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Exploring a Case of Cemento-Osseous Dysplasia: Clinical Insights and Treatments Outcomes

Omar Walha[1*], Maroua Garma[1], Saida Godhbani[1], Dhekra Jlassi[1], Sameh Sioud[1], Habib Hamdi[1]

[1]Department of Oral Medicine and Surgery, Faculty of Dental Medicine of Monastir, Tunisia

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***Corresponding author:** Omar Walha
Department of Oral Medicine and Surgery Departments, Faculty of Dental Medicine of Monastir, Tunisia

Abstract **Case Report**

Cemento-osseous dysplasia is a benign disorder of the maxilla in which cemento-osseous tissue replaces the normal architecture of the bone. It is a benign fibro-osseous lesion of the jaws associated to root apexes and containing amorphous calcifications which would correspond to cementum. It may present in one of three forms: periapical, focal or florid. Usually asymptomatic, it is often fortuitously discovered during routine radiographic examination. However, an infection can occur and the condition progresses to a symptomatic phase which manifests by pain, purulent exudate, fistula formation and sequestration. Various radiological aspects may be observed, depending on the stage of development. In the absence of infectious complications, the therapeutic approach is abstention and monitoring. The aim of this work is to report a case of superinfected focal cemento-osseous dysplasia in the left mandibular bone in a 68-year-old Tunisian woman.

Keywords: Cemento-osseous dysplasia, Fibro-osseous lesion, Maxilla, Benign disorder, Dental radiography.

INTRODUCTION

Cemento-osseous dysplasia (COD) is a benign, non-neoplastic condition characterized by the abnormal development of bone and cementum-like tissue within the jawbone. This disorder, often discovered incidentally through routine dental radiographs, presents a range of clinical manifestations that can complicate diagnosis and management. The condition is classified into three main types: periapical COD (PCOD) which is limited to the apical region of a single tooth of the posterior jaw, focal COD (FCOD) is limited to the apical region of a few adjacent teeth, and florid COD (FLOOD) is more extensive, with multifocal and multi-quadrant involvement of the jaws, COD occurs more commonly in the mandible, with a predilection for middle-aged females of African descent [3].

While COD generally exhibits a benign nature and slow progression, its diagnosis can be challenging due to its overlapping features with other oral pathologies. Clinical and radiographic features may be sufficient to confirm the diagnosis of COD without pathologic examination [2, 10]. However, an early radiolucent FCOD/PCOD may be confused with a periapical granuloma/cyst. Distinguishing FCOD/PCOD from cemento-ossifying fibroma may be challenging.

Other radiographic differential diagnoses such as idiopathic osteosclerosis, condensing osteitis, exostoses, hypercementosis, cementoblastoma, and medication-related osteonecrosis of the jaw (MRONJ) are discussed. Understanding the etiology, clinical presentation, and appropriate management strategies for COD is crucial for dental professionals to ensure accurate diagnosis and effective treatment planning.

This article aims to report a case of COD in a female patient in order to provide a comprehensive overview of cemento-osseous dysplasia, including its classification, clinical features, diagnostic methods, and therapeutic approaches.

CLINICAL CASE

A 68-year-old female patient in good general health consulted for pulsatile pain with productive fistula in the fibromucosa of the left posterior mandibular region, the onset of symptoms having been observed 5 months previously.

The exobuccal examination showed no particular signs, and no swelling was detectable (Fig 1).

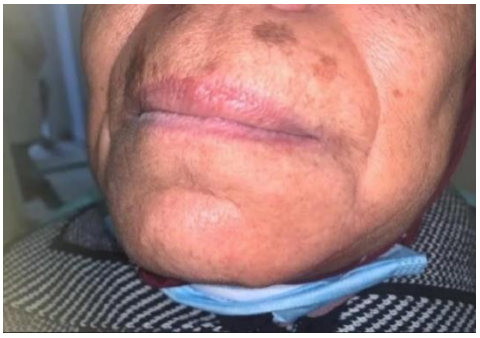

Fig 1: ExoOral examination

The endobuccal examination revealed deposits of plaque and calculus on the incisivo-canine block (Fig 2), on examination of the right-left mandibular molar region, and more specifically at the site of the extracted 36.

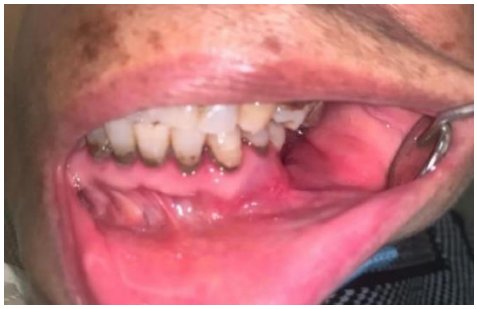

Fig 2: Intrabuccal examination

Ulceration of the buccal mucosa revealed a hard mass at the apex of the alveolar ridge (Fig 3). The surrounding mucosa was normal in appearance, but on palpation there was purulent serositis.

