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Extensive central ossifying fibroma of mandible: case report

Extenso fibroma ossificante central em mandíbula: relato de caso

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ABSTRACT

A 34-year-old female patient attended a referral center for oral diagnosis presenting hardened increased volume in the posterior mandible. Panoramic radiography revealed a mixed unilocular lesion with clear and well-defined limits. The incisional biopsy showed proliferation of mesenchymal cells amid dense fibrous connective tissue. Mineralized material in different degrees of maturation was also observed. Regarding the histopathological aspects associated with the clinical, radiographic, and intraoperative data, the diagnosis of central ossifying fibroma (COF) was performed. COF has similar characteristics to other fibro-osseous lesions, and its diagnosis is a challenge for dental surgeons.

Key words: ossifying fibroma; oral pathology; diagnosis.

RESUMO

Paciente do sexo feminino, 34 anos, compareceu a um centro de referência em diagnóstico oral apresentando aumento de volume endurecido em mandíbula posterior. A radiografia panorâmica revelou lesão unilocular mista, de limites precisos e definidos. A biópsia incisional evidenciou uma proliferação de células mesenquimais em meio a um tecido conjuntivo fibroso denso. Foi observado ainda material mineralizado em diferentes graus de maturação. Considerando os aspectos histopatológicos, associados aos dados clínicos, radiográficos e transcirúrgicos, o diagnóstico de fibroma ossificante central (FOC) foi revelado. O FOC apresenta características similares às de outras lesões fibro-ósseas, e seu diagnóstico é um desafio para os cirurgiões-dentistas.

Unitermos: *fibroma ossificante; patologia bucal; diagnóstico.*

RESUMEN

Paciente de 34 años de edad, acudió a un centro de referencia odontológico con aumento de volumen endurecido en la mandíbula posterior. La radiografía panorámica reveló lesión unilocular mixta, de límites precisos y definidos. La biopsia incisional demostró una proliferación de células mesenquimales en medio de un tejido conectivo fibroso denso. Aún se observó material mineralizado en diferentes grados de maduración. Considerando la histopatología, asociada a los datos clínicos, radiográficos y transquirúrgicos, el diagnóstico de fibroma osificante central (FOC) se reveló. El FOC presenta características similares a las de otras lesiones fibro-óseas, y su diagnóstico es un desafío para los cirujanos dentales.

Palabras clave: *fibroma osificante; patología bucal; diagnóstico.*

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INTRODUCTION

The central ossifying fibroma (COF) was first described in 1842 by Menzel and received this name by Montgomery in 1927[1-3]. It is a rare benign fibro-osseous neoplasia that predominantly affects the craniofacial region[4]. This lesion corresponds to the clinicopathological variant of ossifying fibromas of odontogenic origin, and can also be called cementifying fibroma or cemento-ossifying fibroma[5].

COF has a peak incidence between the third and fourth decades of life[1, 2, 6]. It has a predilection for women, with a female to male ratio of 5:1; the mandible represents the most affected site, with an incidence of 70%-90% of cases[5, 7].

The final diagnosis of COF is based on a detailed analysis of the medical history, radiographic features, intraoperative findings, and histopathological characteristics, since the fibro-osseous lesions present significant overlap in the histological appearance; an appropriate correlation of all these characteristics is essential to achieve a reliable diagnosis[8, 9].

COF treatment includes enucleation and curettage; for larger lesions, surgical resection is indicated[7]. The recurrence rate of these lesions in the mandible is around 6% to 28%. Due to the difficulty of maxillary surgical removal this rate is unknown, but it is believed to be higher[2].

This study aimed to report a case of COF in the mandible, evaluating and emphasizing the importance of clinical, imaging, intraoperative, and histopathological information for the correct diagnosis of benign fibro-osseous lesions.

CASE REPORT

A 34-year-old female patient attended to a dental appointment complaining about increased volume in the mandibular region. On intraoral physical examination an increase asymptomatic hardened slow growing volume was observed in the vestibular region of elements 46, 47, and 48, normal colored mucosa; the average time of evolution was not informed. The extraoral physical examination revealed normal regional lymph nodes.

Panoramic radiography revealed a unilocular mixed lesion, with radiopaque foci, well-defined limits, bone expansion, and displacement of the mandibular canal (**Figure 1**). Based on this finding, an incisional biopsy was performed; the dentist reported easy detachment of the lesion from the adjacent healthy bone. Surgical specimens were sent for histopathological analysis.

