Queratocisto odontogêncio ou cisto dentígero? Desafio no diagnóstico

Odontogenic keratocyst or dentigerous cyst? A diagnostic challenge

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ABSTRACT
The odontogenic keratocyst is a benign cystic lesion that preferentially affects the posterior region of the mandible, having an infiltrative and aggressive feature with unique and varied behavior, high recurrence rates, and distinct molecular findings among odontogenic lesions. Also, its differential diagnosis and treatment is a challenge, due to the presence of other lesions of the jaw with similar aspects. Objective: The present study aims to report a clinical case of odontogenic keratocyst associated with a third molar included in an ectopic position, focusing on its radiographic and tomographic characteristics that are confused with a dentigerous cyst. Conclusion: The combined use of cone beam computed tomography, 3D segmentation and magnetic resonance imaging enable a complete analysis of maxillomandibular lesions from the bone and soft tissue point of view, favoring the development of a more accurate diagnosis of lesions, as in this case, the odontogenic keratocyst it has characteristics that are confused with other types of lesions in which, due to its complexity, the conciliated imaging exams were essential and from this one can also direct the conduct in surgical treatment more effectively.

Key-words: Cone-Beam Computed Tomography; Magnetic Resonance Imaging; Diagnostic Imaging

RESUMO
O queratocisto odontogênico é uma lesão cística benigna que acomete preferencialmente a região posterior da mandíbula possuindo caráter infiltrativo e agressivo com um comportamento único e variado, altas taxas de recorrência e achados moleculares distintos entre as lesões odontogênicas. Além disso, o seu diagnóstico diferencial e tratamento é um desafio, devido à presença de outras lesões da mandíbula com aspectos semelhantes. Objetivo: O presente estudo tem por objetivo relatar um caso clínico de queratocisto odontogênico associado a um terceiro molar incluso em posição ectópica dando enfoque as suas características radiográficas e tomográficas que se confundem com um cisto dentígero. Conclusão: O uso combinado de tomografia computadorizada de feixe cônico, segmentação 3D e ressonância magnética possibilitam uma análise completa das lesões maxilomandibulares do ponto de vista ósseo e de tecidos moles, favorecendo o desenvolvimento de um diagnóstico mais preciso das lesões, como neste caso, o ceratocisto odontogênico possui características que se confundem com outros tipos de lesões em que, pela sua complexidade, os exames de imagem conciliados foram imprescindíveis e a partir disso também pode-se direcionar a conduta no tratamento cirúrgico de forma mais eficaz.

Palavras-chave: Tomografia Computadorizada de Feixe Cônico; Imagem por Ressonância Magnética; Diagnóstico por Imagem

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INTRODUCTION

The odontogenic keratocyst is, by meaning, a benign developmental cyst that presents an aggressive and infiltrative feature affecting the buccomaxillofacial complex. With a predilection for posterior mandible region, male patients, and of variable age, which can occur from the first decade of life to the ninth, there is a predominance in patients aged 20 to 30 years.

Due to its aggressive, infiltrative behavior and high recurrence rates in 2005, it was classified by the World Health Organization (WHO) as an odontogenic tumor. However, in 2017, WHO reclassified it as an odontogenic cyst of development under the justification that there is insufficient evidence to maintain it in the classification of neoplasms.

Radiographically, it is possible to observe a radiolucent area, very delimited, uni or multilocular, which may present a sclerotic halo, regular or wavy margins, with or without involvement with an included tooth. There may be displacement of impacted or erupted teeth, extrusion of erupted teeth and root resorption. These are other possible radiographic findings.

Histologically, its aspect is characterized by a cystic capsule integrated by a stratified squamous epithelium, with ortho-parakeratin, a fibrous tissue wall with satellite cells and a layer of palisaded basal cells.

The form of treatment can be conservative or radical, depending on the aggressiveness of the lesion. Regarding the conservative therapeutic possibilities are marsupialization and decompression. Aggressive approaches range from enucleations to resections with curettage removing the lesion in one piece, with the possibility of adjuvant therapy to the application of carnoy solution, cryotherapy, or peripheral osteotomy.

This study aims to report a clinical case of an odontogenic keratocyst associated with an included third molar in an ectopic position focusing on its radiographic and tomographic characteristics that can be confused with a dentigerous cyst and the characteristics of magnetic resonance images that can favor this differential diagnosis.

CASE REPORT

A 55-year-old male patient sought dental care complaining of a “wound” on the left side of his mouth that did not heal. In the anamnesis, it was reported by the patient that sometimes the involved area swelled and the enlargement disappeared when he felt that a liquid was coming out of the opening (Figure 1).

![Figure 1](image-url) Intraoral examination in which it is possible to observe an opening in the posterior region of the mandible on the left side associated with an edentulous alveolar ridge with a leukoplasic aspect.

Then, a panoramic radiograph was requested (Figure 2), which showed tooth 38 included, projected inverted in the posterior region of the left mandibular branch and presenting a radiolucent image extending from the crown of the dental element to the region of ipsilateral retromolar trine suggesting pathway drainage of a possible dentigerous cyst or attempted surgical access to the tooth.
After analyzing the panoramic radiography, it was indicated to perform a cone-beam computed tomography that showed a hypodense tomographic image associated with the crown of the semi-included tooth 38, located in the lingual region of the left mandibular branch with free roots and an intraosseous crown in close relationship with the mandibular foramen. In the segment of the drainage channel, areas of disruption of the buccal and lingual bone cortex are observed, functioning as possible drainage areas for the intralesional content (Figure 3).