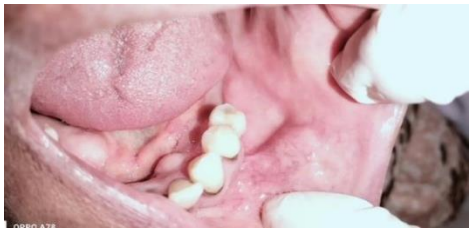

Fig 3: Ulceration of the buccal mucosa at the site of the extracted

There was also a fistula draining pus from the left mandibular crete. The lymph node examination was normal, and mucocutaneous sensitivity on the affected side was preserved. Panoramic radiograph was required and it showed, bone sequestration extended from the site of extraction of the 36 to the wisdom tooth, surrounded by a clear radiolucent halo (Fig 4). A computer tomography (CT) was ordered for further information; it revealed the lesions described above, with the presence of a fistula at the site of extraction of tooth 36 (Fig 5).

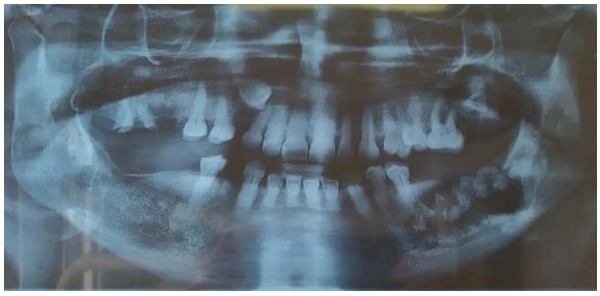

Fig 4: Panoramic radiographic showing bone sequestration extended from the site of extraction of the 36 to the wisdom tooth, surrounded by a clear radiolucent halo

Fig 5: Computer Tomography cross section showing bone sequestration surrounded by a clear radiolucent halo

Possible clinical diagnoses were:

- Osteitis,
- Focal cemento-osseous dysplasia,
- Surinfected focal cemento-osseous dysplasia
- Florid cemento-osseous dysplasia

The management was medico-surgical, involving a prescription of antibiotics for 7 days and local antiseptic treatment with chlorhexidine, followed by the removal of the sequestrum through piezosurgery and curettage of the site (Fig 6 & 7).

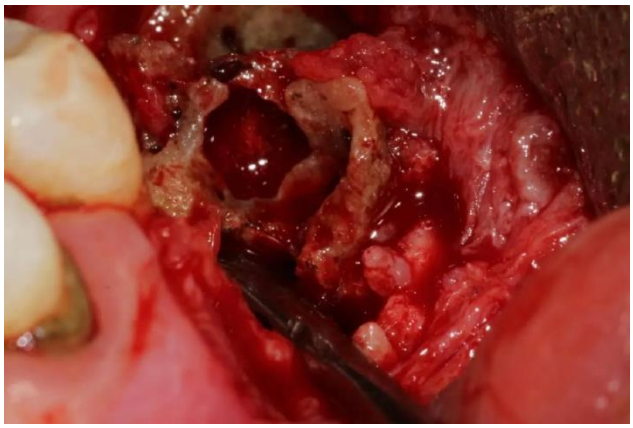

Fig 6: Removal of the sequestrum

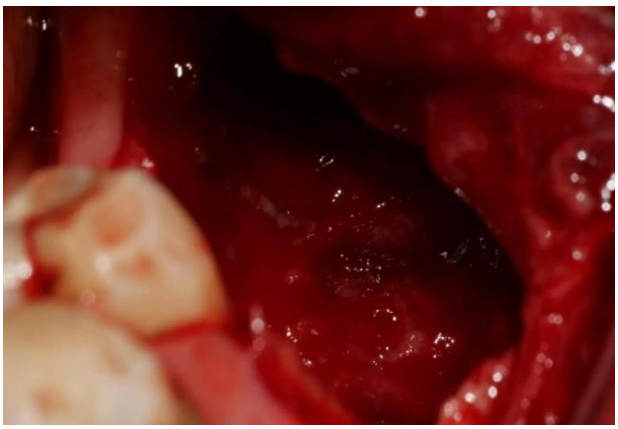

Fig 7: Curettage of the site

The biopsy specimen (Fig 8) was subjected to an anatomopathological examination which found an irregular mass of cemento-osseous tissue within a fibrous stroma.

The diagnosis of cemento-osseous dysplasia was then confirmed.

The patient was reviewed after 7 days for a follow-up and suture removal, it presented a good mucosal healing (Fig 9).

A follow-up radiograph was performed 06 months post-operatively, showing the beginning of site reossification (Fig 10).