Numerous fragments of brown-yellow color have irregular appearance, fibrous and hardened consistency was evidenced in the macroscopic examination.

Histopathological analysis revealed proliferation of mesenchymal cells with morphology varying from spindle to oval shapes amid a dense fibrous connective tissue. Mineralized material was also observed in different degrees of maturation, sometimes forming trabeculae composing entrapped osteocytes and osteoblastic surface layer, sometimes forming basophilic spherules (**Figure 2A-D**).

Therefore, the histopathological findings associated with clinical and radiographic aspects and the intraoperative information allowed the final diagnosis of COF. The patient underwent complete enucleation and curettage of the lesion, after six months, she is currently monitored, with no signs of recurrence.

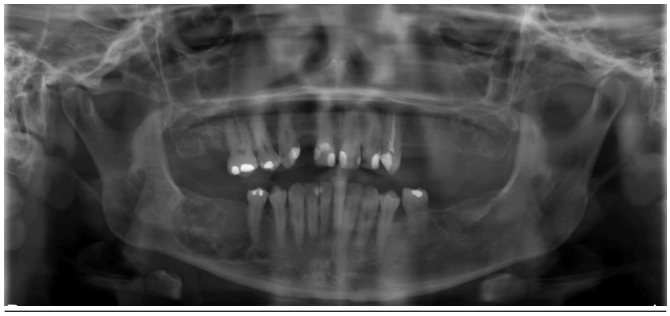

FIGURE 1 – *Radiographic findings of the lesion. Radiolucent image showing radiopaque foci, with well-defined limits, in the right region of the mandibular body*

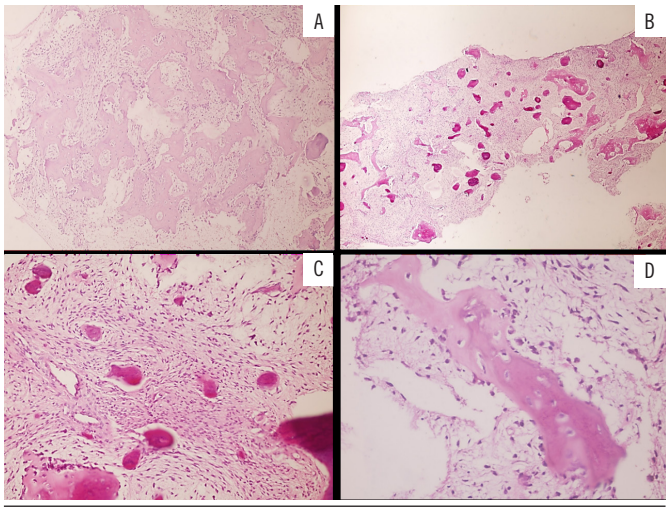

FIGURE 2 – *Histopathological characteristics*

A and B) images in lower magnification showing the presence of mineralized materials in different degrees of mineralization, sometimes forming trabeculae similar to bone tissue (100×) (A), sometimes forming basophilic spherules similar to cementoid material (B); C) stroma of richly cellularized dense fibrous connective tissue showing basophilic spherules (200×); D) trabecular bone showing apparent osteoblastic surface layer (400×).

DISCUSSION

Menzel first described a variant of COF in the mandible in 1842[4]. In 1972, the World Health Organization (WHO) classified COF as cement-forming lesions, which also included fibrous

dysplasia[10]. However, the latest WHO classifications, from 2005 to the last in 2017, changed the term cemento-ossifying fibroma and central ossifying fibroma to ossifying fibroma[5, 11]. Despite this, many authors prefer the term COF to differentiate it from its variant that occurs in soft tissues: the peripheral ossifying fibroma, which is considered a reactive non-neoplastic lesion.

Therefore, COF is considered a true neoplasm, of odontogenic origin, specifically from the periodontal ligament, which is composed of multipotent mesenchymal stem cells capable of forming bone, fibrous tissue, and cement. There is an assumption that this lesion would be the result of some stimulus induced by previous dental extraction or some other traumatic agent, or would simply be linked to a disturbance of bone maturation of congenital origin. However, there is no scientific evidence to justify and support this hypothesis[2, 7, 12].

The epidemiological profile of COF has been extensively studied over the years; it occurs mainly in adult women and in the posterior region of the mandible, especially in the premolars and molars region[13].

