

Exuberant hypercementosis mimicking cementoblastoma

Hipercementose exuberante simulando cementoblastoma

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ABSTRACT

Hypercementosis is a non-neoplastic proliferation involving excessive cementum deposition along the normal root cementum. Here, we report an unusual case of exuberant hypercementosis associated with chronic suppurative inflammation. A 77-year-old man sought dental care presenting a gingival suppurative lesion with several months of evolution. The radiographic examination showed a large radiopaque area involving the root of the tooth #46 surrounded entirely by a radiolucent area, suggesting cementoblastoma associated with infectious process. The histopathological examination of the involved tooth revealed an extensive hypercementosis associated with chronic suppurative inflammation and localized chronic osteomyelitis. This is an unusual presentation and due to the clinicopathological findings, the term “chronic hypercementitis” is proposed. Dentists, especially oral pathologists and endodontists, should be aware of this unusual presentation in order to establishing the correct diagnosis.

Key words: hypercementosis; osteomyelitis; oral pathology; mandible; differential diagnosis.

RESUMO

A hipercementose classifica-se como uma proliferação não neoplásica que envolve deposição excessiva de cimento ao longo do cimento radicular normal. Relatamos o caso de um paciente do sexo masculino, 77 anos de idade, que procurou atendimento odontológico apresentando lesão supurativa em gengiva, com vários meses de evolução. O exame radiográfico evidenciou uma grande área radiopaca envolvendo a raiz do dente 46, que estava circundada inteiramente por uma área radiolúcida, sugerindo cementoblastoma associado ao processo infeccioso. O exame histopatológico do dente envolvido revelou uma extensa hipercementose associada à inflamação supurativa crônica, além de osteomielite crônica localizada. Essa apresentação é incomum e, devido aos achados clinicopatológicos, é proposto o termo “hipercementite crônica”. Dentistas, principalmente patologistas orais e endodontistas, devem estar atentos a essa apresentação incomum para estabelecer o diagnóstico correto.

Unitermos: hipercementose; osteomielite; patologia bucal; mandíbula; diagnóstico diferencial.

RESUMEN

La hipercementosis se clasifica como una proliferación no neoplásica que implica un depósito excesivo de cimento a lo largo del cimento radicular normal. Presentamos el caso de un paciente masculino de 77 años que acude al dentista por una lesión supurativa en la encía que evoluciona desde hace varios meses. El examen radiográfico mostró una gran área radiopaca involucrando la raíz del diente 46, que estaba completamente rodeada por un área radiolúcida, sugiriendo un cementoblastoma asociado con el proceso infeccioso. El examen histopatológico del diente afectado reveló hipercementosis extensa asociada con inflamación supurativa crónica, además de osteomielitis crónica localizada. Esta presentación es infrecuente y, debido a los

hallazgos clínico-patológicos, se propone el término “hipercementitis crónica”. Los dentistas, especialmente los patólogos orales y endodoncistas, deben estar tener conocimiento de esta inusual presentación para establecer el diagnóstico correcto.

Palabras clave: *hipercementosis; osteomielitis; patología bucal; mandíbula; diagnóstico diferencial.*

INTRODUCTION

Hypercementosis is a non-neoplastic condition of unclear etiology, characterized by the excessive formation of cementum in the root of one or more teeth. Radiographically, alterations on the anatomy of the dental roots are observed with preserved-support structures, both the periodontal space and the hard lamina⁽¹⁾. Considering the etiology of hypercementosis, local factors include dysfunctional occlusion and periapical pathosis, whereas systemic factors comprise Paget's disease of bone, acromegaly, rheumatic fever, deforming arthritis and thyroid disease⁽²⁻⁵⁾.

Hypercementosis can be misdiagnosed with other radiopaque lesions in close association with dental roots such as idiopathic osteosclerosis, condensing osteitis, osseous dysplasia, ossifying fibroma, central osteoma and osteoid osteoma. Noteworthy, exuberant hypercementosis can mimic cementoblastoma⁽⁶⁻⁸⁾. The current study reports an extremely rare presentation of hypercementosis associated with chronic suppurative inflammation, emphasizing their diagnostic procedures and differential diagnosis.

