

## Large Odontogenic Keratocyst in the Mandible Treated by Surgical Enucleation: Case Report

*Queratocisto Odontogênico de Grande Extensão em Mandíbula Tratado por Enucleação Cirúrgica: Relato de Caso*

*Queratoquiste Odontogênico de Gran Tamaño en la Mandíbula Tratado Mediante Enucleación Quirúrgica: Informe de Caso*

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### Abstract

Odontogenic keratocyst (OK) is a developmental odontogenic cyst characterized by aggressive clinical behavior, high growth potential, and high recurrence rates, which makes its diagnosis and therapeutic management challenging. The aim of this study was to report and discuss the clinical, radiographic, histopathological characteristics and therapeutic approaches of a case of extensive mandibular OK, initially treated and after recurrence, by enucleation. A 20-year-old female patient was diagnosed with an extensive intraosseous lesion in the posterior region of the right mandible, identified incidentally on radiographic examination. The diagnostic hypothesis of OK was confirmed by histopathological examination after surgical enucleation of the lesion associated with the extraction of the impacted tooth 48. After six months, tomographic examinations showed recurrence, requiring further surgical intervention through enucleation of the recurrent lesion, associated with endodontic treatment of the involved teeth 46 and 47. Clinical and radiographic follow-up after the second surgery showed no signs of recurrence and adequate bone neoformation, confirming the success of the established therapeutic approach. It was possible to conclude that the case reinforces the importance of histopathological diagnosis, careful choice of therapeutic approach, and prolonged clinical and radiographic follow-up, considering the high potential for recurrence of odontogenic keratocyst.

**Descriptors:** Odontogenic Cysts; Treatment Outcome; Recurrence; Cone-Beam Computed Tomography.

### Resumo

O queratocisto odontogênico (CO) é um cisto odontogênico de desenvolvimento caracterizado por comportamento clínico agressivo, alto potencial de crescimento e altas taxas de recorrência, o que torna seu diagnóstico e tratamento desafiadores. O objetivo deste estudo foi relatar e discutir as características clínicas, radiográficas e histopatológicas, bem como as abordagens terapêuticas, de um caso de CO mandibular extenso, tratado inicialmente e, após recorrência, por enucleação. Uma paciente de 20 anos foi diagnosticada com uma extensa lesão intraóssea na região posterior da mandíbula direita, identificada incidentalmente em exame radiográfico. A hipótese diagnóstica de queratocisto odontogênico (CO) foi confirmada pelo exame histopatológico após enucleação cirúrgica da lesão associada à extração do dente impactado 48. Após seis meses, exames tomográficos mostraram recorrência, necessitando de nova intervenção cirúrgica por meio de enucleação da lesão recidivada, associada ao tratamento endodôntico dos dentes envolvidos 46 e 47. O acompanhamento clínico e radiográfico após a segunda cirurgia não apresentou sinais de recidiva e demonstrou neoformação óssea adequada, confirmando o sucesso da abordagem terapêutica estabelecida. Foi possível concluir que o caso reforça a importância do diagnóstico histopatológico, da escolha criteriosa da abordagem terapêutica e do acompanhamento clínico e radiográfico prolongado, considerando o alto potencial de recidiva do queratocisto odontogênico.

**Descritores:** Cisto Odontogênico; Resultado do Tratamento; Recidiva; Tomografia Computadorizada de Feixe Cônico.

### Resumen

El queratoquiste odontogênico (QO) es un quiste odontogênico del desarrollo caracterizado por un comportamiento clínico agresivo, un alto potencial de crecimiento y altas tasas de recurrencia, lo que dificulta su diagnóstico y manejo terapéutico. El objetivo de este estudio fue informar y discutir las características clínicas, radiográficas e histopatológicas, así como los enfoques terapéuticos de un caso de QO mandibular extenso, tratado inicialmente y, tras una recurrencia, mediante enucleación. Una paciente de 20 años fue diagnosticada con una lesión intraóssea extensa en la región posterior de la mandíbula derecha, identificada incidentalmente en un examen radiográfico. La hipótesis diagnóstica de queratoquiste odontogênico (QO) se confirmó mediante examen histopatológico tras la enucleación quirúrgica de la lesión asociada a la extracción del diente impactado 48. Seis meses después, los exámenes tomográficos mostraron recidiva, lo que requirió una nueva intervención quirúrgica mediante la enucleación de la lesión recurrente, junto con el tratamiento endodôntico de los dientes afectados 46 y 47. El seguimiento clínico y radiográfico tras la segunda cirugía no mostró signos de recidiva y se observó una adecuada neoformación ósea, lo que confirmó el éxito del enfoque terapéutico establecido. Se pudo concluir que este caso refuerza la importancia del diagnóstico histopatológico, la cuidadosa selección del enfoque terapéutico y el seguimiento clínico y radiográfico prolongado, dada la alta probabilidad de recidiva del queratoquiste odontogênico.

