Rare disease

Focal cemento-osseous dysplasia of mandible

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Summary

Fibro-osseous lesions are disturbances in bone metabolism in which normal bone is replaced by a connective tissue matrix that then gradually develops into cemento-osseous tissue. Typically, the lesion is asymptomatic and is detected on routine radiographic examination. Radiologically, this lesion has three stages of maturation: pure radiolucent, radiopaque/mixed radiolucent, and radiopaque appearance. During these stages the lesion can be misdiagnosed. In this case report a 69-year-old patient with a complaint of painless swelling of the left mandibular molar and premolar area is presented along with a review of the differential diagnoses considered in order to reach a final diagnosis of focal cemento-osseous dysplasia.

BACKGROUND

Cemento-osseous dysplasias (CODs) are fibro-osseous lesions in which the normal bone architecture is replaced by fibroblasts and collagen fibres containing variable amounts of mineralised material.¹ Focal COD (FCOD) is a benign fibro-osseous lesion of the jaw. The posterior tooth-bearing regions are the usual sites of involvement; the maxilla is rarely affected.² The lesions are typically asymptomatic and are not life threatening, but surgeons should be aware of them. In this article, the case of a 69-year-old patient with FCOD is presented with a review of the literature.

CASE PRESENTATION

A 69-year-old woman was admitted to the University of Istanbul's Department of Oral Surgery for the evaluation of painless swelling in the left mandibular molar and premolar area. She was systemically healthy, and extraoral examination findings were within normal limits. Intraoral examination and palpation revealed a firm, painless, stable mass in the affected area. The patient was not wearing a mandibular denture, so a traumatic aetiology was excluded from the differential diagnosis. She was a non-smoker, recalled no medical history and never had been evaluated for any other fibro-osseous lesion in another part of her body.

INVESTIGATIONS

Panoramic radiographic and dental volumetric tomographic images revealed a 2-cm-diameter sclerotic mass with a radiolucent border and two tooth roots over the mucosa in the posterior left mandible (figure 1A–D).

Macroscopically, the specimen comprised two irregular, pinkish-yellow, hard tissue fragments (figure 2).

On microscopic examination, the lesion exhibited compact bony and cemental calcifications. Resorption and apposition lines could be seen in the tissue. Osteoblastic and osteoclastic cells were observed around the lesion and in the connective tissue islands (figure 3).

DIFFERENTIAL DIAGNOSIS

On the basis of the patient's clinical and radiographic findings, florid or periapical COD, chronic sclerosing osteomyelitis, ossifying/cementifying fibroma, osteoblastoma or odontoma were considered to be the cause of the lesion.

During surgery, according to the surgeons' experience a presumptive diagnosis of FCOD, possibly late-stage was made.

TREATMENT

Surgical intervention and contouring of the swelling were performed under local anaesthesia (figure 4A,B).

The lesion was curetted in two parts, and the curetted material was sent to the pathology department.

OUTCOME AND FOLLOW-UP

On the basis of these clinical, radiological and pathological findings, FCOD was diagnosed. A 6-month follow-up examination revealed that healing was uneventful with no recurrence (figure 5).

DISCUSSION

The current classification of bone-related lesions, released in 2005 by the WHO, is based on age; sex; histopathological, radiographic and clinical characteristics; and location of the lesion. The 2005 WHO edition preferred the use of 'osseous dysplasia' rather than the 'COD', which had been used in the second edition. This classification includes cemento-ossifying fibroma, benign cementoblastoma and the group of benign fibro-osseous lesions in which FCOD is included.³

The aetiology and pathogenesis of FCOD remain unknown, and this lesion is considered to be a reactive or dysplastic process in periapical tissues.⁴ Although the lesion is not considered to be odontogenic, it is undoubtedly related to the presence of teeth. Almost all FCODs appear above the mandibular canal and are thus confined to the alveolar process, suggesting at least some odontogenic influence on their genesis.³

Figure 1 (A) Panoramic view, showing the tooth and the sclerotic mass. (B) 3D view of the tooth apices and the lesion adjacent to it creating expansion towards the buccal bone. (C) Axial view of mandible showing the radioopaque mass surrounding by cortical bone. (D) Cross-sectional view of the lesion; radiolucent area around the sclerotic mass giving the appearance of encapsulated lesion.

Figure 2 Macroscopically two pieces of hard yellowish-reddish bone.

FCOD exhibits a single site of involvement and can occur in any tooth-bearing area, particularly in sites of a former extraction or an edentulous area of the mandible or maxilla, but generally it occurs in the posterior mandible and shows a greater frequency in women than in men (~10:1).^{5 6}

Typically, the lesion is asymptomatic and is detected on routine radiographic examination. Radiologically, this lesion has three stages of maturation: pure radiolucent, radiopaque/mixed radiolucent and radiopaque appearances.⁵ The early stage of FCOD classically presents with a well-defined radiolucency at the apices of mandibular teeth. The radiographic image may be erroneously diagnosed as an endodontic infection. In dentate areas, neither tooth displacement nor root resorption is observed. To assess the lesion, a pulp vitality test can be performed. FCOD lesions at an intermediate stage demonstrate a mixed radiolucent/opaque pattern, with a well-defined radiolucent rim around the radiopacity. At a later stage, these lesions display diffuse radiopacity, often with ill-defined borders and a greater proportion of anastomosing, thick curvilinear, poorly cellular bony trabeculae (the so-called 'ginger root' pattern).^{6 7}

FCODs are usually detected during routine radiographic examinations (periapical and panoramic x-rays), and these

Figure 3 Compact bony and cemental calcifications (black arrows), resorption and apposition lines (red arrows) (H&E, ×100 magnification).

