



UNIVERSIDADE FEDERAL DE UBERLÂNDIA
FACULDADE DE ODONTOLOGIA



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**UNUSUAL CLINICAL CASE REPORT:
BILATERAL DENTIGEROUS CYST**

UBERLÂNDIA

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BILATERAL DENTIGEROUS CYST**

Trabalho de conclusão de curso apresentado a Faculdade de Odontologia da Universidade Federal de Uberlândia, como requisito parcial para obtenção do título de Graduado em Odontologia.

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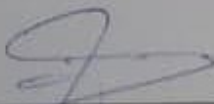


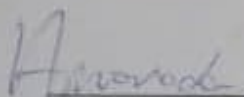
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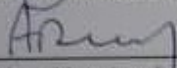
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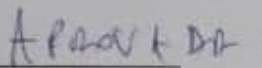
No dia 07 de novembro de 2018, reuniu-se a Comissão Julgadora aprovada pelo Colegiado de Graduação da Faculdade de Odontologia da Universidade Federal de Uberlândia, para o julgamento do Trabalho de Conclusão de Curso apresentado pelo(a) aluno(a) **Beatriz Cristinne Braga Santos**, COM O TÍTULO: "UNUSUAL CLINICAL CASE REPORT: BILATERAL DENTIGEROUS CYST". O julgamento do trabalho foi realizado em sessão pública compreendendo a exposição, seguida de arguição pelos examinadores. Encerrada a arguição, cada examinador, em sessão secreta, exarou o seu parecer. A Comissão Julgadora, após análise do Trabalho, verificou que o mesmo se encontra em condições de ser incorporado ao banco de Trabalhos de Conclusão de Curso desta Faculdade. O competente diploma será expedido após cumprimento dos demais requisitos, conforme as normas da Graduação, legislação e regulamentação da UFU. Nada mais havendo a tratar foram encerrados os trabalhos e lavrada a presente ata, que após lida e achada conforme, foi assinada pela Banca Examinadora.

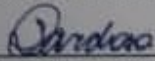
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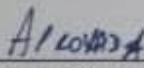

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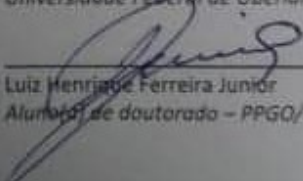

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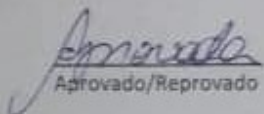

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Abstract

The dentigerous cyst, also known as follicular cyst, is a developmental odontogenic cyst and the second most frequent odontogenic cyst in the jaws. It occurs in a high age of life, which is asymptomatic in the early, and its progression can lead to a facial ablation of the local cortical bone. It is derived from the reduced epithelium of the enamel organ, and is observed in a crown of an unerupted tooth, but with idiopathic etiopathogenesis.

The bilateral dentigerous cyst is generally associated to a syndrome or other pathological alterations, just like the cleidocranial dysplasia, syndromes of Maroteaux Lamy and Gorlin Goltz, therefore in the absence of syndromes, the bilateral condition is rare. This study presents a rare clinical case of bilateral dentigerous cyst in the mandible of a 20-year-old non-syndromic young man of the Program of Specific Cares to Stomatologic Diseases (PROCEDE) of the Dentistry School of the Federal University of Uberlandia. The radiographic image showed a well delimited unilocular radiolucent area in the body and ramus of mandible, and the diagnosis was confirmed by histopathological examination.

Key- words: Bilateral Dentigerous Cyst; Mandible; Non-syndromic

Resumo

O cisto dentífero, também conhecido como cisto folicular, é um cisto odontogênico de desenvolvimento e o segundo cisto odontogênico mais frequente nos maxilares. Ocorre em ampla faixa etária, geralmente é assintomática, e sua progressão pode ocasionar rompimento da cortical ossea. É derivado do epitélio reduzido do órgão do esmalte e é observado envolvendo a coroa de dente não irrompido, mas com etiopatogenia idiopática. O cisto dentífero bilateral é geralmente associado a uma síndrome ou outras alterações patológicas, assim como a displasia cleidocraniana, as síndromes de Maroteaux Lamy e Gorlin Goltz, portanto, na ausência de síndromes, a condição bilateral é rara. Este estudo apresenta um caso clínico raro de cisto dentífero bilateral na mandíbula de um jovem de 20 anos, não síndrômico, do Programa de Cuidados Específicos para Doenças Estomatológicas (PROCEDE) da Faculdade de Odontologia da Universidade Federal de Uberlândia. A imagem radiográfica mostrava uma área radiolúcida unilocular bem delimitada no corpo e no ramo da mandíbula, e o diagnóstico foi confirmado pelo exame histopatológico.