**Descriptores:** Quistes Odontogénicos; Resultado del Tratamiento; Recurrencia; Tomografía Computarizada de Haz Cônico.

## INTRODUCTION

Odontogenic keratocyst (OK) was initially described as an odontogenic tumor or benign intraosseous neoplasm by the 2005 World Health Organization (WHO) Classification due to its aggressive and infiltrative clinical behavior. In 2017, in the fourth edition of the WHO, it was reclassified as a developmental odontogenic cyst, called "odontogenic keratocyst," and no longer as a neoplasm<sup>8,10,11,15</sup>. This cyst accounts for approximately 11% of maxillary cysts and is therefore relatively rare. It is predominantly located in the mandible, most frequently in the mandibular ramus and molar region, with a high potential for expansion and rapid growth. The lesion mainly affects individuals in their second and third decades of life, with a higher prevalence in males<sup>12,22</sup>. High recurrence rates are the main characteristic of this odontogenic cyst, observed in approximately 25% to 60% of cases, in addition to the presence of satellite cysts, which makes complete removal difficult. Its origin is uncertain and may be related to the dental lamina and its remnants, or even to the enamel organ<sup>1,8,15,22</sup>.

Clinically, the lesion is generally asymptomatic, with slow and expansive growth, occurring predominantly in the anteroposterior direction. Only in more advanced stages is progression observed in the lateral-lateral direction, in a non-destructive manner. It is mainly located in the mandible, as previously mentioned, keeping the skin and oral mucosa intact, with a normal appearance, and can be identified by palpation. Only in advanced stages does the increase in volume become evident, with compromise of the bone plate, occasionally causing swelling, pain, abnormal sensations, purulent drainage, and even tooth mobility<sup>3,5,22</sup>. In cases where patients have multiple keratocysts in the jaws, associated with other characteristics such as mild mandibular prognathism, ocular hypertelorism, multiple basal cell carcinomas, and bifid ribs, the condition is consistent with Gorlin-Goltz syndrome. This is a condition that requires a more complex and multidisciplinary therapeutic approach<sup>11,22</sup>.

Radiographically, the lesion may appear as a unilocular or multilocular image, especially when extensive, with an oval or rounded shape. In up to 40% of cases, it is associated with the crown of unerupted teeth<sup>1,5,8,12,22</sup>. Often, OK manifests radiographically as a radiolucent image located in the mandibular ramus, with thin, well-defined edges, which may vary in terms of the presence of a sclerotic halo. In certain cases, the differential diagnosis includes ameloblastoma, dentigerous cyst, adenomatoid odontogenic tumor, calcifying odontogenic cyst, and ameloblastic fibroma. When the lesion is not associated with the crown of a tooth, central giant cell granuloma, traumatic bone

cyst, and lateral periodontal cyst should also be considered as diagnostic hypotheses<sup>2,4,6,13,21,22</sup>. The diagnosis should not be based exclusively on clinical or imaging findings; therefore, histopathological examination is essential for establishing an accurate diagnosis<sup>1</sup>.

Histopathologically, OK is characterized by a cystic wall lined with stratified squamous orthokeratinized or parakeratinized epithelium, with a thickness varying between 6 and 8 cell layers, presenting a basal layer composed of cells arranged in a palisade and dense fibrous connective tissue underneath. The presence of satellite cysts, as well as the flat interface between the lesion epithelium and the connective tissue of the capsule, hinders curettage, making the lesion more friable and increasing susceptibility to recurrence<sup>1,5,11,12,22</sup>. The cystic content may be liquid, straw-colored, or creamy, grayish, consisting of keratin, cholesterol crystals, and hyaline bodies<sup>1</sup>. After confirmation of the diagnosis by histopathological examination, a preventive aspiration is performed before entering the cavity, with the content usually positive and citrus yellow in color, and then the therapeutic option is determined<sup>9</sup>.

