

## Importance of Imaging Exams in the Detection and Enucleation of Odontogenic Keratocyst: a Case Report

### Importância dos Exames de Imagens na Detecção e Enucleação de Queratocisto Odontogênico: Relato de Caso

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#### Abstract

The Odontogenic Keratocyst (QO) is a cyst of epithelial development of the jaws derived from the enamel organ or the dental lamina, which corresponds to approximately 11% of all maxillary cysts. Cysts are benign and generally asymptomatic lesions, but they can reach large sizes, causing tooth displacement, bone expansion, and facial deformity. The objective of this study is to report the total enucleation of a keratocyst in the posterior region of the mandible as an alternative treatment. Patient M.Y.S.B. 20 years old, female, ASA I, was referred to the Dental Specialties Center in Cachoeira-BA, for extraction of tooth unit 48. The panoramic radiographic examination and computed tomography showed a unilocular radiolucent image, extending from the mandibular ramus to the region of tooth 47, with apical displacement of the mandibular canal. The samples were sent for biopsy and a QO with low-grade focal epithelial dysplasia and the presence of satellite cysts was found. We opted for complete enucleation of the lesion and extraction of element 48. Radiographically, QO lesions may present a unilocular or multilocular aspect due to their size. Techniques vary from conservative treatments such as enucleation, decompression, to more aggressive treatments with partial or total resections. With this study, the importance of using image exams and biopsy for the diagnosis was observed in order to choose the effective treatment for QO.

**Keywords:** Odontogenic Cysts. Enucleation Surgery. Maxillofacial Surgery.

#### Resumo

O Queratocisto Odontogênico (QO) é um cisto de desenvolvimento epitelial dos maxilares derivado do órgão do esmalte ou da lâmina dental, que corresponde aproximadamente a 11% de todos os cistos maxilares. Os cistos são lesões benignas e geralmente assintomáticas, mas podem atingir grandes dimensões, provocando afastamentos de dentes, expansão óssea e deformidade facial. O objetivo deste estudo é relatar a enucleação total de um queratocisto na região posterior da mandíbula como alternativa de tratamento. Paciente M.Y.S.B. 20 anos, sexo feminino, ASA I, foi encaminhada ao Centro de Especialidades Odontológicas em Cachoeira-BA, para exodontia da unidade dentária 48. Ao exame radiográfico panorâmico e tomografia computadorizada observa-se imagem radiolúcida unilocular, estendendo-se desde o ramo mandibular até a região do dente 47, com deslocamento apical do canal mandibular. As amostras foram encaminhadas para biópsia e constatou-se um QO com displasia epitelial de baixo grau focal e com a presença de cistos satélites. Optou-se pela enucleação completa da lesão e extração do elemento 48. Radiograficamente, as lesões do QO podem apresentar um aspecto unilocular ou multilocular devido ao seu tamanho. As técnicas variam desde tratamentos conservadores como a enucleação, descompressão, até tratamentos mais agressivos com ressecções parciais ou totais. Com este estudo, observou-se a importância da utilização dos exames de imagem e biópsia para o diagnóstico para assim escolher o tratamento eficaz para o QO.

**Palavras-chave:** Cistos Odontogênicos. Cirurgia de Enucleação. Cirurgia Maxilofacial.

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#### 1 Introduction

The Odontogenic Keratocyst (OKC) is a developmental cyst characterized by a keratinized epithelial lining and aggressive behavior in the maxillomandibular complex. It is most commonly found in the mandibular region and has a high recurrence rate. It may or may not be associated with a dental element<sup>1,2</sup>. The keratocyst or OKC is currently considered again as an odontogenic keratocyst according to the new 2017 WHO classification<sup>3</sup>. Genetic factors are closely related to its development, either linked to its own epithelium or to the enzymatic performance in the fibrous wall of the cyst<sup>4</sup>.

The OKC is an epithelial developmental cyst of the jaws

derived from the enamel organ or dental lamina, accounting for approximately 11% of all jaw cysts<sup>5</sup>. It has a prevalence in patients over the age of 50 in 37.7% of cases and in 28.56% of cases in those under 20 years old. It shows a predilection for the male sex and white ethnicity in 67.8% of cases<sup>6</sup>.

