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CASE REPORT

Diagnostic Procedures and Prosthetic Management of Cemento-Osseous Dysplasia a Case Report

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ABSTRACT

Cement-osseous dysplasia is an uncommon benign fibro-osseous lesion of the maxillae in which cementum tissue replaces the normal architecture of the bone. It is usually asymptomatic, and discovery is incidental on routine radiological examination. Treatment is necessary if there is an infection due to exposure to the lesion in the oral cavity. In these severe cases, pain, cortical blowing, and facial deformity may be present. Thus, patients with cemento-osseous dysplasia have prosthetic bearing surfaces unfavorable for prosthetic rehabilitation.

The management of cemento-osseous dysplasia requires a careful clinical examination to make a correct diagnosis and develop the appropriate treatment plan. The practitioner must inform the patient of the limitations of prosthetic rehabilitation.

The objective of this article is to review, through case report, the clinical, histological, and radiological characteristics and possible complications of this lesion and to highlight its impact on the prosthetic bearing surfaces as well as the problems inherent in the prosthetic rehabilitation.

KEYWORDS: Cemento-osseous dysplasia; Prosthetic bearing surfaces; Prosthetic rehabilitation.

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INTRODUCTION

Cement- osseous dysplasia COD is a rare, benign, osteo-fibrous lesion of the maxilla. It is a non-neoplastic condition in which normal bone is replaced by a fibrous connective tissue matrix containing abnormal bone or cementum.

Histologically, this entity is composed of spindle-shaped fibroblast-like cells and collagen, with only a few trabeculae interspersed with bone and calcified cement-like material **[1]**.

In its 2017 publication, the World Health Organization (WHO) described three clinical presentations of COD: periapical, focal, and florid **[1, 2]**.

- Periapical COD: dysplastic lesion observed in the mandibular canine-incisor area and encompassed only a few adjacent teeth.
- Focal COD: homologous to periapical COD, with fewer lesions involving a posterior quadrant of the maxilla (rather than the anterior mandible).
- Familial florid COD: more extensive forms, occurring bilaterally on the mandible or in all quadrants of the maxilla **[1]**.

This lesion is most commonly found in middle-aged, dark-skinned women, although it can also occur in white-skinned individuals and Asians. The precise etiology of COD remains unknown, and the precipitating factors are not identified, although it appears to represent a form of reactive or dysplastic process.

Since COD lesions are close to the periodontal ligament and have similar histopathology, it has been suggested that their origin and pathogenesis originate from the periodontal ligament. Therefore, few authors have reported that cementum remnants in the bone after extraction could be a cause of COD **[2, 3, 4].**

COD occurs only in the maxilla. It is not associated with any other skeletal involvement or blood count abnormalities.

Clinically, COD may be asymptomatic and detected incidentally during a routine radiographic examination. Usually, the teeth associated with the lesions are alive.
In case of infection, the lesion goes into the symptomatic phase and sometimes manifests as pain, purulent exudate, mucosal fistula, focal expansion, and facial deformities. This infection is due to intraoral exposure of cementum-osseous masses **[4, 5, 6, 7].**
Radiologically, COD is usually limited to the maxillary dental areas or edentulous alveolar processes, especially in the mandibular premolar region, symmetrically and bilaterally; posterior maxillary involvement is less common. Immature lesions are characterized by a paucity of calcified material, whereas more mature lesions are characterized by the development and coalescence of dense, globular masses of calcified material **[1, 3, 4, 6].**
Histological examination shows highly calcified masses resembling secondary cementum, sometimes very basophilic, irregularly shaped, dense, and cell-free, as well as non-lamellar bone masses in the fibroblastic connective tissue. In the case of sequestration of the lesions after an infectious process, necrotic tissue is present **[3, 4, 6].**
This report discusses, through a clinical case, the main challenges related to removable prosthetic rehabilitation in the case of cemento-osseous dysplasia presents the keys to a diagnostic approach splitting the difficulties and evokes the various possible therapeutic solutions.

MATERIALS AND METHODS

Clinical examination

Patient B.J., 50 years old and in good general health, was referred by the surgical pathology department for partial removable prosthetic rehabilitation. His reason for consultation was the extraction of a residual root.
Extra-oral examination revealed an oval, symmetrical face, equality of the facial stages, a slightly convex profile, and a well-supported upper lip, the smile is tooth-dominant (Fig n°1), the volume and tone of the masticatory muscles are normal, the examination of the joints did not show signs of temporomandibular dysfunction.
No abnormality related to cemento-osseous dysplasia was reported on extra-oral examination.