The treatment of OK is controversial, involving both conservative and more aggressive approaches. Conservative therapies include enucleation of the lesion, which may be preceded by decompression or marsupialization in cases of extensive lesions, followed by curettage, with the aim of preserving noble structures and reducing the morbidity of adjacent tissues. However, due to the presence of satellite cysts and the easy detachment of the epithelium and fibrous capsule, these approaches are associated with higher recurrence rates. Thus, more aggressive treatments may be indicated to reduce recurrence, including peripheral ostectomy, cryotherapy, electrocautery, resection, and chemical curettage with Carnoy's solution as an adjunct to enucleation. Despite the effectiveness of these techniques in preventing recurrence, they can cause damage to important structures such as teeth, blood vessels, and nerves<sup>1,3,7,12,16,17,22</sup>. Therefore, the following study aims to report, discuss, and analyze the histopathological, clinical, and radiographic characteristics and therapeutic approaches of an extensive mandibular OK, in which enucleation was performed as an initial proposal and after recurrence.

This work is characterized as a clinical case report, qualitative and descriptive in nature, which gathers and presents relevant information from the literature on the clinical, radiographic, and histopathological characteristics of OK, contributing to a broader understanding and appropriate therapeutic management. It should be noted that all ethical and legal aspects were duly observed, with

the patient signing the Free and Informed Consent Form (FICF) provided by the Araçatuba School of Dentistry – FOA/UNESP, authorizing the diagnosis, treatment, and use of images for scientific purposes and publication in specialized journals.

### CASE REPORT

A 20-year-old female patient with leukoderma attended the Oral and Maxillofacial Surgery and Traumatology Clinic at the Araçatuba School of Dentistry (FOA/UNESP). During a previous radiographic evaluation for orthodontic purposes, an intraosseous lesion was identified in the posterior region of the right mandible, which is why the patient was referred to a specialized service.

In the anamnesis, the patient denied systemic comorbidities, allergies, continuous use of medications, and addictions. Intraoral and extraoral clinical examination revealed no noteworthy changes, and the patient had no complaints of pain or paresthesia in the affected region.

For better diagnostic clarification, a panoramic radiograph was requested, which revealed an extensive intraosseous lesion affecting the body and mandibular ramus on the right side, with a well-defined, multilocular radiolucent image associated with the root apices of teeth 46 and 47 and tooth 48 embedded within the lesion. After careful analysis of the imaging exam and clinical-radiographic correlation, a diagnosis of odontogenic keratocyst was established, and surgical treatment was proposed through cystic enucleation, with the material sent for histopathological analysis.

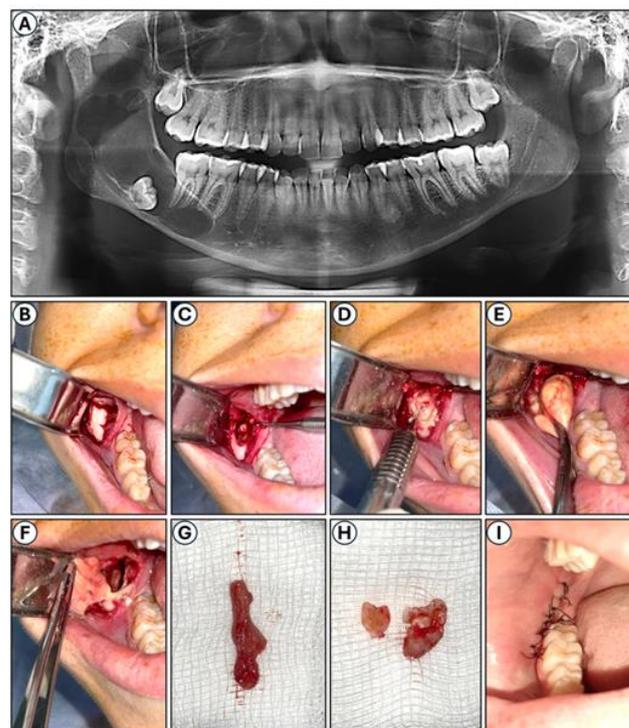
The procedure was performed in an outpatient setting, beginning with extraoral asepsis with 10% polyvinylpyrrolidone-iodine (PVPi) and intraoral asepsis with 0.12% chlorhexidine, followed by isolation of the surgical field. Anesthesia was performed by blocking the inferior alveolar, lingual, and buccal nerves and complementing with terminal infiltrative anesthesia in the mandibular vestibule, using a 2% mepivacaine hydrochloride solution with 1:100,000 adrenaline.

