

Central Cemento-Ossifying Fibroma: Clinical - Imaging and Histopathological Diagnosis

Fibroma Cemento-Osificante Central: Diagnóstico Clínico, Imagenológico e Histológico

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WANZELER, A. M. V.; ROHDEN, D.; ARÚS, N. A.; SILVEIRA, H. L. D. & HILDEBRAND, L. C. Central cemento-ossifying fibroma: clinical-imaging and histopathological diagnosis. *Int. J. Odontostomat.*, 12(3):233-236, 2018.

ABSTRACT: Central ossifying fibroma is a lesion in which diagnosis has proved intriguing and unclear for presenting clinical, radiographic, and even histopathological similarities to other types of lesions such as the fibrous dysplasia of bone and cement-bone dysplasia. It is a benign neoplasm of uncertain etiology and slow development in which the mandible is affected more frequently than the maxilla. We described a case of central cemento-ossifying fibroma involving the right mandible of a thirty-five-year-old female patient by presenting its clinical, radiographic, and histological characteristics and discussing some differential diagnoses.

KEY WORDS: benign neoplasm, central ossifying fibroma; oral pathology; differential diagnosis.

INTRODUCTION

The cemento-ossifying fibroma has been referenced as ossifying fibroma or cementifying fibroma (El-Naggar *et al.*, 2017; Wright & Vered, 2017), but in the fourth issue of the World Health Organization (WHO) for the classification of tumors in the head and neck, published in January 2017, the term "cemento-ossifying fibroma (COF)" (El-Mofty, 2014) was added. The COF has always given way to some controversy regarding terminology and diagnosis criteria (Soluk-Tekkesin & Wright, 2018) included in the group of odontogenic mesenchymal benign tumors. Its pathogenesis remains unknown, but it can be related to congenital issues during the maturation of the dental tissues, which are able to form cement and bone tissue (Chang *et al.*, 2008; Bala *et al.*, 2017).

Clinically, it generally shows as a volumetric asymptomatic increase of slow evolution which affects the region of premolars and molars in the mandible (Chang *et al.*; Trijolet *et al.*, 2011; White & Pharoah, 2014), while radiographically it manifests as a well-defined lesion with radiotransparent and or radiopaque areas which may be associated with divergences or root resorption (Agarwal *et al.*, 2012; Abbas *et al.*, 2017). Treatment consists of surgical removal with a

mostly favorable prognosis (Shimamoto *et al.*, 2011; Sopta *et al.*, 2011). The following case report describes the clinical-imaging and histopathological diagnosis of a central cemento-ossifying fibroma.

CASE REPORT

A thirty-five-year-old female patient without any other relevant medical record entered the service of Pathology of the Dentistry School of the Federal University of Rio Grande do Sul (FO-UFRGS) claiming she had been feeling pain for one year and presenting swelling on the right side of her face. The swelling seemed to grow gradually and had reached its current size at the time, over the past six months. The extraoral physical examination revealed slight facial asymmetry resulting from swelling in the region of the mandible body in the right side as well, with regular color and texture while palpation showed a hard, bone consistency. Intraorally, we observed an increase in the alveolar ridge volume with the expansion of the buccal and lingual cortical; hard palpation, covered with normal-color full mucosa, and absence of the tooth 46 (Fig. 1).



Fig. 1. Increase in the alveolar ridge volume with the expansion of buccal and lingual cortical in the region of lower molar on the right side.



Fig. 2. The panoramic X-ray reveals a well-defined mixed multilocular lesion in the region of the right mandibular body involving the peaks of the teeth 47 and 48, causing external root resorption. We can see the displacement of the mandibular canal to the jaw base.

The digital panoramic X-ray revealed a well-defined radiolucent multilocular lesion in the body of the mandible on the right side extending to the ramification (Fig. 2). The lesion extended beyond the lower limit of the mandible with expansion and grinding of the cortical plates. The internal structure of the lesion is predominantly radiolucent with a diffuse dispersion of radiopacity. The hypotheses of diagnosis included central lesion of giant cells, fibrous dysplasia of bone, and ossifying fibroma.

