

## RELATO DE CASO

## ODONTOGENIC KERATOCYST OF THE MAXILLA: CASE REPORT

## CERATOCISTO ODONTOGÊNICO DA MAXILA: RELATO DE CASO

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ACESSO LIVRE

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## RESUMO

**Introduction:** Odontogenic Keratocyst (OKC) is a pathology that presents high proliferative activity and frequent relapses after treatment. It is a benign but locally aggressive cyst. **Development:** The aim of this study was to report a case of OKC with uncommon location in a 22-year-old patient presenting swelling and drainage of purulent secretion in the region of the third right upper molar. In examinations, a radiolucent density unilocular image involving impacted element 18 was observed. After excisional biopsy accompanied by severe bone curettage, the removed specimen was sent for histopathological analysis, where final diagnosis of odontogenic keratocystic was obtained. The patient has been in clinical and radiographic follow-up for more than 6 months and, to date, there are no signs of relapse. **Final considerations:** This study present to us the relevance of scientific studies in the identification of oral pathologies, always seeking the best treatment and prognosis.

**Keywords:** Odontogenic Keratocyst; Odontogenic Cysts; Nevoid Basal-Cell Carcinoma

## ABSTRACT

**Introduction:** Odontogenic Keratocyst (OKC) is a pathology that presents high proliferative activity and frequent relapses after treatment. It is a benign but locally aggressive cyst. **Development:** The aim of this study was to report a case of OKC with uncommon location in a 22-year-old patient presenting swelling and drainage of purulent secretion in the region of the third right upper molar. In examinations, a radiolucent density unilocular image involving impacted element 18 was observed. After excisional biopsy accompanied by severe bone curettage, the removed specimen was sent for histopathological analysis, where final diagnosis of odontogenic keratocystic was obtained. The patient has been in clinical and radiographic follow-up for more than 6 months and, to date, there are no signs of relapse. **Final considerations:** This study present to us the relevance of scientific studies in the identification of oral pathologies, always seeking the best treatment and prognosis.

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## INTRODUCTION

Odontogenic Keratocyst (OKC) was first described by Philipsen<sup>1</sup> in 1956 and has ever since been well documented and extensively studied, especially for presenting high relapse rate and aggressive biological behavior. The OKC has undergone conceptual and terminological changes in recent decades. There was much discussion about the need for distinguishing the two subtypes of OKC from one another since the parakeratinized one presented a higher recurrence rate. This evidence resulted in the separation of these subtypes into two distinct diseases and the abandonment of the term "OKC" by the WHO in 2005<sup>2</sup>. The parakeratinized subtype became the "keratocystic odontogenic tumor" (KCOT), which integrated the group of odontogenic epithelial tumors into its classification, while the orthokeratinized subtype continued in the group of odontogenic developmental cysts as "orthokeratinized odontogenic cyst". Recently in 2017, the KCOT returned to the WHO classification of odontogenic developmental cysts, retaking the original terminology "OKC"<sup>3</sup>.

Compared to the other odontogenic cysts, this lesion has a high prevalence, generally occupying second place, only behind root cyst. It appears mainly in the second and third decades of life, with a slight predilection for the male gender. They are solitary lesions in most cases, but may also be associated with Nevoid Basal-Cell Carcinoma Syndrome and mainly affect the mandible, especially the posterior part of the ascending body and branch<sup>4</sup>.

Radiographically, it presents as a well-defined unilocular or multilocular radiolucent lesion with corticated margins. In a considerable part of the cases, there is an impacted tooth involved in the injury<sup>4,5</sup>.

On histopathological examination, OKC present stratified squamous epithelium with about 8 to 10 basal layer cells, being cubic or cylindrical, arranged in palisade. Generally, the epithelial surface presents parakeratine of corrugated aspect. However, it may present, in some cases, orthokeratinization, which, for some authors, characterizes a variant of this pathology. The capsule, consisting of fibrous connective tissue, is thin and friable, generally free of inflammation, occasionally exhibiting microcysts. The epithelial-connective interface is flat. Small satellite cysts, cords or islands of odontogenic epithelium can be observed inside the capsule. The lesion lumen may contain a clear liquid that is similar to plasma transudate or may be filled with a caseous substance consisting of keratin remains<sup>6,7</sup>.

Several treatments have been described for OKCs such as decompression, enucleation and marsupialization, with some modifications in techniques, such as the use of Carnoy's solution in order to minimize the possibility of relapse. Treatment protocol based on decompression offers an effective and conservative option, with low morbidity and relapse speed compared to those reported in literature. Long and systematic post-surgical follow-up is considered a key element for a successful outcome. Therefore, the aim of this study was to report the case of OKC in an unusual region, discuss its histopathology and surgical treatment<sup>4,8</sup>.

## CASE REPORT

A 22-year-old male melanoderma patient sought the Oral and Maxillofacial Surgery service at a Reference Surgery Hospital in João Pessoa, Paraíba, Brazil, complaining of drainage of foul-smelling purulent secretion in the region of the third right upper molar, without painful symptomatology.

Clinically, swelling of the posterior region of the right maxilla was observed, with increased soft tissue, measuring approximately 4x4cm. A panoramic radiograph was requested and a 12 / 12h chlorhexidine mouthwash was prescribed due to the diagnostic hypothesis of pericoronaritis. Panoramic radiographic examination revealed a unilocular radiolucent image with defined and sclerotic borders measuring approximately 1.5 cm in its greatest diameter, around the third right molar element (Figure 1).