Next, a straight incision was made along the external mandibular oblique line with a No. 15 scalpel blade, followed by mucoperiosteal detachment and exposure of the bone tissue (Figure 1B). With the aid of a No. 10 spherical carbide drill, access to the cystic cavity was achieved (Figure 1C), and with specific curettes, enucleation of the lesion was performed (Figure 1D, Figure 1E, Figure 1F), revealing a thin cystic capsule (Figure 1G) and whitish pasty contents inside (Figure 1D), consistent with the presence of keratin. Concomitant with enucleation, tooth 48 involved in the lesion was removed with the aid of extractors after tooth sectioning (Figure 1H).

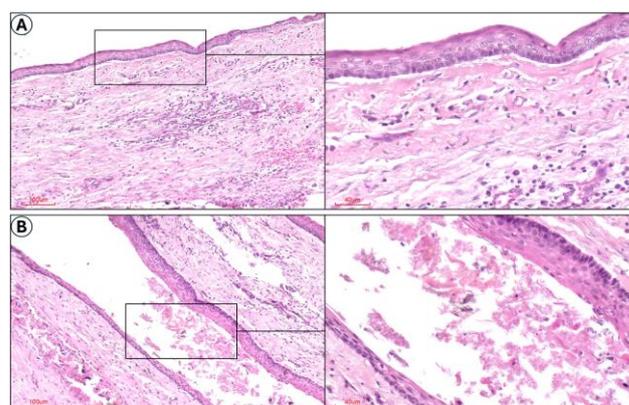
The removed material was fixed in 10% buffered formaldehyde and sent for

histopathological analysis, which confirmed the diagnosis of odontogenic keratocyst (Figure 2A and Figure 2B). The surgical cavity was irrigated with 0.9% saline solution, and the flap was repositioned and sutured with interrupted simple stitches using 5-0 nylon thread (Figure 1I).

At the end of the surgical procedure, amoxicillin 500 mg was prescribed every 8 hours for 7 days; nimesulide 100 mg every 12 hours for 3 days; and dipyron 500 mg every 6 hours, if necessary for pain.



**Figure 1:** Preoperative imaging examination and cystic enucleation procedure. **A:** Panoramic radiograph; **B:** Retraction of the flap and exposure of bone tissue; **C:** Osteotomy and access to cystic cavity; **D, E, and F:** Curettage and cystic enucleation; **G:** Thin cystic capsule removed; **H:** Tooth 48 removed from inside the lesion; **I:** Repositioning of flap with 5-0 nylon suture (Source: authors).



**Figure 2:** Histopathological analysis of odontogenic keratocyst. **A** and **B:** photomicrographs of different regions at 100µm and 40µm magnification (Source: authors).

During postoperative outpatient follow-up, the patient underwent pulp vitality tests on teeth 46 and 47, revealing loss of pulp vitality in tooth 47, which indicated the need for endodontic treatment.

After six months of follow-up, a cone beam computed tomography scan of the mandible was requested (Figure 3A and Figure 3B), which showed recurrence of the lesion in the distal region of tooth 47 and near the periapical region of teeth 46 and 47.

Given the recurrence, a new surgical procedure for cystic enucleation was necessary. Asepsis and anesthesia were performed similarly to the procedure previously described. A low monoangular incision was made with a No. 15 scalpel blade, followed by mucoperiosteal detachment and exposure of the bone tissue (Figure 3C and Figure 3D). Subsequently, two osteotomies were performed with a No. 10 spherical carbide drill, the first in the periapical region of teeth 46 and 47 (Figure 3E) and the second in the distal region of tooth 47 (Figure 3F), allowing access and curettage of the recurrent lesion (Figure 3G and Figure 3H).

After removal of the lesion (Figure 3I), the surgical cavity was irrigated with 0.9% saline solution, and the flap was repositioned and sutured with interrupted simple stitches using 5-0 nylon thread (Figure 3J). The patient was prescribed antibiotics, anti-inflammatory drugs, and analgesics, according to the protocol used in the first surgical approach. Postoperative instructions were provided, and endodontic treatment of tooth 46 was indicated.

