

# A distinct fibro-osseous lesion of the jaws affecting the maxilla

Wilber Edison Bernaola-Paredes<sup>1</sup>, Norberto Nobuo Sugaya<sup>1</sup>, Mariana Lobo Bergamini<sup>2</sup>, Paulo Henrique Braz-Silva<sup>3,4</sup>

Departments of <sup>1</sup>Oral Diagnosis, <sup>2</sup>Oral Pathology and <sup>3</sup>Stomatology, School of Dentistry, University of Sao Paulo, <sup>4</sup>Department of Virology, Institute of Tropical Medicine of São Paulo, University of São Paulo, São Paulo, Brazil  
Pedro Luiz Duarte De Sá equally contributed to this article

## Abstract

The differential diagnosis of fibro-osseous lesions (FOL) presents oral clinician stomatologists with a challenging task because of their similar clinical, radiographic and histopathological behavior that will also hinder the therapeutic planning. Presentation of case to describe the clinical, radiographic and histopathological features of an unusual case of FOL presented by the patient, a Black woman, and the results obtained with the conservative surgical treatment and 3-month follow-up. The presence of cementum in the histopathological analysis of cemento-osseous dysplasias, according to the current literature, is an important factor for the diagnosis of this lesion. Considering the characteristics presented by this particular case, we could suggest another entity that could be named a benign cemento-osseous lesion with adult onset. Some occurrences, as in the present case, do not fit properly into the current World Health Organization classification, thereby generating some doubts concerning the correct management of these patients. Conservative surgical treatment is the first choice in the management of these lesions due to their self-limiting nature, which show a behavior of clinical involution.

**Keywords:** Bone tumors, cemento-osseous dysplasia, conservative management, fibro-osseous lesions, surgical treatment

**Address for correspondence:** Prof. Wilber Edison Bernaola-Paredes, Department of Oral Diagnosis, School of Dentistry, University of Sao Paulo, São Paulo, Brazil.  
E-mail: ebernaola@usp.br

**Received:** 22.11.2019, **Accepted:** 16.01.2020, **Published:** 28.02.2020

## INTRODUCTION

The main characteristic of fibro-osseous lesions (FOLs) is the replacement of normal bone by fibrous connective tissue, which gradually becomes mineralized.<sup>[1]</sup> However, an extensive diversity of diseases is included in this group of lesions, thus requiring accurate diagnosis and individualized treatment.

Histopathological features are essential for a correct diagnosis of FOL but not sufficient *per se*, therefore

demanding additional clinical and imaging characteristics to enable a definitive diagnosis to be made<sup>[2]</sup> such as ossifying fibroma, florid cemento-osseous dysplasia (COD), focal COD and fibrous dysplasia (FD) and even a low-grade osteosarcoma.<sup>[2,3]</sup>

Regarding the present classification of head-and-neck tumors, defined by the World Health Organization (WHO, 2017) in relation to the FOLs and osteochondromatous lesions of the head and neck, the term “cementum,” which was

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10.4103/jomfp.JOMFP\_333\_19

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**How to cite this article:** Bernaola-Paredes WE, Sugaya NN, Bergamini ML, Braz-Silva PH. A distinct fibro-osseous lesion of the jaws affecting the maxilla. J Oral Maxillofac Pathol 2020;24:S23-7.

removed from the 2005 classification, was reinserted in order to recognize these lesions as being of odontogenic origin, specifically from the periodontal ligament. Three variants are still recognized: periapical COD, focal COD and florid COD.<sup>[4]</sup>

CODs originate from the periodontal ligament; they can affect a single tooth or a region involving a larger number of teeth.<sup>[5]</sup> Another important feature is that this lesion has a self-limiting behavior.<sup>[6]</sup> Radiographically, lesions at an early stage or those considered immature show a radiolucent image, whereas more mature lesions have a mixed or completely radiopaque appearance.<sup>[5]</sup>

In the case of CODs, the histopathological features<sup>[2]</sup> consist of mesenchymal tissue fragments composed of fusiform fibroblasts and collagen fibers, as well as presence of variable amounts of bone and cementum. As the lesion evolves to a mature stage, connective tissue expression decreases and mineralization becomes more prominent, sometimes being described as “ginger roots.”<sup>[7]</sup>

