An Unusual Case of Symptomatic Periapical Cemento-Osseous Dysplasia: Case Report

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Abstract

Periapical cemento-osseous dysplasia is a benign fibro-osseous lesion, relatively common and usually asymptomatic with typical clinical, radiographic, incidence and frequency features. Intra-osseous changes are usually observed in the periapical region in the anterior mandibular teeth in black female patients in their fourth decade of life. The purpose of this article is to report the unusual case of a black female patient who presented periapical cemento-osseous dysplasia with painful symptomatology in the anterior mandibular teeth. Endodontic treatment of the involved teeth was performed, and without remission of the painful symptomatology even after 1 year, excision of the intra-osseous lesions was necessary. Ten years after the surgical procedure, the patient is under follow-up with no signs of recurrence of pain and the lesions.

Keywords: Periapical Cemento-Osseous Dysplasia; Orofacial Pain; Endodontic Treatment; Oral Surgery

Introduction

Periapical cemento-osseous dysplasia is a relatively common oral pathology, currently classified as a benign fibro-osseous lesion, in which there is production of mineralized tissue (cementum and bone), located in the periapical regions of the affected teeth [1-6].

Generally the lesions are multiple, and may also be solitary, with a higher incidence in the black female affecting mainly the fourth decade of life. Multilocular symmetrical lesions located in the posterior part of the jaws, affecting or not the four quadrants of the oral cavity may also occur, thus determining a clinical variation called florid cemento-osseous dysplasia [1-5,7].

Typically, periapical cemento-osseous dysplasia is a radiographic finding and is asymptomatic, although the literature presents rare cases of painful symptoms associated with the lesion. Radiographically this lesion can be classified into 3 distinct stages of evolution. It begins with an osteolytic stage, where destruction of the medullary bone occurs, presenting a radiolucent image without the presence of a radiopaque halo. In the intermediate or mixed stage, also called cementoblastic, the radiographic features are composed of radiopaque and radiolucent areas. In the maturation stage, the lesion is radiopaque and bounded by a radiolucent halo [1-8].

Since it is a benign lesion, usually asymptomatic and self-limiting, clinical and radiographic follow-up is generally recommended.

However, when painful symptoms are present, other treatments may become necessary.

**Purpose of the Study**

The purpose of this article is to report the case of a patient who presented periapical cemento-osseous dysplasia in the anterior mandibular teeth whose painful symptomatology remained even after endodontic treatment of the teeth involved, being necessary the tissue excision of the lesions.

**Case Report**

African-descendent female patient, 42 years old, attended the private clinic, complaining of localized pain in the mental region.

Clinically, the teeth were healthy, with absence of restorations, signs of trauma, with normal staining and gingiva in good condition. The mucosa was normal (Figure 1). Pulp vitality tests were positive in the four lower incisors.

Panoramic radiograph showed radiopaque images surrounded by a radiolucent halo located in the periapical region of the mandibular incisors, suggesting the clinical hypothesis of Periapical Cemento-Osseous Dysplasia (Figure 2).

Regarding systemic conditions, surgery and hormone therapy for uterine myoma were related.

**Figure 1**: Initial clinical aspect: healthy mandibular incisors and normal mucosa.

**Figure 2**: Initial radiographic aspect: radiopaque images surrounded by a radiolucent halo located in the periapical region of the mandibular incisors, suggesting the clinical hypothesis of periapical cemento-osseous dysplasia.

Endodontic treatments were performed on the four mandibular incisors (Figure 3), with persistence of the painful symptomatology.

**Figure 3**: Endodontic treatments performed.

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After 12 months of endodontic treatment without remission of the painful symptomatology, it was recommended that the lesions be excised. After clarifying all information to the patient, written consent was obtained for the procedure.

A bilateral mental block was performed, followed by an incision in the mucogingival junction and a full thickness flap (mucoperiosteal), thus exposing the bone tissue (Figure 4). Symphyseal osteotomy was performed until exposure of the lesions, which had firm insertion and were well adhered (Figure 5). After removing the lesions, the procedure was complemented by apicectomy of the four mandibular incisors. The region was sutured (Figure 6). The patient was given analgesic, anti-inflammatory and antibiotic drugs.