Figure 4 (A) Intraoperative view of the lesion; the apices and the expansive mass adjacent to it having a slight yellowish colour that can easily be differentiated than bone. (B) Intraoperative view of the lesion; the conservative approach preserving both the buccal and the lingual cortical bone just as seen in the CT view.

methods are generally adequate for a correct prediagnosis. FCODs rarely cause cortical expansion.⁸ In these situations, cone-beam CT (CBCT) may help to distinguish the extension of the lesion through the cortical layers of the bone. In addition, CBCT can be useful to determine the radiodensity

Figure 5 Postoperative sixth month control panoramic view presenting the healing of bone.

(in Hounsfield units) of the lesion, which is equivalent to those of the cementum or cortical bone.³

The histopathological appearance consists of bone trabeculae and cementum-like material present within a vascular fibrous stroma, findings that are consistent with those of our case.

FCOD is predominantly seen in women, with a mean age in the mid-30s and a slightly higher incidence (64%) among African Americans.⁴ Most (62%) of these lesions are asymptomatic, and they can reach up to 1.5 cm in size.⁷ The mandible is the most frequent site of occurrence (86%).⁹ In the present case, our female patient's lesion was in the left premolar–molar area and was ~2 cm in diameter, which is consistent with findings presented in the literature.

Waldron and colleagues¹⁰ reported that most of their symptomatic cases occurred in edentulous areas, consistent with the finding of Ohkura¹¹ that FCOD was discovered more frequently in these areas. In our case, FCOD was also seen in an edentulous area at the end of a tooth apex.

Periapical cemental dysplasia and FCOD and florid CODs can generally be identified on routine radiographic examinations because of classic radiographic features and the fact that patients are usually asymptomatic. Thus, a biopsy is not usually necessary unless the lesion is infected and/or symptomatic.¹² However, infections sometimes seem to occur after the exposure of cemental masses by resorption of the edentulous alveolus or by extraction of teeth, the roots of which were close to the lesion.¹⁰ In our case, the patient was referred to us with a complaint of pain and swelling in the posterior mandible related to the root apices.

During the formulation of the differential diagnosis, the developmental stage of the lesion should be considered, and the possibilities should include periapical granuloma or cyst and chronic osteomyelitis in the osteolytic stage. In the mixed and radiopaque stages, florid or periapical COD, chronic sclerosing osteomyelitis, ossifying/cementifying fibroma, osteoblastoma and ameloblastoma should all be considered.^{13 14}

FCOD and periapical COD are different names for the same pathological process. Periapical COD is bilateral and is located at the root apices of the mandibular anterior teeth; it shows a predisposition for middle-aged black women. Multiple lesions frequently occur. Microscopic findings are identical to those of FCOD. FCODs, as in our case, are found throughout the alveolar process, but occur most commonly in the posterior mandible in women.⁶

Cemento-ossifying fibroma is an encapsulated benign neoplasm made up of highly cellular fibrous connective tissue that contains varying amounts of osteoid, bone, cementum and cementum-like calcified tissue. Most ossifying fibromas of the craniofacial skeleton grow slowly and symmetrically. They can result in bone expansion, with facial asymmetry and tooth displacement.^{15 16} Ossified fibromas are well-demarcated and show radiolucent features with small radiopaque calcifications. However, FCOD is usually radiopaque, as in our case, which exhibited a 2-cm-diameter sclerotic mass with a radiolucent border on panoramic radiography and CT.

Florid COD may be familial, with an autosomal-dominant inheritance pattern, and dysplastic bone and

cemental tissue is restricted to the jaws.¹⁷ The epidemiology of florid COD is nearly identical to that of FCOD, with a 90% female predominance, a strong predilection for black patients and a mean age at diagnosis of 42 years. The lesions demonstrate three stages of maturation, and most patients are asymptomatic, although some may complain of dull pain, sinus tracts or exposure of avascular bone into the oral cavity due to secondary infection.¹⁸ Florid COD is the most extensive form of this dysplasia, presenting with bilateral lesions in the tooth-bearing areas. Lesions of FCOD are smaller than those of florid COD, and usually present as a single solid lesion.¹⁵

Focal sclerosing osteomyelitis is characterised by an asymptomatic, non-expansile periapical lesion associated with a tooth exhibiting deep caries. The presence of inflammation is critical for the diagnosis of focal sclerosing osteomyelitis because this lesion can resemble several other intrabony processes that produce somewhat similar histological patterns. FCODs are usually not infected.

FCODs can be misevaluated as ameloblastomas during the intermediate stage, in which a mixed radiolucent/opaque pattern is seen. Ameloblastomas are locally destructive lesions that can affect the roots of the teeth and cause resorption, whereas FCODs generally show no root resorption.³

Learning points

- Surgical removal is unnecessary for asymptomatic focal cemento-osseous dysplasia (FCOD) after a final histopathological diagnosis, and follow-up is required to confirm the diagnosis. However, as in the present case, an FCOD should be removed from a patient with pain and other complaints.
- Some authors have suggested that transformation into florid COD is possible and should be considered at follow-up recall visits.

Competing interests None.

Patient consent Obtained.

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