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Cisto dentígero e osteomielite com periostite proliferativa: uma atualização diagnóstica

Dentigerous cyst and Osteomyelitis with proliferative periostitis: a diagnostic update

Quiste dentígero y osteomielitis con periostitis proliferativa: una actualización diagnóstica

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Resumo

A infecção do cisto dentígero é uma condição rara que pode levar a situações de risco de vida. Uma situação incomum é o desenvolvimento de osteomielite com periostite proliferativa por infecção desse cisto, com apenas um caso possível publicado na literatura. O objetivo desse artigo é relatar uma associação rara de um cisto dentígero com osteomielite e periostite proliferativa. Um paciente de 11 anos de idade compareceu para o atendimento odontológico com infecção por pericoronite do segundo molar, que atingiu o germe do terceiro molar e evoluiu para osteomielite com periostite proliferativa, tratada sob anestesia geral. O exame histológico dos tecidos removidos durante a cirurgia revelou cisto dentígero infectado e osteomielite com periostite proliferativa. Após a cirurgia, o paciente evoluiu satisfatoriamente com melhora das queixas clínicas. Essa condição, embora rara, deve ser considerada como um diagnóstico diferencial de várias lesões do sistema estomatognático.

Palavras-chave: Cisto dentígero; Infecção; Osteomielite; Periostite.

Abstract

Dentigerous cyst infection is a rare condition that can lead to life-threatening situations. A situation unusual is the development of osteomyelitis with proliferative periostitis from infection of this cyst, with only one possible case published in the literature. The purpose of this article is to relate a rare association of a dentigerous cyst with osteomyelitis and proliferative periostitis. An 11-year-old patient compared for dental care with infection from second molar pericoronitis, who reached the third molar germ and progressing to osteomyelitis with proliferative periostitis, thatwas treated under general anesthesia. Histological exam of the tissues removed during the surgery revealed infected dentigerous cyst and osteomyelitis with proliferative periostitis. After the surgery, the patient progressed satisfactorily with improvement of the clinical complaints. This condition, although rare, should be considered as a differential diagnosis of several stomatognathic system lesions.

Keywords: Dentigerous cyst; Infection; Osteomyelitis; Periostitis.

Resumen

La infección del quiste dentígero es una condición rara que puede conducir a situaciones potencialmente mortales. Una situación inusual es el desarrollo de osteomielitis con periostitis proliferativa debido a la infección de este quiste, con solo un posible caso publicado en la literatura. El propósito de este artículo es informar una asociación rara de un quiste dentígero con osteomielitis y periostitis proliferativa. Un paciente de 11 años se presentó para recibir

atención dental con infección por pericoronitis del segundo molar, que alcanzó el germen del tercer molar y progresó a osteomielitis con periostitis proliferativa, tratada con anestesia general. El examen histológico de los tejidos extirpados durante la cirugía reveló quiste dentígero infectado y osteomielitis con periostitis proliferativa. Después de la cirugía, el paciente progresó satisfactoriamente con una mejora en las quejas clínicas. Esta condición, aunque rara, debe considerarse como un diagnóstico diferencial de varias lesiones del sistema estomatognático.

Palabras clave: Quiste dentígero; Infeccíon; Osteomielitis; Periostitis.

1. Introduction

Dentigerous Cyst (DC) is the second most common among odontogenic cysts representing about 11,4 to 35,5% in different world populations.(Johnson et al., 2014) Its development is due to the accumulation of fluid between the reduced enamel epithelium and the enamel or within the enamel organ.(Devi et al., 2015) Its wide incidence is accompanied by unusual presentations such as multiple lesions,(Devi et al., 2015) associated with other lesions of the stomatognathic system such as calcifying odontogenic cyst,(Aristizabal Arboleda et al., 2018) as well as infectious processes.(Bas et al., 2012; Luyk & Hunter, 1991)

Infection of this cyst is rare, but when it occurs, it may present different evolutions, from less intense alterations, such as increased local bone resorption,(Osaki et al., 1995) to less common conditions such as necrotizing mediastinitis(Bas et al., 2012)and Osteomyelitis with proliferative periostitis (OPP).(Luyk & Hunter, 1991)OPP, also known as Garrè osteomyelitis is a rare condition associated with low potential infection, in which there is a periosteal reaction with bone formation overlying the local bone.(Liu et al., 2019)It often affects the jaw causing slow-growing facial asymmetry and may present with erythema and fistula. Radiographically, there is a sclerotic image parallel to the bony border(Liu et al., 2019; Luyk & Hunter, 1991; Park & Myoung, 2016) and the usual treatment is antibiotic therapy and removal of the cause.(Luyk & Hunter, 1991; Park & Myoung, 2016)