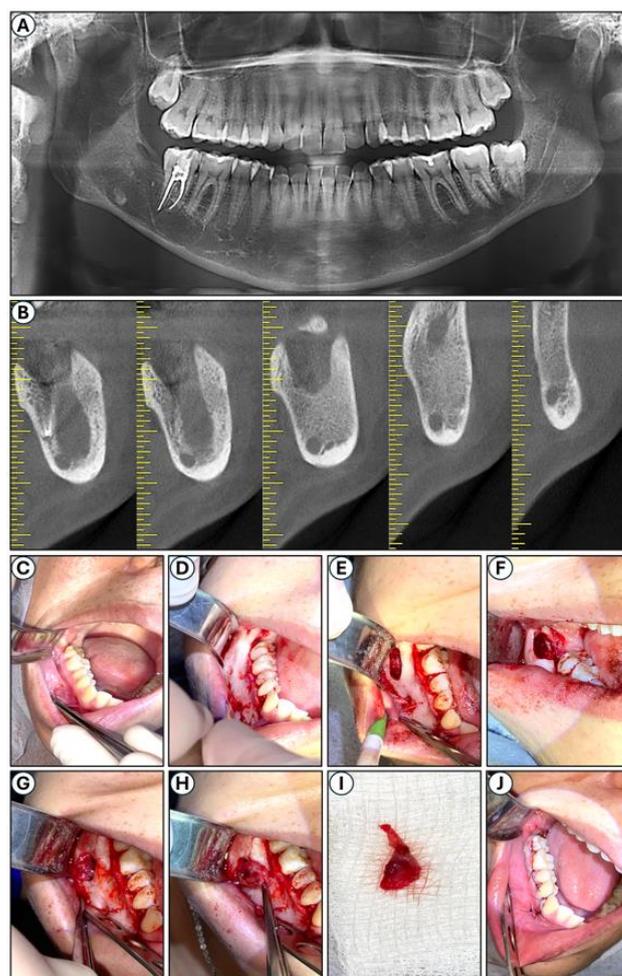
Six months after the second surgical procedure, a new cone beam computed tomography scan of the mandible was requested for follow-up (Figure 4A, Figure 4B, and Figure 4C). The cone beam computed tomography scan showed no signs of recurrence, as well as satisfactory bone neof ormation in the area previously occupied by the lesion. From a clinical standpoint, the patient was asymptomatic, with no complaints of pain or paresthesia, maintaining preserved function and daily activities. Currently, the patient remains under outpatient follow-up, with no evidence of recurrence of the lesion.

## DISCUSSION

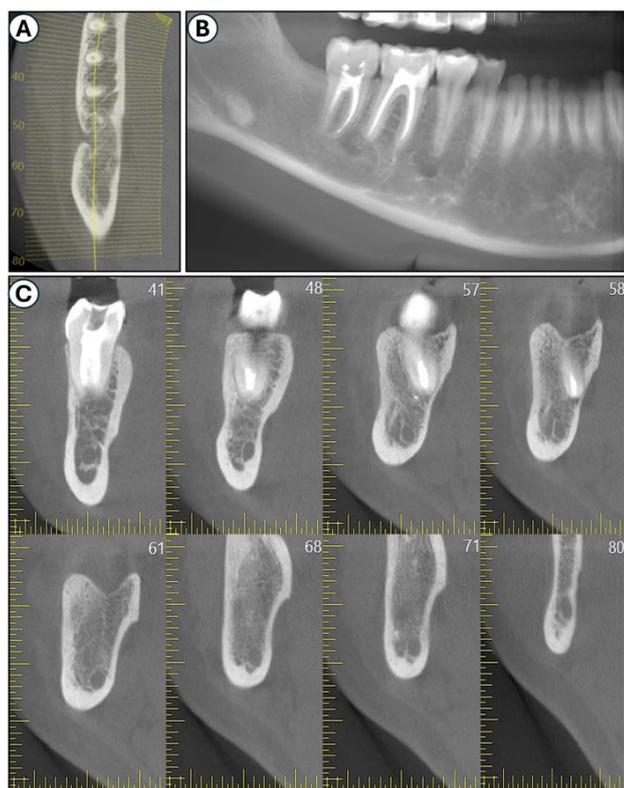
characteristic means that it often goes undiagnosed in its early stages, and its growth can lead to bone fragility, making the individual more susceptible to future pathological fractures. The diagnosis is based on the correlation between clinical, imaging, and histopathological findings, the latter being fundamental for defining the therapeutic approach.