Fig n°1: extra oral view and smile without any impact of the CDO

Intraoral examination showed the presence of a partial Kennedy Applegate Class 1 maxillary edentulous, caries, and amalgam fillings. All teeth near the dysplastic lesions responded positively to vitality tests. The periodontium is thick, and the attached gingiva is sufficient. The fibro mucosa is normal in appearance, adherent, and firm. The maxillary ridges are voluminous, with swelling of the left maxillary ridge. (Fig n°2.3.4)

Fig n°2: intraoral view of the maxilla revealing voluminous ridges

Fig n°3: intraoral view of the arches in occlusion

Fig n°4: intraoral view of the right side (extrusion 34/38)

Occlusal examination indicates an insufficient prosthetic space on the left posterior side (Fig n°5.6), the occlusal plane is disturbed (extrusion: 38.34.44.and 48).

Fig n°5: intraoral view of the left side in occlusion showing the insufficiency of the prosthetic space

Fig n°6: insufficient prosthetic space on articulator-mounted models

Anatomopathological examination of the surgical excision specimen from the biopsy performed in the surgical pathology department confirmed the radiological diagnosis, favoring florid cemento-osseous dysplasia.

The orthopantomogram showed several radioopaque images apically to the mandibular teeth. The left maxillary lesion area showed a larger cementoblasts image. The diagnosis was in favor of florid cemento-osseous dysplasia. (Fig n°7)

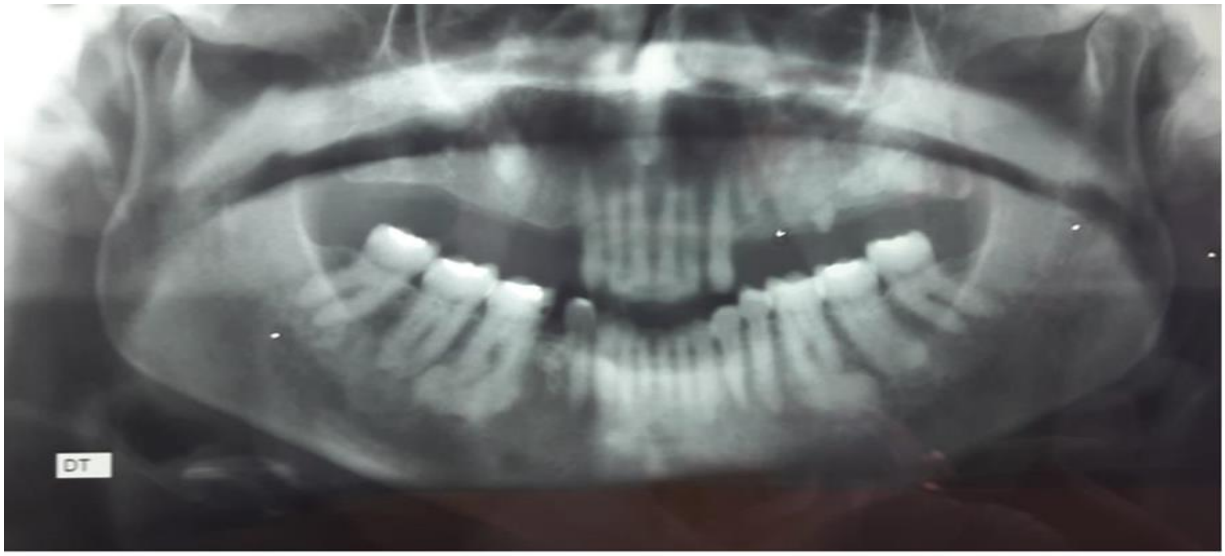

Fig n°7: Orthopantomography shows radiopaque images in the left maxillary region and opposite the mandibular teeth.

Treatment Decision

The maxillary arch has a large distal partial edentulism with hypertrophy of the ridges due to the cemento-osseous lesion. This hypertrophy can be detrimental to prosthetic rehabilitation in bearing surfaces, occlusion, and esthetics, requiring surgical management of the prosthetic space to optimize prosthetic stability.