Such condition has differential diagnosis with various facial pathologies such as osteosarcoma, cherubism, histiocytosis, Ewing's sarcoma,(Liu et al., 2019; Park & Myoung, 2016) fibrous dysplasia, exostosis and osteoma.(Park & Myoung, 2016) However, the association between DC and OPP is extremely rare, with only 1 case reported in the literature.(Luyk & Hunter, 1991).

The purpose of this article is to relate a rare association of a dentigerous cyst with

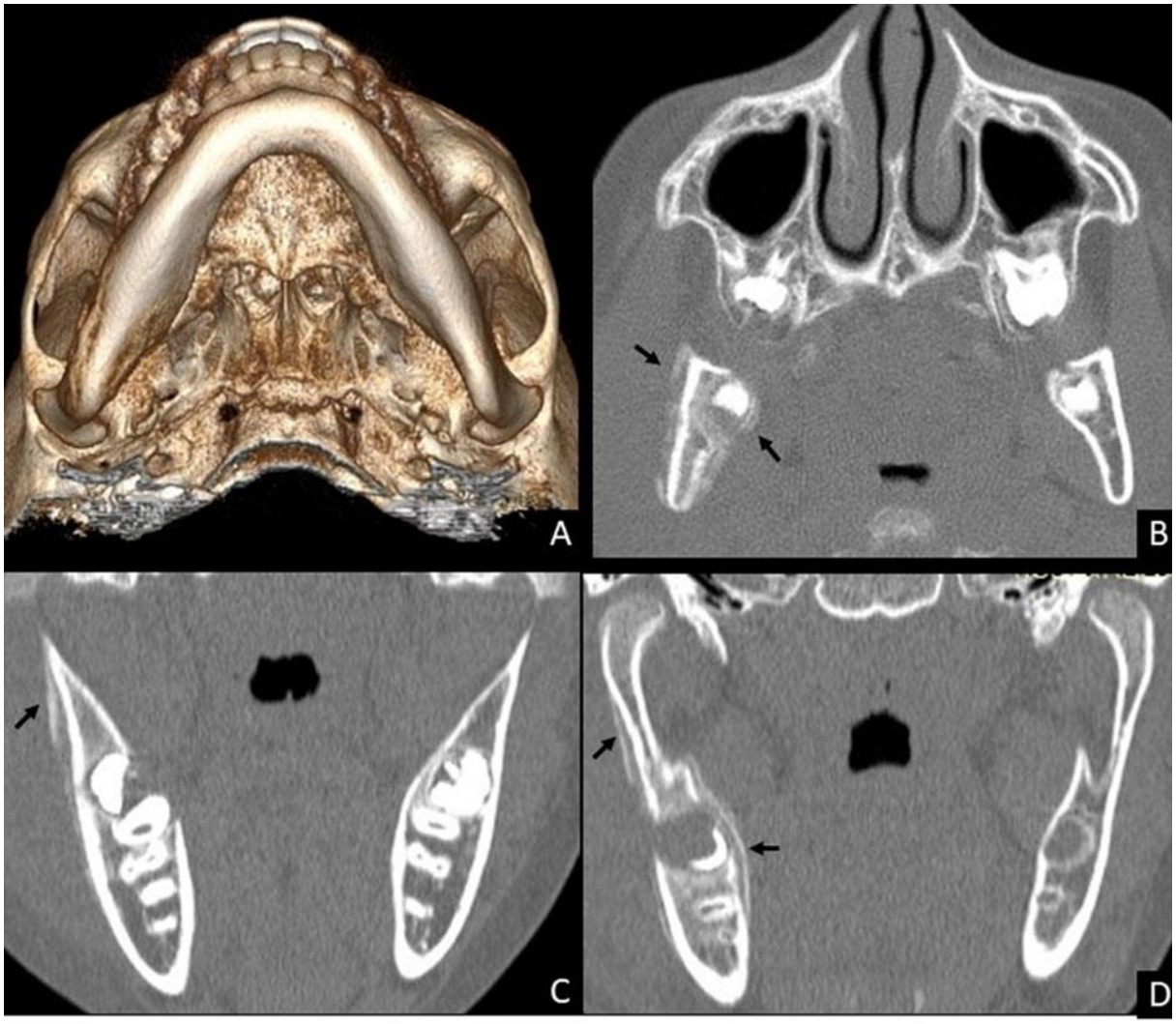
osteomyelitis and proliferative periostitis.