Cysts are benign and generally asymptomatic lesions, but they can reach large sizes, causing teeth displacement, bone expansion, and facial deformity. Therefore, if diagnosed early, the aggressiveness of the surgical treatment will be reduced. Because they are usually asymptomatic, many are accidentally discovered in routine radiographs. To determine the most appropriate surgical treatment, factors of recurrence

of the lesion with destructive and expansive characteristics are discussed<sup>6-8</sup>.

Radiographically, the lesions may appear unilocular or multilocular, needing more effective imaging exams for detection and treatment planning. Most of the images may appear as well-defined unilocular radiolucent areas related to the crown of a tooth, raising questions about the differential diagnosis, which should include entities such as the dentigerous cyst, ameloblastoma, and calcifying odontogenic cyst. However, when not associated with a tooth, the traumatic bone cyst, central giant cell granuloma, and lateral periodontal cyst can be considered. It is important to emphasize that the definitive diagnosis should not be made solely based on clinical and radiographic information; histopathological analysis is of fundamental importance<sup>4,5</sup>.

Diagnosis plays a fundamental role on guiding treatment and determining the prognosis, knowing that all epithelium responds to the inflammatory process<sup>5</sup>. A definitive diagnosis is given by biopsy, characterized by the presence of the histopathological variant known as the parakeratinized form, since the orthokeratinized variant is classified as a separate entity, receiving the name orthokeratinized odontogenic cyst<sup>9</sup>.

Its treatment is still controversial. Techniques range from conservative treatments such as enucleation and decompression to less conservative treatments like partial or total resections. Enucleation and decompression have proven effective in treating keratocysts and present lower morbidity. To reduce recurrences, there are adjunct treatments such as the use of Carnoy's solution and cryotherapy, which penetrate the adjacent bone, causing chemical destruction of the remaining cells<sup>4</sup>.

The recommended treatment is surgical, and various techniques have been reported, among which the most important are the total removal of the cystic capsule and the cyst peripheral ostectomy<sup>6</sup>. Decompression treatment through marsupialization, followed or not by enucleation, is used for large cysts and in young patients. The technique of cauterization, together with electrocautery or the application of Carnoy's solution, is considered. According to Aciole (2010)<sup>5</sup>, lesions that involve adjacent soft tissues, with rupture of the bony cortices, indicate a more aggressive treatment, such as bone resection with clear margins, justified by the risk of recurrence.

The objective of this study is to report the total enucleation of a keratocyst in the posterior region of the mandible as an alternative treatment. This work presents the clinical and radiographic characteristics of the lesion.

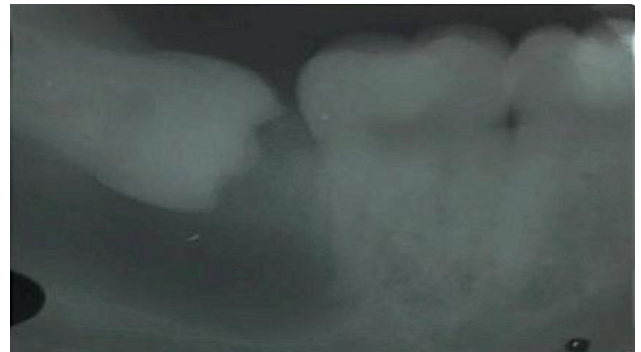
## 2 Case Report

This case report was submitted to the evaluation of the Research Ethics Committee of Adventist College

of Bahia through the Plataforma Brasil (CAEE No. 74702622.0.0000.0042) and was approved by Opinion No. 6.417.867 according to regulated guidelines in research involving human beings (Resolution 466/2012).