The treatment plan consists of maintaining good oral hygiene, periodontal and dental sanitation, and periodic monitoring of the cemento-osseous lesion to avoid superinfection and transition to the symptomatic form.

For the prosthetic rehabilitation of partial edentulism, we have chosen to perform a removable partial denture with a metal framework, given the contraindication of implants in a dysplastic focus.

Therapeutic Approach

Pre-prosthetic stage

Before starting the prosthetic rehabilitation, the maxillary casts were transferred to the articulator. The analysis of the models on the articulator allowed us to determine the occlusal plane, appreciate the available prosthetic space, and evaluate the surgical adjustments to be performed. (Fig n° 8;9)

Adjusting the occlusal plane required grinding of tooth 34, coroplast of tooth 44, and extraction of the wisdom teeth that touched the maxillary ridge.

For a good balance of the metallic partial prosthesis, we decided to remodel the ridge by subtractive corrective surgery after realizing a surgical guide. (Fig n°10)

A previously made transitional prosthesis was placed to guide the healing process. (Fig n°11)

Fig n °8: simulation of surgical remodeling on articulator

Fig n °9: simulation of the teeth arrangement on the articulator and preparation of the provisional prosthesis

Fig n °10: preparation of the surgical guide before the subtractive surgery

Prosthetic Stage

After healing, the prosthesis's fabrication (Fig n°12) was started by preparing the abutment teeth according to the chosen design, recording the peripheral joint with Kerr paste. The anatomical-functional impression was made under digital pressure, using a polyether given the quality of the fibro mucosa.

The occlusal models were made on the secondary model to facilitate recording the intermaxillary relationships in maximum intercuspation and at the right vertical dimension and the transfer of the models to the articulator. We could thus visualize the gain in prosthetic space that the surgical remodeling offered us.

The choice and the arranging of prosthetic teeth respect the aesthetic and biomechanical requirements for successfully integrating the partial metal denture. The arranging is tried in the mouth and validated before polymerizing and placing. (Fig n°13; 14)

The patient is seen regularly for follow-up sessions.

Fig n°11: view of the maxillary arch (the surgery day)

Fig n°12: left intraoral view after healing

Fig n°13: view of the maxillary arch (on the day the prosthesis was inserted)

Fig n° 14: extraoral view after placement of the prosthesis

DISCUSSION

COD is an uncommon benign fibro-osseous lesion. It was first reported by Bhaskar and Cutright in 1968, then by Melrose in 1975. These authors were the first to describe the lesion's clinical, pathological, and radiological criteria and to use the term cemento-osseous dysplasia **[5].**

It has a clear predominance in middle-aged women. Toledano & al. Report that about 90% of cases are diagnosed between the third and fourth decade of life. African women seem to be the most affected group, which coincides with our cases, followed by Asian women and Caucasians in a lesser proportion **[8]**.

COD is a clinically asymptomatic lesion most of the time, but some patients may present more or less severe symptoms.

In this regard, a systematic review of 158 cases of COD revealed 17 cases of superinfection with purulent discharge, and 63% of cases in a series of 54 patients were asymptomatic **[9,10]**.

Our patient presents as the only symptom the hypertrophy of the ridge. The lesion was discovered during a routine radiological examination. This hypertrophy can be detrimental to prosthetic rehabilitation.

The expansion caused by the lesion has long been a debatable topic for pathologists. Some published pathology reviews have described the lesions as non-expandable; however, in other studies, a slight expansion of at least one of the cortical plates has been reported with intact but thinned cortices **[1]**. Using occlusal radiographs, Kawai & al. 11 noted mild mandibular expansion in 3 of 54 cases of COD (periapical and florid) **[11]**.

The asymptomatic forms of COD don't require treatment. They are monitored clinically and radiologically with attention to good oral hygiene to detect a possible change in the behavior of the lesions. According to Ducommun & al, the "wait and see" concept is usually sufficient **[5, 12]**.

In symptomatic forms, two therapeutic attitudes can be envisaged. For some, antibiotic therapy would be sufficient to ensure the sclerotic, avascular, cementum-like lesions sequestration before their surgical removal.

For others, immediate surgical intervention is recommended because partial resection does not control the infection and may promote its spread to other areas **[3, 4, 7]**.

In all cases, once the diagnosis is made, patients should be monitored regularly to prevent infection from setting in. Thus, early management of cavities, fissures, and periodontal pockets can prevent infection and avoid complications.