The removed lesions (Figure 7) were fixed in 10% formalin and sent to the Laboratory of Surgical Pathology of the School of Dentistry of the University of São Paulo for anatomicopathological examination. Microscopically, the fragment revealed a mineralized tissue with irregular and well-defined margins similar to cementum. Basophilic lines of apposition and reversion composed the structure of this material, as well as areas of dense connective tissue consisting of spindle-shaped cells with basophilic nuclei and cytoplasm without precise boundaries, scattered randomly throughout the specimen. Blood vessels and hemorrhagic areas completed the histological image. The final diagnosis was periapical cemento-osseous dysplasia.
After 7 days, the remaining sutures were removed (Figure 8 and 9). The patient was evaluated weekly until 30 days, in which satisfactory mucogingival repair was observed. During this visit, a follow-up periapical radiograph was taken (Figure 10).

The patient was followed up radiographically at 3 months (Figure 11) and 12 months (Figure 12). After 12 months, satisfactory bone repair and cessation of painful symptoms were observed.

The patient has been followed for 10 years with no signs of recurrence of the lesion and pain (Figure 13).
Discussion

Initially classified as a lesion of odontogenic origin, in 1971 by the WHO, Periapical Cemento-Osseous Dysplasia was included in one of the 4 groups of cementomas and later, in 1992, it was re-classified as a benign fibro-osseous lesion. Periapical Cemento-Osseous Dysplasia are probably the most common included in this group followed by cementoma, fibrous periapical dysplasia, cementifying fibroma, cementoblastoma, fibrocerementoma, periapical osteofibrosis, local osteofibroma, and others [2,4,5].

There is a wide variety of confusion pertinent to the term periapical cemento-osseous dysplasia, as already cited: cementoma; cementoblastoma; fibrous periapical dysplasia; fibrocerementoma; cementifying fibroma; periapical osteofibrosis; focal/florid or focal/florid cemento-osseous dysplasia; local or peripical osteofibroma; ossifying fibroma; periapical fibroosteoma; fibroostecementoma; sclerosing cementoma; cemento-ossifying fibroma; giant cementoma [4,5,9-11].

Periapical cemento-osseous dysplasia is characterized by three stages of evolution. The first stage, also called osteolytic phase, occurs when the bone tissue of the periapical region is replaced by fibrous tissue, characterized radiographically by a radiolucent lesion, usually delimited. The second, intermediate phase, called cementoblastic, the deposition of mineralized tissue (cementum) within this tissue mass is observed, characterized radiographically by a radiolucent lesion with a slightly radiopaque mass (at the beginning of mineralization). The third stage, of maturation, occurs the increment of mineralization, whose radiographic characteristic is a radiopaque lesion surrounded by a radiolucent halo, usually well delimited. This last stage of maturation is considered pathognomonic of periapical cemento-osseous dysplasia. These stages are also pertinent to the clinical variation florid cemento-osseous dysplasia, a condition that presents as multilocular mineralized tissue masses, usually symmetrical, and may also affect the four quadrants [1-8].

Diagnosis occurs, most of the time, incidentally, by means of routine radiographic examinations, usually by periapical and panoramic radiographs [1-9,11]. However, panoramic radiographs should not be used as a means to diagnose periapical cemento-osseous dysplasia, particularly in its early stage when the lesion is ra-
diolucent. Moreover, panoramic radiography presents distortions and overlaps, underestimating the diagnosis of periapical cemen- 
to-osseous dysplasia and other oral pathologies [6]. Contradicto-
rily, in the present report, the characteristics of lesions in matu-
ration stage favored visualization and diagnosis. Additionally, the 
use of CT scanning in the diagnosis of periapical cemento-osseous 
dysplasia also improves not only the diagnosis but also the locali-
case using laser Doppler blood flowmetry as a more accurate diag-
nostic method than other tests or when pulp vitality tests were not 
elucidative. The exam was based on the evaluation of blood flow, 
determining pulp vitality by the movement of erythrocytes in the 
periapical circulatory system.