CODs show an important and considerable predilection for Black women, especially in the fourth and fifth decades of life.<sup>[5]</sup> Most cases are asymptomatic, are diagnosed on routine radiographic assessments and rarely present expansion. The cases that manifest some discomfort are mostly related to secondary infections.<sup>[6]</sup> These lesions most commonly affect the anterior and premolar regions of the mandible, whereas they are more rarely found in the maxilla.<sup>[6]</sup>

CODs require no treatments other than clinical and radiographic follow-up, unless they are complicated by infection and osteomyelitis. Periodic prophylaxis, reinforcement of oral hygiene, maintenance of a healthy periodontal condition and adequate control of removable prosthesis in use are procedures recommended to prevent complications.<sup>[8]</sup>

### CASE REPORT

The patient, a 46-year-old afro-descendant brazilian woman, who consulted our outpatient clinic, reported abnormal

growth in the posterior region of her right maxilla; it was asymptomatic and had an evolution time of 5 years [Figure 1]. Intraoral examination revealed a swelling in the posterior region of the upper jaw with a bony consistency, smooth surface, asymptomatic and normal soft-tissue covering. A slight asymmetry of the right hemi-face could be clinically observed. Radiographic assessment revealed a poorly delimited radiopaque lesion, with the appearance of ground glass, involving the region from the premolar up to that of the tuberosity. A root remnant of tooth 16 showing periapical radiolucency could be also observed in a close relationship to the dysplastic lesion [Figure 2]. Other residual roots, teeth in poor condition and an extensive deformity produced by the dysplastic bone could be noted in the panoramic X-ray that showed evidence of a remarkable expansion of the right maxilla in all directions, in addition to projecting downward into the space provided by the absence of mandibular molars on that side.

A clinical diagnosis of FOL led to a surgical procedure involving the removal of the remnant root and collection of a sample of the surrounding bone tissue for histopathological analysis. The consistency of the tissue removed was much softer than that of normal bone, suggesting a high content of fibrous component.

Histopathological examination revealed connective tissue fragments with immature bone tissue, trabeculae and spherules of material similar to dental cementum, providing the diagnosis of COD as shown in Figure 3.

The microscopic image of the benign FOL and the nature of the intraoperative tissue observed in the biopsy procedure allowed a surgical intervention to be performed with the aim of recontouring the buccal aspect of the affected maxilla to achieve an improvement of patient’s facial symmetry, as may be visualized in Figure 4.

Histopathological assessment after surgical osteoplasty, reassured the diagnosis of COD [Figure 5].

At 3-month follow-up consultation, the patient showed an optimal condition, without signs of infection and/or



**Figure 1:** Clinical diagnosis: (a) clinical features of extraoral and (b) intraoral evaluation in the patient, a afro-descendant brazilian woman), who showed (c) swelling on the right hemi-face

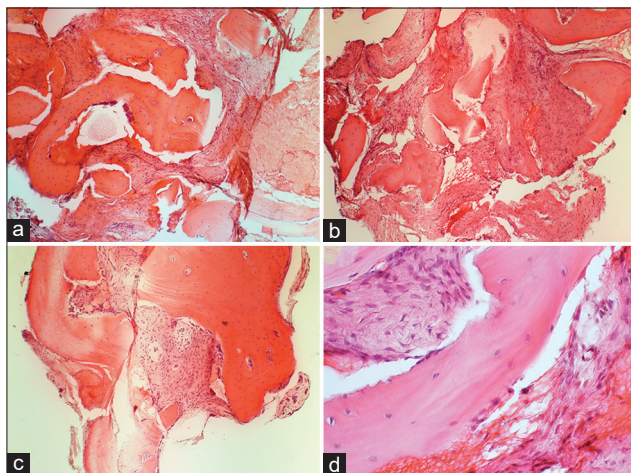
recurrence of the lesion. A second surgical intervention is being planning to achieve an ideal field for prosthetic rehabilitation of the patient [Figure 6].