We proceeded with an incisional biopsy in which both the soft tissue and the bone tissue were removed from the affected region. After the histopathological result, the lesion was diagnosed as central ossifying fibroma and followed by our decision to carry out an excision biopsy with curettage of the lesion and extraction of the teeth involved (Fig. 3). The piece removed measured 20mm at the largest diameter. Another histopathological examination was conducted and confirmed the diagnosis of central ossifying fibroma (Fig. 4).

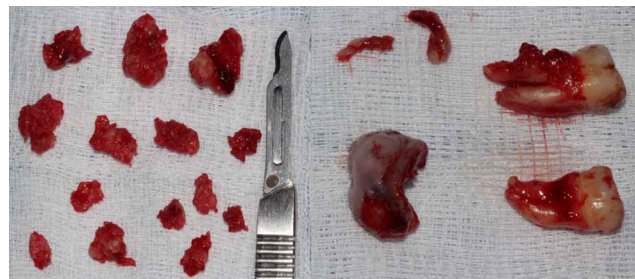


Fig. 3. Surgical piece and the teeth involved in the lesion (47 and 48).

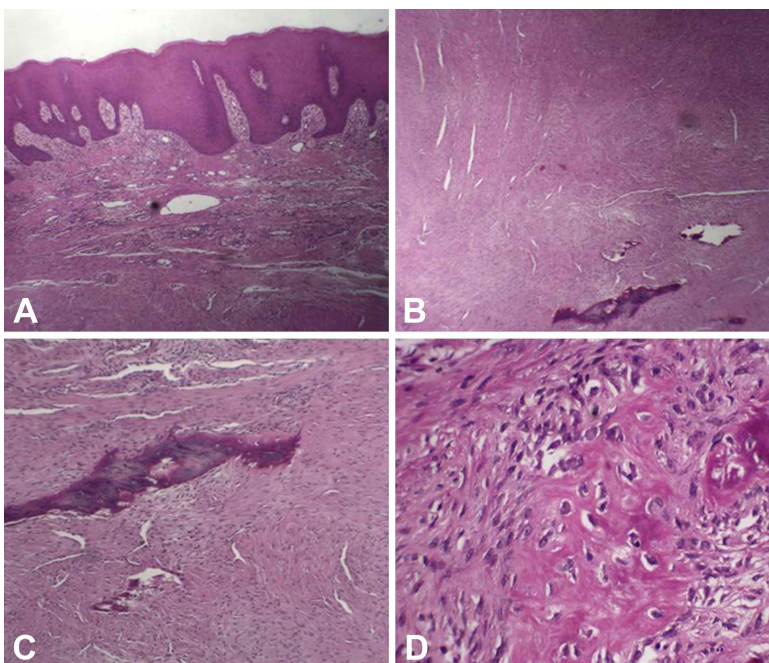


Fig. 4. Histopathological aspects indicate a) higher increase of the epithelial tissue of the mucosa coating, connective tissue fibrous of the submucosa, and tissue proliferation of the lesion; b) highly cellularized connective tissue and calcified material (osteoid), c) tissue proliferation in the storiform pattern, and d) non-mineralized osteoid trabecula

DISCUSSION

The cemento-ossifying fibroma was initially classified by the WHO as a fibrous neoplasia (El-Naggar *et al.*; Wright & Vered); however, it does not appear on the long bones, but especially in the areas of maxilla teeth. It is similar to some cemento-bone dysplasia, which suggests an odontogenic origin (Trijoleit *et al.*) and derives from mesenchymal cells showing a potential to form fibrous tissue, cement, bone or a combination of such elements (Agarwal *et al.*).

It occurs especially in patients aged from 20 to 40 years old, even though it may also manifest in children and adolescents, in addition to older adults (White & Pharoah). Women are more commonly affected than men ranging a proportion of 5: 1 (Chang *et al.*) by frequently affecting the posterior region of the mandible (Trijoleit *et al.*; El-Naggar *et al.*). The report presented occurred in a thirty-five-year-old woman and was located in the region of the mandible body on the right side.

