermediate or cementoblastic stage, there is a characteristic radiolucent rim around the radiolucent area. The mixed appearance in further levels of the lesion may be confused with chronic sclerosing osteomyelitis, cemento-ossifying fibroma, odontoma and osteoblastoma. In the final stage, the tissue becomes more mature and osteosclerotic. This stage is characterised by sclerotic radiopacity with poorly defined borders (Summerlin and Tomich, 1994; Su et al., 1997; Alsufyani and Lam, 2011; Salvi et al., 2020). As the lesion progresses, the ratio of fibrous connective tissue to mineralised tissue ratio decreases. A thick curvilinear structure is formed by the bony trabeculae as they mature. In the final radiopaque stage, the individual trabeculae fuse to form lobular masses composed of relatively acellular and disorganised cemento-osseous sheets (Ohkura, 2001; Cho et al., 2007; Oh et al., 2019; Olgac et al., 2021; Gumru et al., 2021).

Early detection of the lesion and a detailed clinical and radiological examination are essential to make a correct initial diagnosis, leading to the decision to perform a biopsy, a total excision or a follow-up (Seifi et al., 2022). According to the review by Alsufyani and Lam (Alsufyani and Lam, 2011), the majority of cemento-osseous dysplasia cases were diagnosed in the mixed radiolucent-radiopaque stage (72.0%), and the internal radiopacities were described as dense and cementum-like (61.4%). This case represents a mixed stage after the initial osteolytic period. Furthermore, the maturing process was consistent with that reported in the literature and all stages of maturation have been presented (Kawai et al., 1999; Cho et al., 2007; Oh et al., 2019; Olgac et al., 2021).

Previous studies have shown that cortical plate expansion and thinning are common findings in periapical and florid cemento-osseous dysplasia, but rare in FCOD (Bulut et al., 2012; Cankaya et al., 2012; Rao et al., 2014; Cavalcanti et al., 2018; Kato et al., 2020; Thakur et al., 2021). Additionally, the most common age, gender, region and suggested / applied treatments of FCOD were presented in Table 1. In the present case, a minimal amount of expansion was observed in the buccal cortical plate of the mandible. A minimal thinning of the buccal cortical plate was also noted. Tooth extraction or surgery was not required due to the asymptomatic nature and minimal changes of the lesion. Surgery was considered unnecessary as long as the patient cooperated and the lesion was asymptomatic. The lack of histopathology may seem to be limiting, but the long-term observation nature of this case may have prevented the patient from above-mentioned complications (Drazić and Minić, 1999; Galgano et al., 2003; Cankaya et al., 2012; Bhandari et al., 2012; Rao et al., 2014). The patient is being followed-up.

CONCLUSION

FCOD is a benign fibro-osseous bone lesion, and most cases are asymptomatic. This case report has demonstrated a long follow-up in accordance with the principle of "first do no harm" and a complete process of maturation of the FCOD.

Table 1: Selected previous studies and case reports.

Author(s)	Year	Country	Case(s)	(Mean) Age	Sex (%)	Location	Treatment/ Suggestions
Su et al.	1997	USA	247	38	Female (86.6%)	Posterior mandible (61.5%)	Inadequate information
Dražić and Minić	1999	Yugoslavia	1	19	Male	Posterior maxilla	Biopsy / Radiological follow-up
Ohkura	2001	Japan	51	52	Female (86.2%)	Posterior mandible (2/3 of total cases)	Radiological follow-up
Galgano et al.	2003	Italy	1	47	Female	Anterior mandible	Excision / Radiological follow-up
Cho et al.	2007	Korea	33	55.9	Female (84.8%)	Mandible	Inadequate information
Bulut et al.	2012	Turkey	1	24	Female	Posterior mandible	Excision / Radiological follow-up
Cankaya et al.	2012	Turkey	1	69	Female	Posterior mandible	Excision / Radiological follow-up
Bhandari et al.	2012	India	1	25	Female	Posterior mandible	Biopsy / Radiological follow-up
Rao et al.	2014	India	1	47	Female	Posterior mandible	Excision / Radiological follow-up
Oh et al.	2019	Australia	29	35.7	Female (89.6%)	Posterior mandible	None / Radiological follow-up
Olgac et al.	2020	Turkey	83	40.36	Female (89.2%)	Posterior mandible	Biopsy / Radiological follow-up
Gumru et al.	2021	Turkey	55	46.84	Female (92.7%)	Posterior mandible (98.2%)	None / Radiological follow-up
Seifi et al.	2022	Iran	1	28	Male	Posterior mandible	Excision / Radiological follow-up
Ocbe and Yalcinkaya	2023	Turkey	1	37	Female	Posterior mandible	None / Radiological follow-up

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None

CONFLICT OF INTEREST

None

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