Figure 2. Panoramic radiography showing tooth 38 inverted and radiolucent region suggesting drainage route or an attempt of surgical access to the tooth.

Figure 3. A - Panoramic reconstruction showing tooth 38 included and the entire length of the drainage channel; B - 3D reconstruction with lingual view; C - Tooth 38 semi-enclosed with intraosseous crown and free roots; D - Relationship of proximity of the crown to the mandibular foramen; E - Disruption of the buccal and lingual bone cortex in different regions of the continuity of the drainage channel; F - Distance of the drainage channel in relation to the continuity of the mandibular canal.
As an auxiliary method to better characterize the atypical context of the lesion, the software InVesalius4 v. 3.1.1 for volume calculation, morphological analysis and assistance in virtual planning of the clinical case (Figure 4). InVesaluis is free and public for the health area, which performs analysis and segmentation of virtual anatomical models and is available for download at www.cti.gov.br/invesalius. The final value of the cyst volume was 2.504 mm³ of the drainage channel and 1,223 mm³ of the tooth involved. After performing the 3D reconstruction of the lesion using a manual segmentation process, it was possible to observe more clearly which regions the cyst caused the cortical rupture.

**Figure 4:** InVersalius software; A, B and C - Reconstruction of the lesion using the method of manual segmentation of slices from computed tomography and the bone structure reconstructed using the automatic segmentation method. D and E - Representative images of the cyst and tooth, with superficial details and presentation of the final volume of the lesion and tooth involved.

As requested by the temporomandibular dysfunction professional, the patient underwent magnetic resonance scan (Figure 5) whose images were used to assess the lesion. The sagittal T2W revealed an intermediate signaling image occupying the entire left branch of the mandible, rupture of the lingual and buccal cortex and extension to the oral cavity. It was possible to observe well-defined edges of the lesion without invasion of muscle tissue. The images showed evidence of image of hypersignal parts inside the lesion, also corresponding to possible secondary infection due to clinical findings of swelling sensation and then drainage of fluid by the patient.
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Figure 5: A, B and C showing sagittal T2W sections showing an intermediate signal with foci of hypersignal.

After analyzing the images, tooth 38 was removed and the lesion was excised. In the histopathological examination (Figure 6), the histological sections revealed an epithelium with uniform thickness and parakeratosis, often showing a wavy and palisade surface of the basal cell nuclei, which present a flat interface with the connective tissue, fragments of keratin fill the lumen. Such characteristics led to the diagnosis of odontogenic keratocyst.

Discussion

In the diagnosis of these types of lesions, panoramic radiography is usually the first choice of image examination by dental surgeons. However, because it is a two-dimensional image, the overlapping craniofacial structures generate images with limitations such as distortions and enlargements ample. And for a diagnosis of a lesion to be possible, it depends on the association of many criteria such as the location, demarcation of the lesion, involvement of the cortical bone, periosteum, density and soft tissue changes related to the adjacent structures within the mandible. Therefore, the Cone Beam Computed Tomography generates images that are dimensionally accurate and anatomically accurate for the diagnosis of lesions in the gnathic bones, such as the odontogenic keratocyst.
During the analysis of the images obtained from the CBCT, the risk of injury to adjacent anatomical structures, such as teeth and nerves, is assessed, and the risk of mandibular fracture is observed depending on the extent of the injury. There is also the advantage of the reduced radiation dose compared to the medical tomography combined with the high quality of the images that enable the dental surgeon to have accurate three-dimensional images about the extent, limits and the exact measurement of the lesion to be observed\textsuperscript{11,12}.

Furthermore, 3D segmentation of anatomical structures plays an essential role during planning for the treatment of craniofacial injuries. Also, the mandibular structural complexity makes the process of manual segmentation in CT scans a challenge\textsuperscript{13,14}. In the present case, the association with the included third molar (tooth 38) suggested as a differential diagnosis of the dentigerous cyst, on tomographic images and 3D reconstruction by manual segmentation showed rupture of the cortical bone by lingual and buccal and proximity to the mandibular foramen of the tooth involved with the injury. Although the CBCT provides details on the extent of the lesion at the bone level, the limitations of soft tissue information can restrict the course of action to be taken for the treatment of the lesion.

In this context, magnetic resonance imaging is a non-invasive method that offers superior images of the internal composition of a lesion and greater details about the soft tissues\textsuperscript{15}. The investigation of the lesion content may be essential to distinguish keratocysts from other odontogenic lesions, such as the dentigerous cyst in this case, which tend to present intermediate T1WI homogeneous signal intensity and high homogeneous signal intensity in the T2WI sequence\textsuperscript{16}.

As in keratocysts, the intermediate or high-intensity signal in the T1WI sequence and heterogeneous low to high signal intensity in the T2WI sequence may reflect the presence of material in the cystic lumen that in the case of the odontogenic keratocyst consists of keratin fragments, or components of secondary infection on microscopic examination\textsuperscript{15-17}.

In this case, the patient had undergone an MRI to evaluate the temporomandibular joint as indicated by the TMD professional and the images were used to analyze the lesion and the aspects described in the literature to distinguish the type of cyst were consistent with the histopathological examination of odontogenic keratocyst.

**CONCLUSION**

The combined use of cone beam computed tomography, 3D segmentation and magnetic resonance imaging enable a complete analysis of maxillomandibular lesions from the bone and soft tissue point of view, favoring the development of a more accurate diagnosis of lesions, as in this case, the odontogenic keratocyst it has characteristics that are confused with other types of lesions in which, due to its complexity, the conciliated imaging exams were essential and from this one can also direct the conduct in surgical treatment more effectively.

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