Palavras-chave: Cisto dentífero bilateral, mandíbula, não síndrômico

Introduction

Dentigerous cyst (DC) is classified as an odontogenic cyst that is attached to the cervical region of an unerupted tooth and envelops its own crown¹. Regarding its pathogenesis, it has been suggested that the pressure exerted by an erupting tooth on the follicle may obstruct venous flow inducing accumulation of exudates between the reduced enamel epithelium and the tooth crown². This is the most common type of odontogenic developmental cyst, accounting for about 20% of all epithelium-lined cysts in gnathic bones³. DC is frequently associated with impacted/unerupted mandibular third molars, followed by maxillary canines and then the maxillary third molars⁴. Although the DC can be found in patients with a wide range of age, they are found more often in patients between 10 and 30 years. There is a slight male predilection and a higher prevalence in whites than in blacks³.

Frequently the DC is discovered when radiographs which are taken to investigate a failure of dental eruption, a missing tooth or misalignment. There is no pain or discomfort associated with the cyst unless it becomes secondarily infected. Radiographs shows an unilocular radiolucent lesion characterized by well-defined sclerotic margins, and associated with the crown of an unerupted tooth. While normal follicular space is 3–4 mm, a DC can be suspected when the space is more than 5 mm⁵. However, differential diagnosis must be made with alterations such as a dental follicle or a hyperplastic dental follicle and even an odontogenic keratocyst or unicystic ameloblastoma⁶.

Multiple bilateral cysts or bilateral dentigerous cyst (BDC) usually are associated with some of syndromes including cleidocranial dysplasia,

Maroteaux Lamy and Gorlin Goltz syndromes, therefore, in the absence of these syndromes, the occurrence of this condition is extremely rare⁷⁻⁸.

The purpose of this study is to relate a rare clinical case of bilateral dentigerous cyst non-syndromic in mandible, involving the left second molar and the right third molar, both impacted and unerupted, in a young adult male.

Case report

Male patient, feoderma, 20 years old, attended the Special Care Program for Stomatological Diseases (PROCEDE) of the School of Dentistry of the Federal University of Uberlandia (FOUFU), sent by him orthodontist, with radiographic findings of bilateral radiolucent images in the mandible, associated with second and third right molars and the left second molar tooth and, being all unerupted, besides the unerupted upper canines teeth. Intraoral examination revealed absence of the second and third lower molars, on both sides, with no bilateral increase in volume of the bone cortical in the mandible, and absence of the upper canines (Figures 1, 2A and 2B). The patient does not have any of the syndromes reported in the literature. The panoramic radiography (Figure 3) revealed in the right side a unilocular radiolucent area delimited by a radiopaque halo, which was associated with the crown of the second and third lower molars, being the third molar inverted toward the crown of the unerupted second molar, and in the left side another radiolucent area associated in the crown of the lower second molar with a horizontal position in the mesial direction toward to the distal root of the left first molar, with resorption signs in this root. The upper canines were revealed impacted by radiographic

examination, with no signs of pathological alteration. Computed tomography (CT) revealed on the right side an intimate relation of the lesion to the crown of the lower right third molar, with thinning and rupture of the bone lingual plate (Figures 4 and 5). Imaging and clinical examinations led us to the first hypothesis of diagnosis of bilateral dentigerous cyst, with secondary hypotheses of odontogenic keratocyst or unicystic ameloblastoma. Aspiration puncture was performed, with negative result. Then incisional biopsy was performed, at the same time as surgical decompression under local anesthesia, on both sides (Figures 6A and 6B), and the histopathological analysis confirmed the diagnosis of bilateral dentigerous cyst (Figures 7A and 7B). Histopathological analysis of the right lesion revealed an apparent stratification of mucosal lining epithelium, with congestive capillaries, oedema and some leukocytes scattered on the connective tissue, with no signs of differentiation (Figure 7A), and the left lesion revealed a stratified epithelium with no pavement differentiation apparent, presence of leukocytes in loose connective tissue (Figure 7B).



Figure 1: Absence of the right and left upper canines.