Patient M.Y.S.B., 20 years old, female, ASA I, after a routine consultation at a dental clinic, was referred to the Centro de Especialidades Odontológicas - Centro/Cachoeira - BA for extraction of tooth 48. During the anamnesis, the patient did not report any painful symptoms, denied drug allergies, and systemic alterations. Clinically, tooth 48 was partially retained. On the periapical radiographic examination (Figure 1), a radiolucent image was observed around the tooth, followed by requests for panoramic radiographic examinations (Figure 2) and cone-beam computed tomography (Figure 3) for better evaluation, which showed a unilocular radiolucent image extending from the mandibular ramus to the region of tooth 47, with apical displacement of the mandibular canal. The tomographic examination revealed in the axial cuts thinning and rupture of the lingual cortex and slight bulging of the buccal cortex.

**Figure 1** - Periapical radiograph



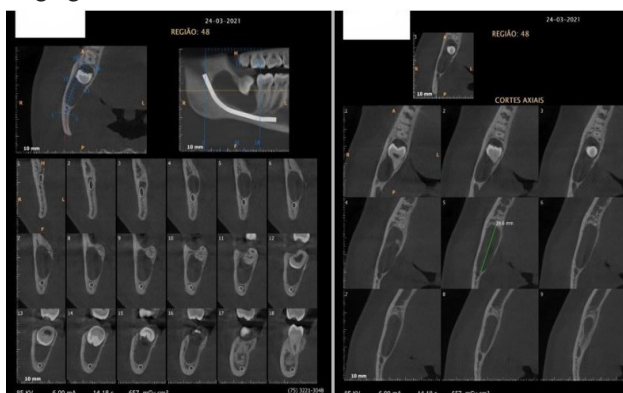
Source: the authors.

**Figure 2** - Initial panoramic radiograph: a unilocular radiolucent image is observed, extending from the mandibular ramus to tooth 47, with apical displacement of the mandibular canal



Source: the authors.

**Figure 3** - Computed tomography: axial sections show thinning and rupture of the lingual cortical bone and slight bulging of the buccal cortical bone



Source: the authors.

An attempt at aspiration was performed with negative results, followed by exploratory surgery, opting for complete enucleation of the lesion (Figure 4) and extraction of the impacted tooth 48 (Figure 5). Samples were sent for histopathological analysis, which revealed a keratocystic odontogenic tumor with focal low-grade epithelial dysplasia and the presence of satellite cysts. Following enucleation, the patient underwent follow-up every six months.

**Figure 4** - Odontogenic Keratocyst



Source: the authors.

**Figure 5** - Extracted tooth unit, element 48



Source: the authors.

## 2.2 Discussion

keratocyst is an odontogenic cyst that generates significant interest among surgeons due to its aggressive biological behavior, as well as specific clinical manifestations. Because of these characteristics, WHO has considered it a true neoplasm, referred to as the odontogenic keratocyst tumor<sup>6</sup>. The expansion process can lead to morbidities such as dental resorption and displacement, cortical bulging, asymmetry, pain, and drainage<sup>1</sup>.

The diagnosis of OKC is considered challenging due to its aggressive and asymptomatic nature and is often identified incidentally through imaging studies, as occurred in this case. Conventional radiographs cannot distinguish OKC from other maxillomandibular lesions, such as dentigerous cyst, ameloblastoma, early stages of calcifying odontogenic cyst, adenomatoid odontogenic tumor, and ameloblastic fibroma<sup>10</sup>.

Panoramic and computed tomography scans may be requested to assist in diagnosis and therapeutic planning. Detection of this condition can be challenging due to its similarity to other cystic and neoplastic lesions. Clinical and radiographic differences between the keratocyst and other conditions can result in incorrect or delayed diagnoses<sup>11</sup>. Radiographic features commonly found are radiolucent images, either unilocular or multilocular, with circular trabeculations and surrounded by radiopaque areas. However, these findings are only suggestive, and biopsy is recommended for a definitive diagnosis<sup>6,12</sup>.

Panoramic radiography has various applications and is used for an initial evaluation of this complex, providing detailed information about structures and potential anatomical anomalies. It helps identify pathological regions and understand their relationship with adjacent tissues<sup>13</sup>.

In addition to panoramic radiography, a computed tomography scan may be requested, allowing the surgeon to visualize the odontogenic keratocyst in all its dimensions. This exam reveals critical structures that might be affected by the aggressive nature of the lesion, such as the inferior alveolar nerve region, the nasal cavity, and the maxillary sinus. Tomography also helps determine the size of the bone cortical perforation, which is crucial for surgical excision. In this regard, computed tomography is more effective than conventional radiography as an auxiliary diagnostic tool<sup>10</sup>.