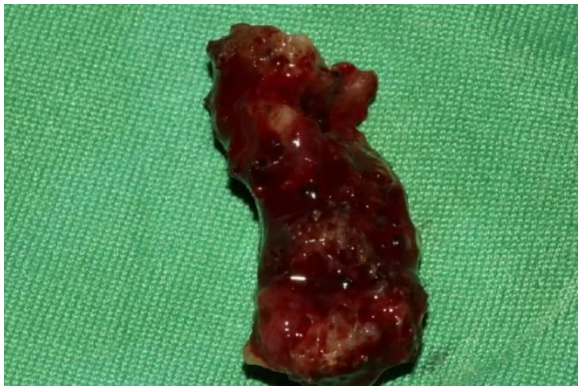

Fig 8: Biopsy specimen

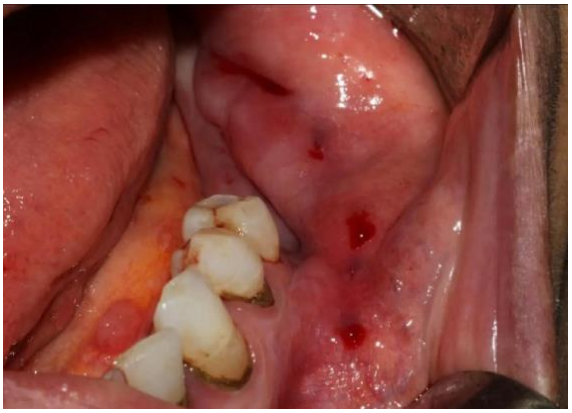

Fig 9: Post operative check up for suture removal showing a good healing of the mucosa[2]

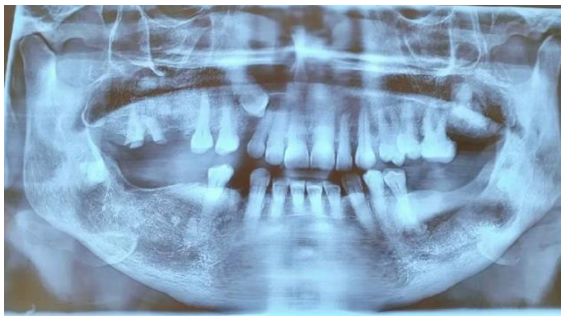

Fig 10: A panoramic radiographic post operative showing the beginning of site reossification

DISCUSSION

Cemento-osseous dysplasia or bone dysplasia belong to the group of benign non-neoplastic fibro-osseous lesions affecting the dentate areas of the jaws (reference article classification oms 2022) [10, 11].

When the lesion is confined to the anterior mandibular region, it is referred to as Periapical Bone Dysplasia.

Focal Bone Dysplasia refers to lesions limited to a posterior mandibular sector.

The term Florid Bone Dysplasia' is used if multiple sectors are affected. 'Gigantic Familial Cementoma' is also an extensive dysplasia that can involve multiple sectors, has a familial component, and an earlier age of onset compared to Focal Bone Dysplasia [1].

Focal Bone Dysplasia (FBD) was first described by Melrose *et al*., in 1976. It predominantly affects females and Africans in 59% of cases, with an average age of onset between 40 and 50 years A few rare cases have been reported in males [3, 7].

It is most often discovered incidentally during routine radiological examination. Sometimes, patients consult due to an infectious complication [10], as was the case for the reported patient. Bone dysplasia generally does not lead to complications: in a 1999 study, 63% of cases from a series of 54 patients were asymptomatic [6]. However, a systematic review of 158 cases of FBD revealed 17 cases of superinfection with purulent discharge [6].

Progression to a symptomatic phase occurs when inflammation superimposes on the pre-existing bone dysplasia. This is related to the exposure of sclerotic masses in the oral cavity due to dental extraction, trauma, alveolar surgery, or traumatic mucosal ulceration [8]. This phase can manifest as throbbing pain, swelling, fistulization with purulent exudate, or even sequestration [8]. Several of these symptoms were present in the reported case.

The etiology of these cemento-osseous dysplasia remains poorly defined to date. The hypothesis of a desmodontal origin seems to predominate, although genetic and environmental components are not excluded.

Most of these lesions are asymptomatic and adjacent to vital teeth.

Most of them are discovered by chance following a check-up radiographic examination combined with a clinical examination, these lesions can lead to a diagnosis.

The use of biopsy and histopathological analysis is possible but is rarely performed given the diagnostic arguments provided by clinical examination and imaging, as well as the risks of secondary infection that such an intervention might entail.