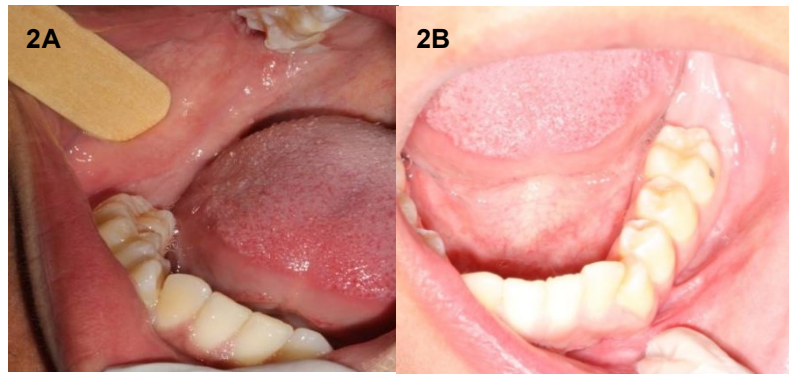


Figure 2: (A) Clinical examination in the right side with absence of the second and third molars, with no volumetric increase of the vestibular bone plate; (B) Clinical examination in the left side with absence of the second and third molars, with no volumetric increase of the vestibular bone plate.



Figure 3: Panoramic radiography revealing a well delimited radiolucent area with an unilocular aspect involving the crown of the inverted right third molar and the right second molar, and a similar image in the left second molar, with a slight resorption sign in the distal root of the left first molar.

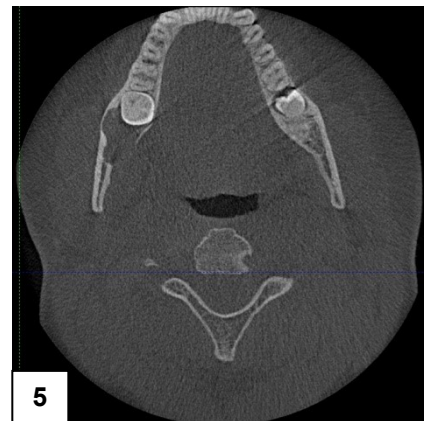


Figure 4: Sagittal section showing the relationship of the lesion with the crown of the inverted right third molar.

Figure 5: Axial section showing thinning and rupture of the lingual bone plate on the right side.

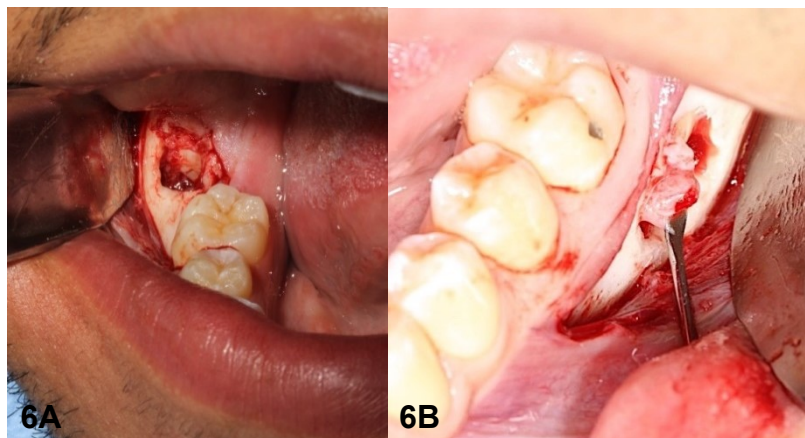


Figure 6: (A e B) incisional biopsy of the right and left sides, respectively.

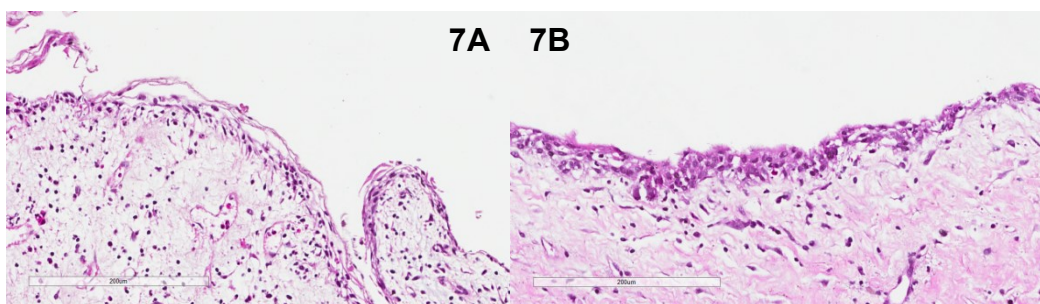


Figure 7A: Histological image revealing an apparent stratification of the mucosa lining epithelium, with congestive capillaries, oedema and some leukocytes scattered on the connective tissue, with no signs of differentiation. H /E coloration.