During clinical analysis, these lesions appear as an slow-growing increased volume, usually with no symptoms; facial asymmetry is rare[7]. When they exhibit extensive dimensions, they can displace tooth roots, cause root resorption of adjacent teeth, and expand the lower edge of the mandible or the floor of the maxillary sinus[6].

Therefore, this case is in line with the demographic characteristics of gender, age, and lesion location, classically described for this neoplasm. The clinical presentation and the pattern of tumor growth observed also fit the descriptions reported in the literature, which refer to slow and asymptomatic tumor growth[3].

Radiographically, COFs appear as single well-defined radiolucency with radiopaque foci, whose radiopacity varies according to the amount of mineralized material that has been deposited. On the other hand, it is important to highlight that there is no relationship between intensity of mineralization and tumor growth rate, not even with the time of evolution of the lesion[2, 14]. Eversole *et al*. (1985)[15] described two main radiographic patterns: expandable unilocular radiolucency and multilocular configuration. These types tend to expand the surrounding cortical bone without perforating the cortical, featuring a centrifugal growth pattern, with equal expansion in all directions. This is a diagnostic feature for COF[4].

A retrospective study by Titinchi and Morkel (2016)[1] revealed almost 49.2% of COF lesions as radiopaque, 34.9% as mixed, and only15.9% as radiolucent. Furthermore, 84.1% of the lesions were unilocular on panoramic radiographs and only 15.9% of the cases were multilocular; these lesions were more prominent in the lower posterior regions and in patients younger than 20 years. The authors also found about 93.6% of lesions with well-defined margins, easily differentiating them from the healthy bone. The results of this work corroborate the imaging findings of the case presented, which demonstrated a mixed unilocular pattern in the well-defined posterior mandibular region, with easy detachment of the adjacent healthy bone.

Several lesions may show clinical and radiographic features similar to COF. The differential diagnosis is essential to provide strategic data for establishing the final diagnosis. Fibrous dysplasia represents the main lesion to be distinguished radiographically from COF, but it is classically observed as a lesion consisting of diffuse and irregular radiopacity, with "ground-glass" appearance and very imprecise margins, often found in the posterior region of the maxilla[4, 13]. Also some lesions do not differ from COF by radiographic examination, such as calcifying odontogenic cyst and calcifying epithelial odontogenic tumor; thus, histopathological characteristics are essential for the final diagnosis[8].

Histologically, COF is presented as a proliferation of fibrous cellular tissue that contains a variable amount of mineralized material. The degree of cellularity varies widely, with hypocellular and collagenized areas that merge with hypercellular areas. The mineralized material may be presented as bone trabeculae or basophilic ovoid cementum-like structures. Tumors usually have a combination of these calcified structures. COF bone trabeculae may still present surface layer of osteoblasts[8, 9].

The histopathological diagnosis of COF in the absence of solid clinical and radiographic information is a challenge, even for experienced pathologists. This difficulty occurs due to histopathological similarities found between COF and other fibro-osseous lesions, such as fibrous dysplasia (FD) and osseous dysplasia (OD)[16]. In FD, osteoblastic surface layer is usually absent; fibrous connective tissue contains less cellularity; there is a minimal amount or absence of spherical basophilic mineralized deposits; and the lesion is present in continuity with the adjacent bone, unlike the COF. Whereas, OD presents stroma with hemorrhagic foci; osteoblastic surface layer is scarcely observed; and the lesion is in continuity with the adjacent bone, as well as in the FD. However, cementoid masses are irregular, with no brush border, which differentiates it from COF, whose masses are generally ovoid, with brush border[8, 13].

It is difficult to obtain an accurate final diagnosis of COF, as errors can arise when all characteristics are not carefully considered, leading to therapeutic and prognostic effects[17]. Some authors consider that the histopathological diagnosis with no clinical and radiographic correlation is only possible if the

biopsy sample includes the interface between lesion and healthy adjacent tissue. However, in the absence of clinicoradiographic information, it is very common that the pathologist is unable to present a definitive diagnosis, preferring to assign the biopsy as a benign fibro-osseous lesion[18]. Unfortunately for the clinician, this non-specific diagnosis does not provide much guidance to direct him to appropriate treatment[8].

CONCLUSION

COF has similarities with other benign fibro-osseous lesions, and its diagnosis is often a challenge for dental surgeons. Therefore, a reasonable correlation of clinical, radiographic and histopathological features is necessary to establish a definitive diagnosis and an appropriate surgical intervention.

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