The lesion progresses through 3 stages which can be distinguished radiographically and depending on the patient's age [9]:

- Osteolytic stage: well-defined radiolucent lesion composed of well-vascularised fibrous tissue.
- Intermediate or cementoblastic stage: presence of radio-dense areas within a radiolucent lesion giving a mixed image representing the appearance of a cementoid deposit on the existing fibrous matrix.
- Mature, osteosclerotic and inactive stage: almost complete radiopaque lesion surrounded by a radiolucent halo.

The differential diagnosis of focal or periapical lesions includes endodontic lesions, cementoblastoma, and ossifying fibroma. For Focal Bone Dysplasia (FBD), the differential diagnosis mainly involves Diffuse Sclerosing Osteomyelitis (DSO) and Gardner's syndrome or SAPHO syndrome [4].

Schneider *et al*., emphasize that bone dysplasia should be considered when there is multilobular osteosclerosis affecting different quadrants of the mandible rather than a single segment.

Histologically, these lesions are defined as the replacement of healthy bone tissue by tissue rich in fibroblasts and collagen fibers.

A variable amount of mineralized tissue (bone-like and cement-like) is found within this fibrous stroma, evolving with the age of the lesion. Indeed, as they progress, the mineral component increases at the expense of other components and bone lacunae, thereby reducing vascular supply and, consequently, the healing and regenerative capacity of the bone.

Management of asymptomatic focal or florid cemento-osseous dysplasia is not necessary, and the patient should receive regular radiological follow-up to detect any potential changes in the behavior of the lesions, as well as maintain good oral hygiene. Biopsy is not indicated as it may trigger progression to the symptomatic phase. It is now accepted that no local treatment should be considered outside of infection phases.

However, they can become symptomatic also if their evolution is marked by secondary infectious complications, with osteomyelitis sometimes described as a symptomatic evolution of cement-osseous dysplasia. Since sclerotic bone is poorly vascularized, it has a low potential for responding to infections. The patient may then describe experience pain associated with swelling, fistulization with purulent exudate, or even the presence of a bone sequestrum.

Two therapeutic approaches can be considered: for some: antibiotic therapy alone may be sufficient to ensure the sequestration of sclerotic, avascular lesions resembling cement before their surgical removal. For others, Antibiotic therapy alone is not sufficient and extensive surgical intervention is recommended from the outset because partial resection does not control the infection and may promote its spread to other areas of Focal Bone Dysplasia (FBD)."

Piezosurgery can facilitate the surgical approach to these lesions; this technique was used for the first time by Vercelotti in 1988 for implant purposes, and has since been introduced in oral surgery as well as in other fields such as neurosurgery, ophthalmology, otorhinolaryngology, orthopaedics and traumatology [1].

This technique is based on the piezoelectric effect, which involves the deformation of certain crystals and ceramics when placed in an electric field, generating ultrasonic waves that are amplified and transferred to a vibrating tip with a cutting effect on mineralized tissues. The piezosurgery device used consists of a handpiece connected to the main unit and various types and shapes of inserts, each suited to a specific clinical situation [7]. In our case, we used the bone surgery kit with the BS6 insert, shaped like a curved scalpel, which helped us separate two surfaces: healthy bone and dysplastic bone. Piezosurgery offers precise cutting without damaging adjacent teeth and neighboring noble anatomical structures.

Additionally, thanks to the integrated irrigation system, it prevents bone overheating caused by conventional rotary instruments, which could lead to iatrogenic bone necrosis

Bencharit *et al*., [3] demonstrated the ineffectiveness of antibiotic therapy in the context of Diffuse Cemento-Osseous Dysplasia (DCO) in dealing with secondary infection. The vascular deficiency of the lesions makes it impossible for antibacterial molecules to diffuse into the affected area, necessitating the use of resective surgery in case of infection.

Osteointegration of implants is unlikely to be successful in these patients because the bone is abnormal and poorly vascularized.

Also; According to Park *et al*., [2], the risks of peri-implantitis and its consequences are increased. The formation of a bone sequestrum occurred following peri-implant infection, which may have led to infection of the dysplastic bone around the implant. Before this event, the implants remained osteointegrated in this pathological tissue.

In light of these findings, many authors, such as Shin *et al*., [5], contraindicate surgical management and thus implantation within a DCO context.

CONCLUSION

In conclusion, cemento-osseous dysplasia is a benign condition which, although it can sometimes present diagnostic challenges, is generally associated with a good prognosis when correctly identified and monitored. Close collaboration between clinicians, radiologists and pathologists is essential if it is to be fully understood and properly managed. Advances in imaging techniques and diagnostic approaches now make it possible to assess patients more accurately and monitor them more effectively. By adopting a multidisciplinary approach and keeping abreast of the latest research, healthcare professionals can offer optimal care and improve the quality of life of people affected by this condition. Ongoing vigilance and patient education remain essential to the successful management of cemento-osseous dysplasia.

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