The most affected age group is between the 2nd and 3rd decades of life<sup>19</sup>. Most cases of OK are in the posterior region of the mandible<sup>9</sup>, accounting for about 60% of cases occurring in the posterior mandible, but it can also recur in the maxilla (13%) and, even more rarely, in the maxillary sinus (1%)<sup>21</sup>. In the clinical case in

question, the patient was included in the group with the highest predilection, presenting typical characteristics of the lesion, both in the absence of symptoms and in the radiographic aspect, which was confirmed histopathologically. Initially, the patient underwent complete enucleation of the lesion, associated with extraction of the third molar involved, an approach considered conservative and indicated in well-defined lesions without signs of invasion of adjacent structures, especially considering the patient's age. Although OK is more aggressive than other benign bone lesions, it is a commonly asymptomatic cyst<sup>8</sup>. This characteristic means that it often goes undiagnosed in its early stages, and its growth can lead to bone fragility, making the individual more susceptible to future pathological fractures. The diagnosis is based on the correlation between clinical, imaging, and histopathological findings, the latter being fundamental for defining the therapeutic approach.



**Figure 3:** Imaging exam for follow-up after six months and surgical procedure for enucleation of recurrent lesion. **A:** Cone beam computed tomography of the mandible in panoramic reconstruction; **B:** Cone beam computed tomography of the mandible in coronal sections; **C:** Initial clinical appearance; **D:** Flap reflection and exposure of bone tissue; **E:** Osteotomy performed near the periapical region of teeth 46 and 47; **F:** Osteotomy performed distally to tooth 47; **G** and **H:** Curettage and enucleation of the recurrent lesion; **I:** Recurrent lesion removed; **J:** Repositioning of the flap with 5-0 nylon suture (Source: authors).



**Figure 4:** Cone beam computed tomography of the mandible six months after enucleation of the recurrent cystic lesion. **A:** Axial plane with markings of the coronal sections performed; **B:** Sagittal plane; **C:** Coronal plane (Source: authors).

The most affected age group is between the 2nd and 3rd decades of life<sup>12</sup>. Most cases of OK are in the posterior region of the mandible<sup>9</sup>, accounting for about 60% of cases occurring in the posterior mandible, but it can also recur in the maxilla (13%) and, even more rarely, in the maxillary sinus (1%)<sup>21</sup>. In the clinical case in question, the patient was included in the group with the highest predilection, presenting typical characteristics of the lesion, both in the absence of symptoms and in the radiographic aspect, which was confirmed histopathologically. Initially, the patient underwent complete enucleation of the lesion, associated with extraction of the third molar involved, an approach considered conservative and indicated in well-defined lesions without signs of invasion of adjacent structures, especially considering the patient's age.

Although the surgical approach has proven to be highly effective, OK is known for its high recurrence rate, which is higher than that of other odontogenic cysts, often due to epithelial remnants from the lesion<sup>15</sup>. Such biological aggressiveness was observed in the case in question, where there was recurrence of the lesion after the first surgical procedure performed. Other techniques such as decompression and marsupialization can be used in combination with surgical removal of the cystic content depending on the size of the lesion. However, in the present case, a second enucleation associated with endodontic

treatment of teeth 46 and 47 was chosen. The literature also reports therapeutic options such as peripheral osteotomy, Carnoy's solution, and scarification, which are sometimes more invasive and difficult to perform, a fact that corroborates the choice of more conservative treatment measures, such as surgical enucleation<sup>14,18,23</sup>.

The clinical evolution after the second surgical approach was favorable, with complete remission of the lesion and no signs of postoperative complications. In addition, clinical and radiographic follow-up over one year showed no signs of recurrence, suggesting that the new therapeutic approach was necessary and effective. The absence of recurrence to date reinforces the importance of careful enucleation, associated with the correct removal of the lesion, especially when combined with complementary therapies, such as endodontics, to remove possible sources of irritation or contamination. However, the patient continues to undergo periodic clinical and radiographic follow-up, as the most recent literature shows recurrence even at a late stage<sup>20</sup>.

## CONCLUSION

The correlation between clinical, radiographic, and histopathological findings was essential for the diagnosis of OK in the presented case. The therapeutic approach adopted, based on surgical enucleation of the lesion, proved effective, although a second intervention was necessary due to recurrence. After the surgical reapproach, favorable clinical and radiographic evolution was observed, with adequate bone neof ormation and no signs of recurrence during the follow-up period, reinforcing the importance of careful management and long-term follow-up in these cases.

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#### CONFLICT OF INTERESTS

The authors declare no conflict of interest.

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