## DISCUSSION

The main purpose of this case report was to discuss the clinical, radiographic and histopathological features that the case presented, which apparently did not fit into the last WHO classification of FOLs of the jaws.<sup>[4]</sup>



**Figure 2:** Imaging method: poorly delimited radiopaque image associated with a molar showing a periapical radiolucency



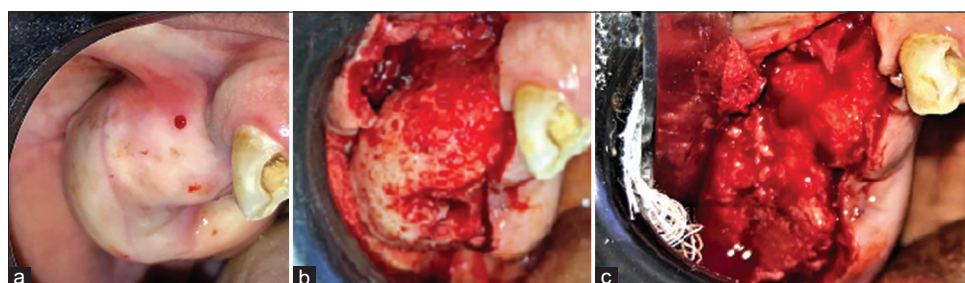
**Figure 3:** Staining method: (a) histological sections showing trabeculae of immature bone surrounded (b) by fibrous tissue and (c) osteocytes trapped in small gaps, as well as structures (d) resembling dental cementum tissue

The patient, a Black woman in her forties, sought assistance in order to resolve her facial asymmetry caused by a maxillary asymptomatic swelling of 5 years' duration. The panoramic radiographic showed a lesion with a classical fibro-osseous appearance compatible with a FD of the jaws: in the maxilla, with the appearance of ground glass and asymptomatic expansion. The obstacles to this hypothesis were the patient's age and duration of the lesion, which would correspond to an FD with the onset of its evolution in the fifth decade of the patient's life.

The COD fitted in with the sex, age range and ethnicity of the patient but was far more common in the mandible and rarely caused expansion at the level observed in the case presented. Other factors that opposed the hypothesis were relative to the homogeneous and diffuse radiographic appearance (if a focal COD was being considered because the case did not fulfill the criteria for classification as a periapical or florid COD) and the fibrous consistency of the lesion observed intraoperatively. Characteristically, focal COD in its radiopaque phase shows a very hard consistency due to a lack of trabecular spaces and vascularization that makes this type of lesion susceptible to the development of infections and osteomyelitis, and in the case presented, a dental root remnant with a periapical lesion was evident, in close relationship with the lesion, causing no symptoms at all.

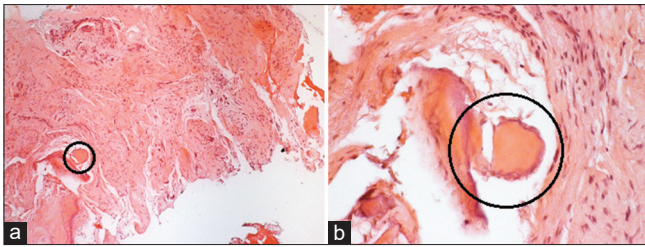
A familial gigantiform cementoma was completely out of the question, in this case, because it is characterized by huge and early expansive lesions affecting all quadrants of the maxillofacial region.

An ossifying fibroma could be considered due to the radiographic aspect and the extensive expansion; however, this odontogenic neoplasia usually affects younger patients, is more common in the mandible and is clearly delimited from the surrounding bone. The radiographic appearance was not compatible with these characteristics, and the surgical intervention did not disclose any type of capsule or delimitation between the marginal bone and the pathological tissue. Moreover, the cementum tissue



**Figure 4:** Surgical approach: (a-c) intraoperative aspect of the lesion site





**Figure 5:** Histopathological analysis: (a) presence of cementum-like tissue associated with fibrous dysplastic tissue. (b) Higher magnification to enable visualization of cementum surrounding the fibrous tissue

identified in the microscopic slides of the case presented, ruled out this diagnosis.

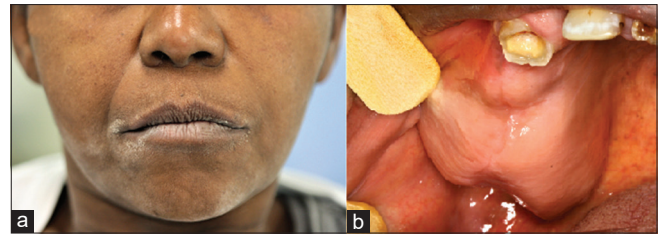
Some reports have described recurrences that are more aggressive after surgical interventions in FOLs (mainly immature FDs and gigantiform cementomas), which led us to adopting a conservative approach in this unusual case, by limiting the surgical recontouring to the buccal aspect of the lesion. The facial asymmetry was the patient's main concern. As no sign of recurrence was noted 3 months after the intervention, more comprehensive plastic surgery is being planned.