Figure 7B: Histological image revealing a stratified epithelium, with no apparent pavement differentiation, scarce presence of leukocytes in loose connective tissue. H/E coloration.

Discussion

Although the dentigerous cyst is the most common type of odontogenic developmental cyst, the occurrence of multiple cases or even bilateral cysts is extremely rare in non-syndromic patients. As noted in Table 1, there have been only 36 cases of multiple non-syndromic reported in the world literature since 1943⁹, to 2018.

Table 1- Review of literature regarding to the bilateral or multiple dentigerous cysts⁹, updated until to 2018, totalling the 36 rare cases.

Authors/ Year	Gender	Age (years)	Location	Treatment
Meyers/ 1943 ⁷	F	19	Md. third molars	Enucleation
Henefer/ 1964 ⁹	F	52	Mx. third molars	Enucleation
Stanback/ 1970 ⁹	M	9	Md. first molars	Enucleation
Callaghan/ 1973 ⁹	M	38	Md. third molars	Enucleation
Burton, Scheffer 1980 ⁹	F	57	Md. third molars	Enucleation
Swerdloff, Alexander, Ceen, Fergusson/ 1980 ⁹	F	7	Md. first molars	Enucleation
Crinzi/ 1982 ⁹	F	15	Md. third molars	Enucleation
McDonnell/ 1989 ⁹	M	15	Md. second premolar and second molar	Enucleation
Eidinger/ 1989 ⁹	M	15	Md. first molars	Enucleation
O'Neil, Mosby, Lowel/ 1989 ⁹	M	5	Md. first molars	Enucleation
Banderas, Gonzalez, Ramirez, Arroyo/ 1996 ⁹	M	38	Md. third molars	Enucleation
Sands, Tocchio/ 1998 ⁹	F	3	Md. central incisors and first molars	Enucleation
Ko, Dover, Jordan/ 1999 ⁹	M	42	Md. third molars	Enucleation
De Biase, Ottolenghi, Pollimeni, Benvenuto, Lubrano, Magliocca/ 2001 ⁹	M	8	Md. first molars	Enucleation
Shah, Thuau, Beale/ 2002 ⁹	M	39	Md. third molars	No treatment
Ustuner, Fitoz, Atasoy, Erden, Akyar/ 2003 ⁹	M	6	Mx. Canines	Enucleation
Batra, Roychoudhury, Balakrishan,	F	15	Md. third molars and second	Enucleation

Parkash/ 2004 ⁹			premolar	
Garcia RG, Hernandez VE, Moreno AC, Diaz PLM, Perez JS, Campo FJR/ 2005 ¹⁰	M	28	Md. third molars	Extraction molars
Freitas DQ, Tempest LM, Sicoli E, Lopes- Neto FC/ 2006 ⁹	M	14	Mx. third molars and Md. Second molar	Enucleation
Turkiye/ 2007 ¹¹	M	51	Md. third molars	Enucleation
Fregnani ER, <i>et al.</i> / 2008 ¹²	M	5	Md. first molars	Enucleation
Maurette PE, Allais M, Morais M, Jorge J./ 2008 ¹³	M	7	Md. first molars	Descompression
Chew YS, Aghabeigi B./ 2008 ¹⁴	F	30	Md. third molars	Spontaneous regression
Cury S, Cury M, Cury S, Pontes F, Pontes H, Rodini C, Pinto D./ 2009 ¹⁵	M	5	Md. permanent first molars	Enucleation
Kannan N, Patil Rajendra, Sreenivasulu P./ 2010 ¹⁶	M	32	Mx. Canines	Enucleation
Saluja JS, Ramakrishnan MJ, Vinit GB, Jaiswara C/ 2010 ¹⁷	M	22	Mx. Incisors and second premolars and Md. canines, right first premolar and second premolars	Enucleation
Prasad LK, Chakravarthi PS, Sridhar M, Ramakumar Y, Kattimani V/ 2010 ¹⁸	F	12	Mx. canines and Md. lateral incisors	Enucleation
Reddy PM, Nanjuda, Tavane PM, Kailash, Gupta PK/ 2011 ²	F	11	Md. first and second premolars	Enucleation
Shirazian S, Agha-Hosseini F/ 2011 ¹⁹	M	10	Md. second premolars	Marsupialization
Tamgadge A, Tamgadge S, Bhatt D, Bhalerao S, Pereira T, Padhye M./ 2011 ²⁰	M	10	Mx. right second premolar and left canine.	Enucleation
Ishihara Y, Kamioka H, Takano-Yamamoto T, YamashiroT / 2012 ²¹	M	13	Md. second premolars, Md. left third molar and Mx. right second premolar	Marsupialization
Imada TSN, Neto VT, Bernini GF, Silva Santos PS, Rubira-Bullen I, Bravo-Calderon D, <i>et al.</i> / 2014 ²²	F	42	Md. third molars	Enucleation after marsupialization
Parvathi Devi, V. B. Thimmarasa,	M	17	Mx. central and lateral incisors,	Enucleation