2. Patients and Methods

For this case study the literature search was performed using the strategy: (osteomyelitis OR periostitis) AND dentigerous cyst, without using any search filter, conducted by two independent examiners, which found 151 articles published in the PubMed (20 articles), Embase (96 articles), Scopus (25 articles) and Web of Science (10 articles), however, only one presented a similar case.(Luyk & Hunter, 1991).

An 11-year-old male patient, healthy, was referred to the department of oral and maxillofacial surgery complaining o fright mandibular volume increase (5 months) associated with episode feverish and no history of local trauma. Clinical exam revealed swelling in the right mandibular ramus associated with mouth opening limitation (15 mm). Oroscopy revealed a right lower second molar erupting and swelling in the right retromolar region. Computed tomography showed buccal and lingual bone cortical perforation in 48 dental germ region and periosteal reaction process with sclerotic areas extending from the right mandibular body to the subcondylar portion (Fig. 1).

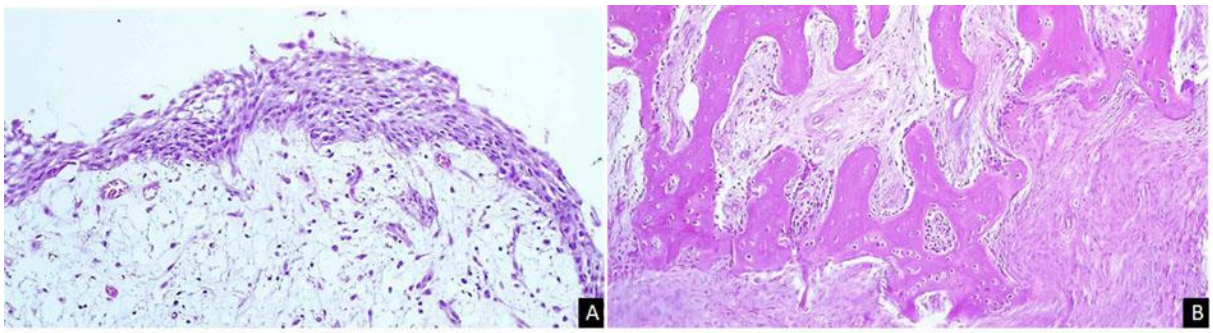
Figure 1 - Computed tomography exam in A (3D reconstruction), B (axial) and C-D (coronal) showing new bone formation (arrow) over lateral and medial cortical surface extending from the mandibular body to the subcondylar region.



Source: Authors.

Under general anesthesia, the patient underwent removal of the third molar and adjacent tissue, as well as part of the mineralized tissue deposited in the vestibular portion. In the intraoperative period, a small amount of purulent content was observed near the newly formed bone. The collected material was sent for histopathological analysis which revealed infected DC and young bone tissue deposited in collagen fibers, with osteoblastic margination (Fig. 2), characterizing, in association with the tomographic examination, the presence of osteomyelitis with periostitis proliferative.

Figure 2 – Histologic features. A) cystic capsule with non-keratinized stratified pavement epithelium of few layers displaying areas of hyperplasia (hematoxylin-eosin, 200×); B) Newly formed bone trabeculae containing typical osteocytes and sometimes exhibiting conspicuous osteoblastic margination, arranged in the midst of inflammatory infiltrate of varying intensity (hematoxylin-eosin,100×).



Source: Authors.

The patient evolved with improvement in mouth opening (21 to 41mm) and submandibular edema reduction from the 30th postoperative day, which was maintained for 2year of follow-up.

3. Results and Discussion

Dentigerous cyst infection is a rare condition and may represent 2.1% of severe head and neck infections.(Smith & Kellman, 2005) With clinical presentation different from that usually found in cysts, it can evolve with trismus,(Bas et al., 2012; Luyk & Hunter, 1991) nausea, vomiting, dysphagia, dysphonia and presence of purulent discharge with leukocytosis and fever,(Bas et al., 2012) characteristics common to odontogenic infections.