Kato *et al.*, 2018,<sup>[9]</sup> reported a large case series of 383 FOLs, using data obtained from a Brazilian laboratory service. COD accounted for 48.8% of the cases and 31.6% presented signs of swelling. Unfortunately, there were no data concerning the extent of the aspect of growth and a significant percentage of cases were associated with osteomyelitis (30.5%).

The diagnosis of FOLs is often difficult to establish without the sum of all data from microscopic studies, imaging characteristics and clinical assessment. Even areas of malignant osteoid neof ormation in the case of low- to high-grade osteosarcoma may resemble a benign FOL.<sup>[3,10,11]</sup> The presence of cementum and/or cementum tissue in the areas associated with proliferative fibrous tissue is a key feature that differentiates CODs from FD and other osseous tumors.<sup>[5]</sup>

Since the last edition of the WHO classification of head-and-neck tumors (2017), the denomination of COD was reinstated, with the aim of clearly establishing the odontogenic origin of the lesion.<sup>[4,10]</sup>

An interesting clinical aspect in this case was the relatively extensive deformity that the expansion process produced, associated with the rather classical radiological dysplastic aspect of FD of the jaw. Focal and florid CODs are not characterized by large expansion or deformity. The



**Figure 6:** Clinical results: (a) clinical appearance after 3 months of follow-up (b) without any sign of recurrence

presence of a tooth root with periapical lesion inside the dysplastic bone could have influenced the lesion behavior and be related to the expansion clinically observed. On the other hand, tooth extractions are the main cause of osteomyelitis in CODs,<sup>[9]</sup> while this type of event is not related to cases of FD.

These facts make the lesion in this report an unusual occurrence. An alternative would be to consider it as a condition of FD that had not remodeled after the conclusion of the individual's skeletal growth, in contrast to the information given by the patient. The asymptomatic characteristic of these cases often makes the history reported by patients inaccurate. We also had no access to more accurate information about the patient's dental and medical history, although the patient informed no relevant up-to-date medical history. Moreover, we noted early loss of several teeth and the presence of root remains related to the lesion in the contralateral hemi-maxilla.

Another aspect that always raises discussion in relation to FOLs in the jaws concerns to the viability of dental implant placement in these areas. For our patient, the treatment strategy did not include dental implants, but only a conventional removable prosthesis after final surgical reshaping of the maxillary alveolar ridge.

While the present report is certainly not unique, it may serve as a point of discussion and re-evaluation of the current classification criteria of FOLs. Considering the characteristics presented in this particular case, we could suggest another entity that could be named benign cemento-osseous lesion with adult onset.

FOLs continue to be a group of diseases that deserve a larger amount of research in order to clarify their etiopathogenesis and efficient therapeutic protocols, especially concerning the variants of CODs and FD. Some occurrences, such as that in the present case, do not fit properly into the current WHO classification, thus generating some doubts concerning the correct management of such patients.

## CONCLUSION

The differential diagnosis of FOLs has become fundamental for the purpose of accurately establishing and planning the treatment protocol, mainly because of their clinical, radiographic and histopathological behavior. Conservative surgical management and enhanced clinical and radiographic follow-up of cases will improve the patient's functional and esthetic conditions and may enable future rehabilitation of the respective areas involved.

## Ethical approval

This report was exempt from requiring ethical approval because of the use of medical data records of the patient, without exposure of her identity or photographs that showed her facial identification or characteristics. We are including the Portuguese version of a document entitled "Patient Release Form" that consists of detailed explanations given to the patient, about further publication and about the journal. This form was signed by the patient, and represents her agreement with its content.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

## Acknowledgment

This work was supported by the Department of Oral Diagnosis, School of Dentistry, Sao Paulo, Brazil, mainly by the Department of Stomatology, including the technical team who participated in the work at the outpatient clinic. We would like to thank Ivan Solani Martins, Giovanna

Dantas Aguiar and Margery Jacoba Galbraith for their contributions to the process of writing and revising this manuscript.

## Financial support and sponsorship

This work was supported by the National Council for Scientific and Technological Development of Brazil (870091/2001-8).

## Conflicts of interest

There are no conflicts of interest.

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