



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.



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

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.

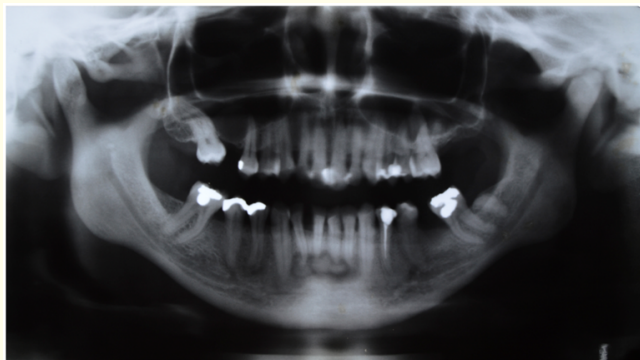


Figure 2: Initial radiographic aspect: radiopaque images surrounded by a radiolucent halo located in the periapical region of the mandibular incisors, suggesting the clinical hypothesis of periapical cemento-osseous dysplasia.

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



Figure 3: Endodontic treatments performed.

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.

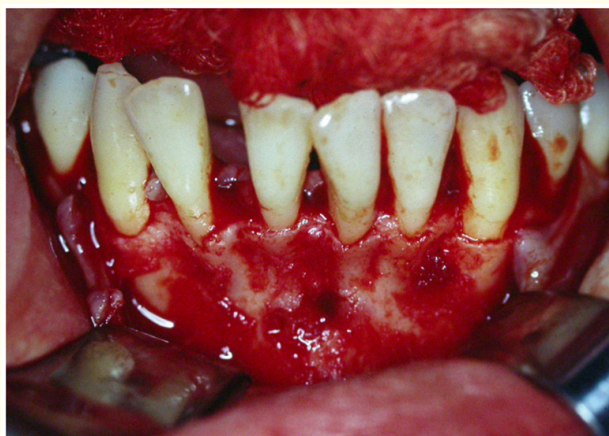


Figure 4: Mucoperiosteal flap and exposure of bone tissue.

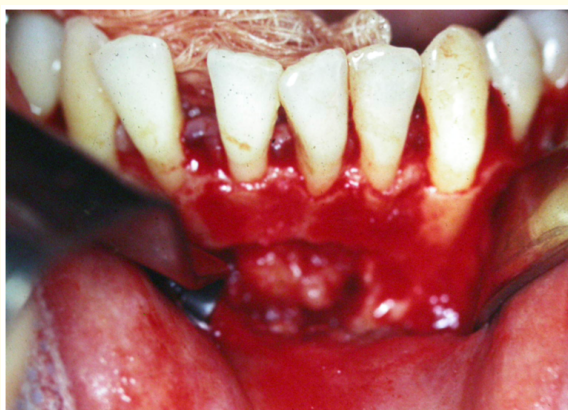


Figure 5: Exposure of lesions after symphyseal osteotomy.



Figure 6: Sutured region.

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 anatomopathological 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.



Figure 7: Removed fragments.

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).



Figure 8: Post-surgery (7 days): remaining sutures.



Figure 9: Post-surgery (7 days): removal of sutures.

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).



Figure 10: Radiographic evaluation after 30 days.



Figure 11: Radiographic evaluation after 3 months.

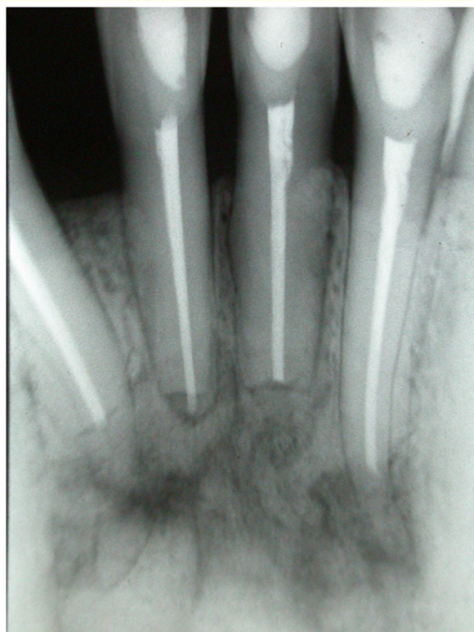


Figure 12: Radiographic evaluation after 12 months.

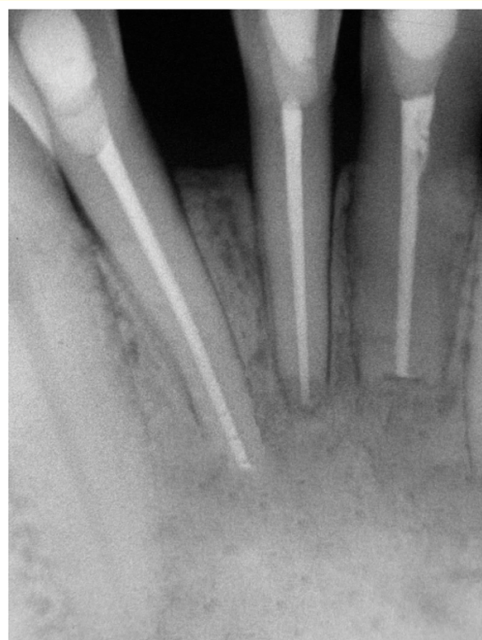


Figure 13: Radiographic evaluation after 10 years.

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, fibrocementoma, 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; fibrocementoma; cementifying fibroma; periapical osteofibrosis; focal/florid or focal/florid cemento-osseous dysplasia; local or peripical osteofibroma; ossifying fibroma; periapical fibroosteoma; fibroosteocementoma; 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 cemento-osseous dysplasia and other oral pathologies [6]. Contradictorily, in the present report, the characteristics of lesions in maturation 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 localization of the lesions [4-5,8]. Chandler, *et al.* (1999) [9] evaluated a case using laser Doppler blood flowmetry as a more accurate diagnostic 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 osteolytic 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 (cementoblastic), there is a progressive deposition of bone tissue and cementiform material among the fibroblastic proliferation. The maturation phase presents dense mineralized areas, almost completely taking over the histological field [1-7].

The differential diagnosis of periapical cemento-osseous dysplasia 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 theories. Usually, some authors have cited odontogenic origin or reactive process of periapical tissue [1,2,4,5,7,8]. Mild chronic trauma, caries, periodontal diseases, systemic diseases, infections, hormonal influences (which alter bone remodeling), genetic factors, could also be associated with periapical cemento-osseous dysplasia. History of uterine polyps under hormone therapy was still reported as an etiopathogenic factor [2,5], as well as found in the present report. In contrast, Consolaro, *et al.* (2020) [6] postulate that there is no inflammation or pulpal 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 involvement 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 dubious 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 [3,8,11]. Stabholz, *et al.* (1988) [10] reported, in a case similar 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 apicectomy, 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 cementary dysplasia, presenting an appearance of a "sandy" lesion [7].

Conclusion

Periapical cemento-osseous dysplasia is a benign and self-limiting 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.

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