CASE REPORT

A 77-year-old black man was referred to Oral and Maxillofacial Surgery Service, presenting a “lesion in the right side of the mandible”. The intraoral examination showed grade III mobility of the tooth #46, as well as gingival suppuration. Radiographic analysis revealed a large radiopaque lesion involving the root of the tooth #46, surrounded by a radiolucent area (**Figure 1A**). The surgical approach included the extraction of the tooth #46 and local curettage. On macroscopic examination, the involved tooth showed irregular surface, yellowish-brown color and small hemorrhagic foci (**Figure 2A-C**). After decalcification, the cut surface exhibited gutta percha filling root canal and radicular dentin in close relationship and continuous with large areas of hard tissue (**Figure 2D**). Microscopic examination revealed extensive hypercementosis delimiting peripherally irregular cavities (**Figure 2E**). Moreover, peripheral areas exhibited liquefactive necrosis, cellular detritus and numerous bacterial

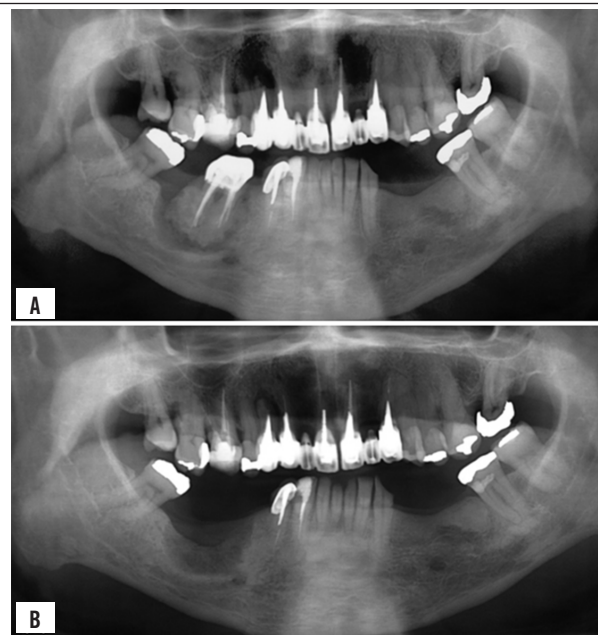


FIGURE 1 – A) pre-operative panoramic radiograph showing a radiopaque lesion, surrounded by a radiolucent halo, involving the root of the tooth #46; B) after 1-year of follow-up, notice the progressive healing of the lesional area

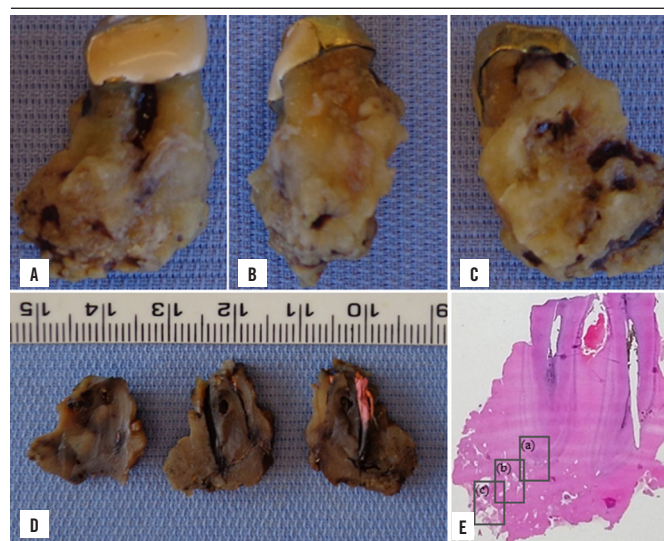


FIGURE 2 – Permanent mandibular right first molar; A, B, and C: macroscopic view of the extracted tooth with lesional hard tissue adhered to root

A) facial surface; B) mesial surface; C) distal surface; D) macroscopic view of the cut surface, showing hard lesional tissue adhered to root and gutta percha filling the root canal; E) microscopic whole view of the lesional tissue adhered to root (HE stain, $\times 2.5$).

HE: hematoxylin and eosin.

colonies occupying the cavities, which also presented varying degrees of cementum resorption (**Figure 3A-C**). The Grocott-Gomori and periodic acid-Schiff stains highlighted numerous filamentous bacteria surrounding the hypercementosis areas, as well as piece of cementum-like mass sequestration (**Figure 3D-G**). Moreover, necrotic bone trabeculae covered by debris and bacterial colonies were evident. The clinicopathological correlation was consistent with the diagnosis of hypercementosis associated with chronic suppurative inflammation (chronic hypercementitis) and localized chronic osteomyelitis. After 2-year of follow-up, the patient is well, without alterations, showing complete repair of the lesional area (**Figure 1B**).