According to Gonçalves et al.<sup>14</sup>, flat radiographs often do not provide adequate information for surgical planning. Therefore, computed tomography is essential as it overcomes the limitations of 2D image overlap, allowing for a more accurate and effective assessment. Due to its invasive-destructive nature and potential to reach large sizes with high recurrence rates<sup>15</sup>, once diagnosed, treatment should be prompt to avoid damage and deformities to the stomatognathic system<sup>16</sup>.

This condition accounts for approximately 10% of all cystic jaws lesions. Keratocysts are more prevalent between the second and third decades of life, with a higher prevalence

in males, and the posterior region of the mandible is the most commonly affected area<sup>17</sup>. Melo et al.<sup>18</sup> also state that there is a higher incidence in men, involving approximately 60% to 80% of cases, with the greatest frequency in the posterior body and mandibular ramus.

Oliveira et al.<sup>4</sup> also noted in their study that the odontogenic keratocyst occurs in 37.7% of patients over 50 years old and in 28.56% of those under 20 years old. There is a preference for males and for the white ethnicity, representing 67.8% of the cases. Additionally, 40% of the cases are associated with an unerupted tooth.

The treatment of OKC is determined based on its clinical and imaging presentation<sup>6</sup>, ranging from simpler methods such as marsupialization and enucleation to more aggressive approaches like block resection. However, there is still no consensus in the literature regarding the optimal management for this type of lesion<sup>19,20</sup>. For lesions of significant extent, marsupialization or preliminary decompression are employed to facilitate enucleation and manage risks of pathological fracture or to preserve important anatomical structures initially involved by the lesion<sup>21</sup>.

According to Oliveira et al.<sup>4</sup>, treatment remains a controversial issue. The choice of the appropriate method often depends on the individual experience of each professional. Surgical approaches range from conservative methods, such as enucleation, to more aggressive options, like partial or total resections.

Histologically, this lesion is characterized by a cystic capsule of connective tissue with a stratified squamous parakeratinized epithelial lining. The basal layer exhibits a palisaded arrangement of cells with hyperchromatic nuclei, and the connective tissue wall contains “daughter cysts” or transverse sections of the cystic capsule. The cystic content is either liquid (straw-colored) or creamy (thick and grayish), containing keratin, cholesterol crystals, and hyaline bodies, as observed in the reported case<sup>5</sup>. The cells of the odontogenic keratocyst show higher proliferative capacity, related to unknown inherent factors of the epithelium or enzymatic activity in the fibrous capsule<sup>6,22</sup>.

Regarding limitations, various studies in the literature discuss the recurrences associated with different treatment types. Techniques such as enucleation, curettage, and marsupialization are more frequently linked to higher recurrence rates due to residual tissue from the lesion. In contrast, radical resection shows the lowest recurrence rates<sup>4</sup>. The recurrence of the keratocyst is more related to the nature of the lesion than to the surgical technique used. Research indicates that recurrence rates can be significant, ranging from 10% to 60%, depending on the therapeutic approach and the quality of postoperative follow-up. The presence of a thin and fragile capsule, as well as the formation of septa and irregular borders during lesion growth, makes complete removal of the bone cavity through enucleation and curettage more challenging<sup>23</sup>.

Its prognosis is generally good; however, treatment is prolonged and requires extended follow-up to manage recurrences. Recurrences may occur mainly due to the presence of satellite cysts or smaller cysts connected to the main lesion, as well as technical difficulties in the surgical removal of the lesion<sup>6</sup>.

### 3 Conclusion

The clinical characteristics of OKC reaffirm the importance of routine imaging exams. The significant value of CT in visualizing the extent of the lesion and adjacent structures is evident, emphasizing the importance of anatomopathological analysis for definitive diagnosis. It is important to note that the surgical procedure achieved is objective, but ongoing monitoring of this patient is necessary due to its high recurrence potential.

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