Moussaoui & al. Reported that mucosal ulceration, alveolar bone atrophy, extraction, pulp necrosis, endodontic treatment, periodontal pocketing, or biopsy of the lesion might cause the onset of symptomatology following exposure of the lesions to the oral environment **[3]**. The indication for endodontic treatment must be discussed according to the benefit/risk ratio of the treatment. Biopsy and dental extractions should be avoided owing to the reduced power of mucosal healing **[7]**. The consequences of orthodontic movement in the context of COD are poorly documented. In their case report, Kato & al. suggest that the presence of cemento-osseous dysplasia does not prevent successful orthodontic treatment **[13]**.

Fixed prosthetic rehabilitation is recommended whenever possible, but implant-supported prosthetic rehabilitation seems difficult, and pre-implant surgery is because of abnormal bone with poor vascularity in the cementum lesions. However, an implant success was described by Bencharit and al who reported the placement of five implants in the anterior region in a patient with COD with a 3-year follow-up **[7, 14]**.

A similar experience has been reported by Chadid & al. In a recent publication. They describe the successful osseointegration of dental implants in the areas of florid cemento-osseous dysplasia after eight years of follow-up **[15]**.

However, in a report published in 2014, Oliveira & al. Present the case of a patient with COD in whom a dental implant was placed but lost six months after surgery and suggest that COD should be considered a limiting factor in the indication of dental implants **[16]**.

Meanwhile, Shin & al. Reported a case of chronic osteomyelitis induced by dental implant placement on cemento-osseous dysplasia **[17]**.

Gerlach & al. Presented a unique case of florid cemento-osseous dysplasia with concomitant cemento-ossifying fibroma discovered during explantation of an implant. These two diseases could appear successively, coexist in the same patient, and complicate the long-term success of an implant-supported prosthesis **[18]**.

According to MacDonald-Jankowski, we currently have very little information about the post-surgical healing of COD sites. We do not know if they recur, and even if they do, we do not know if the healed bone will be of adequate quality for implants **[10]**.

Therefore, we have chosen to rehabilitate aesthetically and functionally the clinical case presented in this article with a removable partial denture, allowing us to stabilize and monitor the lesion and avoid any ulceration or trauma before and throughout the prosthetic post-prosthetic stages.

The patient had an asymptomatic form of COD with a swelling of the left maxillary ridge inauspicious to the prosthetic balance and reducing the prosthetic space. The rehabilitation by partial removable prosthesis was carried out by remodeling the left maxillary ridge **[19]**.

The quality of the impressions and the choice of material play a primordial role in the tissue and prosthetic stability. The fragile fibromucosa implies a careful choice of impression material and its viscosity. Thus, in the face of a firm and adherent fibromucosa, the use of Kerr's paste, because of its compressive effect, allowed to capture functional movements of the intra-oral musculature. The presence of cortical bone swelling could be addressed by using polyethers and their thixotropic properties (elastic seal) [20].

Compliance with occlusal requirements ensures the prevention and limitation of mechanical stresses on the fibromucosa and underlying bone. The choice and mounting of the prosthetic teeth are of paramount importance in maintaining stable occlusal relationships over time and thus prosthetic and tissue balance.

Strict occlusal balancing is recommended on the day of placement of the removable prosthesis. Otherwise, an ill-fitting prosthesis could lead to alveolar bone resorption and contribute to the exposure of sclerotic masses to the oral environment **[21]**.

During the post-prosthetic phase, prevention of superinfection requires prosthetic maintenance and tissue hygiene. Regular brushing of the teeth and fibromucosa and weekly cleaning of the prostheses should be prescribed by the practitioner. Any inflammation or traumatic lesion of the fibromucosa must be treated immediately. Otherwise, the lesions will be exposed to the oral environment.

CONCLUSION

Since there is insufficient evidence on the long-term success of rehabilitation with removable prostheses in cemento-osseous dysplasia, we recommend regular clinical and radiographic follow-up with the patient maintaining good oral hygiene and regularly attending for follow-up and prophylaxis.

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AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors. Indeed, all the authors have actively participated in the redaction, the revision of the manuscript, and provided approval for this final revised version.

COMPETING INTERESTS

The authors declare no competing interests with this case.

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PATIENT'S CONSENT

Written informed consent was obtained from the patient for the publication of this case report.

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