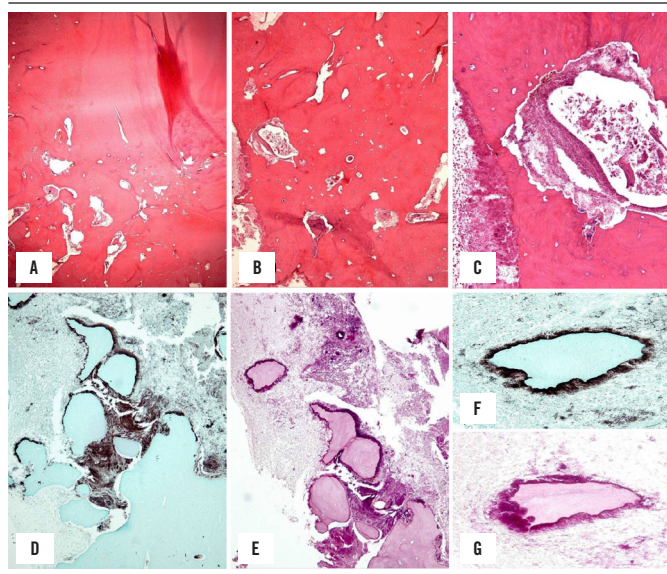


FIGURE 3 – Microscopic view (HE stain)

A) interface showing the intimate contact between the dental root and cementum hyperplasia ($\times 5$); B) hypercementosis exhibiting irregular cavities, some of them empty ($\times 5$); C) peripheral area of the lesional tissue showing numerous bacteria, inflammatory cells and liquefactive necrosis ($\times 20$); D) Grocott-Gomori stain evidencing the presence of numerous filamentous bacteria in close contact with lesional hard tissue ($\times 10$); E) periodic acid-Schiff stain showing bacterial colonies surrounding the lesional hard tissue ($\times 10$); F) Grocott-Gomori; G) periodic acid-Schiff stains showing cementum-like mass sequestration surrounded by filamentous bacteria ($\times 40$).

HE: hematoxylin and eosin.

DISCUSSION

The current study presents an uncommon case of exuberant hypercementosis associated with chronic suppurative inflammation. A previous publication shows that in some cases of hypercementosis, a bacterial contamination is favored due to the rough surface⁽⁵⁾. The histopathological features of the current case show a chronic suppurative inflammatory process

in close association with hypercementosis areas, including foci of cementum resorption and sequestration of cementum-like masses, which are rare findings. The etiology of hypercementosis is not well understood; however, this condition can show association with several factors⁽²⁾. Among local factors, Zhou *et al.* (2012)⁽⁵⁾ reported a case of chronic periodontitis, affecting the teeth #45 and #46, as an probable etiological factor in the hypercementosis formation, suggesting that chronic periodontitis once established, could induce the formation of more layers of cementum (hypercementosis), so as to ensure the permanence of the tooth in the alveolar bone⁽⁵⁾.

Hypercementosis associated with osteomyelitis is a rare event, not previously reported in the English-language literature. In the current case, we report the association of hypercementosis and localized chronic osteomyelitis. Moreover, cementum-like sequestration foci, which were evident at the periphery of the lesion, were also observed. It is noteworthy the degree of hypercementosis and periodontal involvement from previously reported case by Zhou *et al.* (2012)⁽⁵⁾ and the current case, which appears to be gradually and directly involved with the extension of the inflammatory process. Thus, it is important to take into account these clinicopathological features before endodontic treatment planning and/or surgical approach of the teeth affected by hypercementosis.

Such as above mentioned, the differential diagnosis of hypercementosis includes any radiopaque lesion in close relationship with the dental root. However, a distinctive finding is the preservation of the periodontal ligament space surrounding the hypercementosis, which is also visualized in cementoblastoma. Thus, in some cases, the differential diagnosis on imaginological analysis between hypercementosis and cementoblastoma is difficult⁽⁶⁻⁹⁾. In these cases, the histopathological analysis is fundamental to establishing the conclusive diagnosis. In fact, different from hypercementosis, the cementoblastoma is a rare benign odontogenic tumor constituted by plump, active-looking cementoblasts rimming trabeculae and supported by vascular connective tissue⁽⁷⁻⁹⁾. Noteworthy, occasionally, some hypercementosis cases can have the appearance of a severely torn root⁽¹⁾.

In summary, we report an unusual clinicopathological presentation of exuberant hypercementosis associated with chronic suppurative inflammation, for which we think that the term “chronic hypercementitis” is more appropriated. Dentists, especially oral pathologists and endodontists, should be aware of this unusual presentation in order to establishing the correct diagnosis.

SIGNIFICANCE OF THE STUDY

The current case shows, for the first time, an unusual presentation of exuberant hypercementosis associated with chronic suppurative inflammation, clinically mimicking a cementoblastoma. Due to the clinicopathological findings, the term “chronic hypercementitis” is proposed. Dentists, especially oral pathologists and endodontists, should be aware of this unusual presentation in order to establishing the correct diagnosis.

CONFLICT OF INTEREST

All authors declare that they have no conflict of interest.

ETHICAL APPROVAL

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

INFORMED CONSENT

Informed consent was obtained from all individual participants included in the study.

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