Figure 1: Initial panoramic radiography

Excisional biopsy of the lesion was performed, accompanied by severe bone curettage. The specimen consisted of five soft tissue fragments, with its largest fragment measuring 3.0 x 2.0 x 1.0 cm and a cavity of white-gray coloration and fibroelastic consistency, containing the element 18, which presented integrity (Figure 2).



Figure 2: Intraoperative Procedure

Microscopically, fragments of cystic capsule covered by little thick and uniform stratified squamous epithelium were observed. Basal layer cells were cubic, hyperchromatic and

arranged in palisade. The luminal surface was covered by corrugated-looking parakeratinized epithelial cells. In the dense fibrous connective tissue capsule, intense infiltration of chronic inflammatory cells and hemorrhage areas were observed (Figure 3). The histopathological diagnosis was OKC.

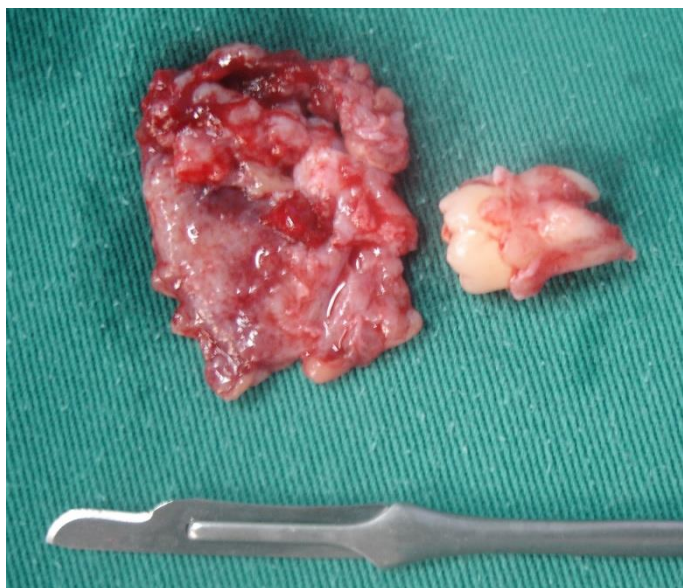


Figure 3: Specimen and element 18 referred for histopathological examination.

The patient has been in clinical and radiographic follow-up for more than 6 months and to date, there are no signs of relapse.

## DISCUSSION

Odontogenic keratocyst is unique among odontogenic cysts, especially due to its high risk of relapse.

Epidemiological studies have shown a predilection of the lesion for males, and for the 2<sup>nd</sup> and 3<sup>rd</sup> decades of life<sup>8,9</sup>, which findings corroborate those of the present case. However, most lesions affect the mandibular branch and body<sup>4,10</sup>, differently from the present case, which affected the posterior region of the maxilla, a rare location for this type of lesion. In an epidemiological study by Servato<sup>11</sup> carried out during the years 1978-2009, it was found that the prevalence of OKC represented 31% of all diagnosed odontogenic cysts.

The main features of the Nevoid Basal-Cell Carcinoma Syndrome, or Gorlin-Goltz syndrome, are the presence of multiple odontogenic keratocysts in the maxillo-mandibular complex, basal cell carcinomas and skin epidermoid cysts. It is a hereditary disease and is believed to be caused by mutation of a tumor suppressor gene. In the present case, the patient presented only a tumoral lesion in the maxilla and there were no other skin characteristics common to the syndrome<sup>10,12</sup>.

Radiographically, OKCs present as a well-defined unilocular lesion, usually with corticated margins. In a study carried out by Fadi, it was observed that 52% of the radiographic OKC images have an impacted dental element and there is usually no resorption of the adjacent tooth roots<sup>5</sup>. The radiographic features of OKC in literature were compatible with the case presented here.

The histopathological features found in the present study, such as presence of a cystic capsule of fibrous connective tissue, lining epithelium presenting a corrugated surface and basal cell layer arranged in palisade, are in agreement with literature. The presence of intense inflammatory infiltrate and hemorrhage areas are unusual histopathological processes<sup>4,6,10</sup>.

Due to its clinical behavior, OKC treatment may be notably important. Therapy is closely related to the size of lesions; in smaller ones, enucleation with peripheral ostectomy is chosen, which was performed in the above case, while in larger lesions, marginal or segmental resection is performed due to their great relapsing potential. Other alternatives such as surgical site treatment with Carnoy's solution, electrocauterization, cryotherapy, decompression and / or marsupialization are also reported in literature<sup>13-16</sup>. The fact that the patient, in this case, resides in a rural area, has caused the chosen approach to be adequate to his displacement conditions, and only one surgical moment was chosen. Enucleation treatment associated with curettage was positive. The patient is undergoing preservation, and so far, at 6 months, there are no signs of relapse.

## CONCLUSION

OKCs are lesions of epithelial development and most of the lesions affect the branch and mandibular body. The clinical examination and its radiographic characteristics help the surgeon to obtain an accurate diagnosis and appropriate treatment. Conservative surgical treatment may be an option to reduce recurrence and morbidity and increase the likelihood of secondary cure without complications.

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