Vishal Mehrotra, Mayuri Agarwal/ 2015 ²³			and left canine. Md. left canine and second premolar and right first and second premolars	
Jeon, Park, Cho, Hwang/2016 ²⁴	M	15	Mx. and Md. third molars	Enucleation
Dhupar A, Yadav S, Dhupar V, Mittal HC, Malik S, Rana P/ 2017 ²⁵	M	8	Mx. right canine and first premolar; Md. left first and second premolars	Enucleation
Khandeparker RV, Khandeparker PV1 Virginkar A, Savant K/ 2018 ²⁶	M	10	Mx. right second premolar permanent and left canine permanent	Enucleation

The age range reported cases varies from 3 years to 57 years of age⁹, the mean age of the 36 cases was 20.1 years, with male predilection (2.3: 1). Of these, 24 cases^{2, 5, 7, 9-15, 19} affected the mandible, 5 cases^{9, 16, 20, 22, 26} affected the maxilla and 7 cases^{9, 17, 18, 21, 23-25} affected both gnathic bones. Regarding to the treatment, enucleation was performed in 29 cases^{5, 7, 9, 11, 12, 15- 18, 20, 23-26}, marsupialization in 2 cases^{19, 21}, spontaneous regression occurred in 1 case¹⁴, marsupialization was performed and enucleation was performed in 1 case²², decompression was performed in 1 case¹³, extraction of the involved elements was performed in 1 case¹⁰ and an untreated case was reported⁹. In the present case affects second and third mandibular molars, in a patient with 20 years of age, who was submitted to bilateral decompression (Fig.: 7 e 8).

Multiple dentigerous cysts can also be induced by prescribed drugs, the combined effect of cyclosporine and a calcium channel blocker is reported to cause bilateral dentigerous cyst^{20, 22, 25}. In our case, there were no clinically evident syndromes, the patient did not report cyclosporine or calcium blockers, however, he has hypothyroidism and uses Puran 150mg (sodium levothyroxine). No relationship was found in the world literature between

hypothyroidism and dentigerous cysts. In all cases found in the literature, including the present case, the radiographic examination were described as unilocular radiolucent area, delimited by a radiopaque halo, associated with the crown of unerupted dental elements. In the present case, the upper canines are impacted, however, with normal follicular space, without evidence of a dentigerous cyst.

Computed tomography is necessary to obtain information about the origin, size, content, cortical plates and the relationship with the adjacent anatomical structures.^{9, 23} In this case, through computed tomography it was possible to identify thinning and cortical rupture of the lingual bone plate on the right side, and the relationship of the lesion with the right lower third molar.

Although the radiographic images offer important information, the definitive diagnosis can be only confirmed through the histopathological analysis, seeing that odontogenic keratocysts and unicystic ameloblastoma present similar radiographic characteristics.^{5, 9, 17} It is not possible to differentiate the dentigerous cyst of ameloblastoma through clinical examination and radiographic examination. As for odontogenic keratocysts, are they more likely to have periphery scalloped, while dentigerous cysts are more likely to have smooth periphery.^{17, 25}

Conclusion

The occurrence of de bilateral dentigerous cyst is rare in non-syndromic patients and that anatomopathological examination is essential for its diagnosis, besides its clinical and radiographical features.

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