The lesions are small and asymptomatic and cause a painless swelling as they grow and expand (Dominguete *et al.*, 2014; da Silveira *et al.*, 2016). In the reported case the patient felt pain and had a swelling on the right side of her face. Radiographically the early lesions may appear as uni- or multilocular radio transparency (Sopta *et al.*; Agarwal *et al.*; Soluk-Tekkes in & Wright). The early stage progresses gradually to a mixed lesion as the calcified material settles in the tumor (White & Pharoah; da Silveira *et al.*; Bala *et al.*). Mature lesions may consist of a dense radiopaque mass surrounded by a well-defined radiotransparent line (Titinchi & Morkel, 2016; Bala *et al.*). Considering the radiographic characteristics, the reported case shows well-defined mixed lesion. In general, displacements and reabsorption of the tooth roots may appear (Chang *et al.*; Oliveira *et al.*, 2008; El-Mofty; Bala *et al.*), such as in the case reported. Radiographically it is also shown the displacement of the mandibular can alto the mandible base, which are characteristics that suggest a benign lesion.

The CFO differential diagnosis includes lesions with a radiolucent-radiopaque internal structure (Oliveira *et al.*). It can be difficult to distinguish a fibrous dysplasia of bone; the limits of a CFO lesion are normally well-defined and occasionally present a fibrous and a cortical capsule, while a fibrous dysplasia often mixes with the adjacent bone (Agarwal *et al.*).

The internal structure of a fibrous dysplasia of bone can be more homogenous and show less variations (Shimamoto *et al.*; Abbas *et al.*).

In turn, the central granuloma of giant cells (CGGC) was introduced as a feasible diagnostic hypothesis for presenting radiographically well-defined limits in addition to in some cases having as light granular pattern of a very thin calcification internally with poorly-defined internal septa. The CGGC normally displaces or resorbs tooth roots. The lesion develops more often in the mandible by affecting especially adolescents and young adults aged below 20 years old (White & Pharoah). The case reported involves a thirty-five-year-old patient whose final diagnosis was confirmed through histological analysis.

The histopathological report confirmed the lesion as benign and compatible to a cemento-ossifying fibroma scenario whose characteristics are fusocellular proliferation of fascicular or diffuse organization alternately, immersed in the stroma of fibrous connective tissue moderately collagenized and vascularized, in addition to an alternately trabecular and psammomatoid position of mineralized tissue.

The prognosis is favorable (Chang *et al.*) upon adequate treatment (Sopta *et al.*), which is commonly agreed to consist of surgical removal of the lesion (White & Pharoah). Many authors describe that either the enucleation or curettage of the lesion is the initial choice of treatment for cemento-ossifying fibroma (da Silveira *et al.*). Whenever the lesion leads to a greater damage of structures, the implementation of treatment to remove the lesion with a five-millimeter safety margin is indicated (Abbas *et al.*; Soluk-Tekke in *et al.*). For the reported case, the lesion was treated through curettage because of its size.

CONCLUSION

The diagnosis of the central cemento-ossifying fibroma is oriented by the clinical and radiological aspect of the lesion; however, the histopathological aspect corroborates the diagnosis. Its treatment is surgical, but curettage or enucleation are indicated for smaller sizes, while larger lesions need surgical resection with a safety margin – favorable prognosis and low recurrence rate.

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RESUMEN: El fibroma cemento-osificante central es una lesión que requiere un diagnóstico diferencial ya que muestran similitud clínica, histológica y radiológica con la displasia fibrosa y con la displasia cemento-ósea. Esta lesión es un tumor benigno de etiología incierta, presenta crecimiento lento y afecta principalmente la mandíbula más que el maxilar. Se reporta un caso de un paciente de sexo femenino de 35 años, diagnosticada con fibroma cemento-osificante central que le afectó el lado derecho de la mandíbula. Se describen las características clínicas, histológicas y radiológicas de la paciente y se discuten los diversos diagnósticos diferenciales.

PALABRAS CLAVE: Neoplasia benigna; Fibroma cemento-osificante; patología bucal, diagnóstico diferencial.

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Received: 26-03-2018

Accepted: 22-05-2018