The histopathological pattern follows the different stages of 
periapical cemento-osseous dysplasia development. In the os-
teolytic stage, there is an intense fibroblastic proliferation where 
small areas of osteoid formation can be found intermingled with 
this stroma, with no signs of inflammation. In the next stage (ce-
mentoblastic), there is a progressive deposition of bone tissue and 
cementiform material among the fibroblastic proliferation. The 
maturation phase presents dense mineralized areas, almost com-
pletely taking over the histological field [1-7].

The differential diagnosis of periapical cemento-osseous dys-
plasia includes several lesions such as ossifying fibroma, Paget’s 
disease, odontogenic keratocyst, ameloblastoma and central giant 
cell granuloma [4,7]. Radiographically and depending on the stage 
of the lesion, similarity or mimicking with periapical lesions of 
endodontic origin such as periapical abscess, granuloma and cyst 
is observed [3,5,7,8]. In the present case, the lesions showed the 
characteristics inherent to the maturation stage.

The etiopathogenesis of periapical cemento-osseous dysplasia 
is still unknown, and there are, however, some questionable theo-
ries. Usually, some authors have cited odontogenic origin or reac-
tive process of periapical tissue [1,2,4,5,7,8]. Mild chronic trauma, 
caries, periodontal diseases, systemic diseases, infections, hormo-
nal influences (which alter bone remodeling), genetic factors, cou-
l'd also be associated with periapical cemento-osseous dysplasia. 
History of uterine polyps under hormone therapy was still repor-
ted as an etiopathogenic factor [2,5], as well as found in the present 
is no inflammation or pulp necrosis.

Female gender, black race, median age between the 3rd and 6th 
decades of life, usually affecting the 4th decade, and involvement 
of the mandibular incisors were unanimous referring to incidence 
[1,2,4-7,11]. These characteristics were markedly observed in 
the present case. However, it should be noted that the region of 
involve ment may vary according to ethnic group [1,2,6,7]. There 
are reports of a higher incidence in the premolar and molar region 
(multilocular) in Asians and Caucasians when compared to other 
ethnic groups, although the frequency regarding age and gender 
were compatible [4,5,7].

Since this is a generally asymptomatic and self-limiting lesion, 
simple preservation is indicated, therefore not requiring any type 
of intervention. There is a need for evaluation of cortical integrity, 
existence or not of bone resorption or rare tooth movement [1- 
5,8]. In the present report, we suppose that the symptomatology 
occurred due to the possible compression of nerves (incisors) of 
the region.

Due to failures in diagnosis, particularly related to radiographic 
interpretation, unemployment of pulp vitality tests, or even the du-
bious results when employed, endodontic treatment was instituted 
in some cases [2-7]. However, knowing the nature of the lesion, 
it is essential to avoid endodontic treatment of the teeth involved 
to the present one, with symptomatology located in the anterior 
mandible, the maintenance of the pulp vitality of two of the four 
affected incisors, after the procedure of surgical excision and api-
cectomy, thus opposing our conduct (endodontic treatment prior 
to surgical excision associated with apicectomy).

Surgical removal is not necessary in cases of asymptomatic 
Periapical Cemento-Osseous Dysplasia. However, some authors 
have submitted their patients to the procedure and subsequent 
pathological examination [1-5,7,11]. Surgical excision is indicated 
in symptomatic cases including pain, paresthesia, and infection; 
with expansive growth of the lesion (although it is self-limiting); 
or in candidate areas for dental implant installation [5]. Exeresis 
is easily accomplished by curettage and removal of the fragment 
of periapical cemental dysplasia, presenting an appearance of a 
“sandy” lesion [7].

**Conclusion**

Periapical cemento-osseous dysplasia is a benign and self-limi-
ting lesion, usually asymptomatic. However, in some cases it can
be symptomatic and require endodontic or surgical treatment. Most of the time, in asymptomatic cases, clinical and radiographic follow-up is sufficient, as well as patient orientation to avoid diagnostic confusion.

**Bibliography**