In young patients this infection may start in the pericoronal region of the erupting lower second molar, spreading and reaching the third molar germ and may develop into abscesses (Park & Myoung, 2016) and osteomyelitis.(Fukuda et al., 2017; Liu et al., 2019; Luyk & Hunter, 1991; Park & Myoung, 2016) However, there are few published cases of OPP resulting from this type of infection, most of them unrelated to the concomitant presence of cysts (Fukuda et al., 2017; Liu et al., 2019; Park & Myoung, 2016) Only one similar case was reported by Luyk and Hunter (1991) in which there was a DC associated with OPP.

This infectious process has low virulence, which is evidenced by the absence of acute infectious process' classic signs. Usually, there is a slow evolution, absence of changes in

leukogram and C-reactive protein, in addition to the presence of bacteria from the normal oral flora (Luyk & Hunter, 1991; Park & Myoung, 2016) or without any bacterial growth strain.(Liu et al., 2019)

They often reach the jaw and present as a chronic infection with firm swelling on palpation, which leads to facial asymmetry and limited mouth opening. Imaging, whether radiographic or tomographic, reveals a sclerotic area lateral to the mandibular vestibular cortex (Fukuda et al., 2017; Liu et al., 2019; Luyk & Hunter, 1991; Park & Myoung, 2016) and may also reach the lingual region (Park & Myoung, 2016) as seen in the present case. In addition, Fukuda et al. (2017), report the possibility of submandibular lymphadenopathy. When the process is acute, the presence of erythema, fever and fistula with drainage of purulent discharge may be observed (Liu et al., 2019; Park & Myoung, 2016). But, as in the case presented, even at a chronic stage, it is not uncommon to find purulent content when the region is accessed.(Fukuda et al., 2017)

Since the clinical characteristics of cases not associated with DC are similar those observed in associated cases, we reinforce the need for histopathological evaluation of the removed tissue. The importance of this analysis was emphasized by Smith and Kellman (2005) who observed that the infected DC may represent a larger portion than expected in cases of infection requiring hospitalization.(Smith & Kellman, 2005) The hospital is the place of choice for the treatment of OPP, especially in young patients, whether it is associated(Fukuda et al., 2017; Liu et al., 2019; Park & Myoung, 2016) or not with the DC,(Luyk & Hunter, 1991) because this environment enables the procedure under general anesthesia, which is more comfortable for them.

This treatment is often performed with broad-spectrum empirical antibiotic therapy and removal of the cause. Liu et al. (2019) performed a more aggressive approach with cutaneous access and osteotomy to regularize the area.(Liu et al., 2019) In the present case, intraoral access to the lesion was chosen, which progresses with a gradual reduction of edema, as evidenced in other studies.(Fukuda et al., 2017; Luyk & Hunter, 1991; Park & Myoung, 2016)

The histological aspect represented by young bone trabeculae, with the presence of osteoblasts, which are arranged in the midst of fibrous tissue with inflammatory infiltrate, is common to the reaction bone regardless of association with the dentigerous cyst.(Fukuda et al., 2017; Liu et al., 2019; Luyk & Hunter, 1991) These features corroborate to the findings of the present case; however, the histological characteristics of the dentigerous cyst associated with OPP have not yet been showed in the literature.

Lyuk and Hunter (1991), also described an association between OPP and infected DC in a patient of similar sex and age as in the present case. However, these authors did not observe the presence of DC in their histopathological analysis, justifying that such lining was lost due to the evolution of the infection. Thus, this situation allows the interpretation of unpublished of the present report. As found in the present case, the histological aspect of the DC is characterized by the presence of a thin cystic capsule formed by about 2-5 layers of stratified non-keratinized squamous epithelium,(Aristizabal Arboleda et al., 2018; Bas et al., 2012) and areas of mucous metaplasia can be observed, as described by Arboleda et al. (2018).

4. Final Considerations

Thus, stands out the importance of dental follow-up of teeth erupting and highlight the relevance of infected dentigerous cyst as a differential diagnosis of head and neck infections. We highlight the importance of future publication of cases with rare associations, for a better understanding of the pathology.

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