SURGICAL MANAGEMENT AND FOLLOW-UP OF ODONTOGENIC KERATOCYSTIC IN MARILARY SINUS WITH PROXIMITY TO ORBITAL CAVITY: CASE REPORT

TRATAMENTO CIRÚRGICO E ACOMPANHAMENTO DE QUERATOCÍSTICO ODONTOGÊNICO EM SEIO MARILAR COM PROXIMIDADE DA CAVIDADE ORBITAL: RELATO DE CASO

AGNALDO ROCHA **PRATA-JÚNIOR**[1*], DEYVID DE CAMARGO **SANTANA**[1], GABRIELLA DANTAS DE **ALMEIDA**[2], ISABELA GOMES DE SENA **RIBEIRO**[1], LAURA BEATRIZ PIMENTA **ALVES**[3], LILIANE CECÍLIA DA **SILVA**[1], PÉROLA DE CASTRO **BARRETO**[4], ÍTALO CORDEITO DE **TOLEDO**[5]

1. Dentist, Resident of Oral and Maxillofacial Surgery, State Emergency Hospital of the Northwest Region of Goiânia Governador Otávio Lage de Siqueira (HUGOL), Goiania, Goias, Brazil. **2.** Dentistry Student, Universidade Tiradentes, Aracaju, Sergipe, Brazil. **3.** Dentistry Student, Centro Universitário Goyazes, Trindade, Goiás, Brazil. **4.** Dentist, Universidade Paulista, Goiânia, Goiás, Brazil **5.** Dentist, Professor and Oral and Maxillofacial Surgeon, State Emergency Hospital of the Northwest Region of Goiânia Governador Otávio Lage de Siqueira (HUGOL), Goiânia, Goias, Brazil.

* Anhanguera Avenue, 14527, Santos Dumont St., Goiânia, Goias, Brazil. ZIP CODE: 74463-350. prata.aiesec@gmail.com

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ABSTRACT

Odontogenic Keratocystic (OKC) is considered one of the most aggressive odontogenic lesions derived from the dental lamina, presenting high recurrence rate. It is equivalent to 11% of all odontogenic cysts, and only 1% of them affect the maxillary sinus. Clinically, in larger lesions, they may present with pain, swelling and drainage, where their diagnosis is confirmed by imaging together with histopathology. Treatments range from the more conservative, such as marsupialization, to the more aggressive, such enucleation and bloc resection. Radical treatments are commonly associated with fewer recurrences, as they allow greater exploration of the lesion, often with safety margins. The purpose of this article was to report the case of a 17-year-old female with OKC in maxillary sinus with proximity to orbital cavity associated to ectopic third molar treated successfully with enucleation. After 12 months of follow-up, patient remained with no recurrence.

KEYWORDS: Maxillary sinus; Odontogenic cysts; Jaw cysts.

1. INTRODUCTION

The odontogenic keratocystic (OKC), now classified as a developmental cyst of odontogenic origin, was first described in 1956 by Philipsen[1]. Later, World Health Organization (WHO) classified odontogenic keratocyst as an odontogenic tumor - between 2005 and 2017 - due to its aggressive behavior. However, considering that tumors do not regress spontaneously (and there are reports of OKC that completely regressed after decompression) the WHO reclassified it as a cyst[2]. Of all odontogenic cysts, only 11% are diagnosed as OKC. It occurs in a wide age group, with peaks of incidence in the second and third decades of life, however 60% of cases are seen in people between 10 and 40 years old[3].

OKC primarily involves the mandibular ramus and angle region. It's rare when presented in maxilla, as this region is affected in only 13% of the cases and the most extensive lesions compromise the maxillary sinus (only 1% of maxilla cases)[3]. Radiographically, OKC appears as a well-defined unilocular or multilocular lesion with scalloped borders, these characteristics make difficult to differentiate from other cyst and tumors of jaws.[4] OKC, dentigerous cyst and ameloblastoma are considered in differential diagnosis.

Histologically, it is a cyst characterized by a thin layer of parakeratinized stratified squamous epithelium with hyperchromatic basic cells in palisade.

OKC are commonly asymptomatic, being discovered only during routine imaging. The larger ones may cause pain, swelling or drainage when infected. The lesion usually has a fibrous, thin, and friable capsule, making it difficult to remove the entire epithelial lining.[5] Because of this, OKC is one of the most aggressive lesions of odontogenic origin, with a high recurrence rate and tendency to invade adjacent tissues[4].

The treatment modalities vary from aggressive management - peripheral ostectomy or enucleation - to more conservative options like marsupialization and decompression followed by secondary enucleation.[6]

The purpose of the present case report is to describe the management of OKC in maxillary sinus with proximity to orbital cavity in a 17-years-old female with enucleation and its outcome over a 1-year period.

2. CASE REPORT

A 17-year-old female reported to the Maxillofacial Department of an Emergency Hospital, with the chief complaint of sinusitis pain for the last 06 months. The pain was intermittent, dull aching, of moderate intensity and was radiating to the ear and eye. She

denied any trauma, swelling or pus discharge. Medical history was non-significant with no history of any systemic illness or long-term medication.

On examination, there were no pathological changes. Intraorally, her four third molars were retained and on palpation of the right superior vestibule she felt pain. Oral mucosa was normal in color and texture.

A computed tomography scan of the face (figure1) showed a single extensive destructive lesion associated to ectopic third-molar crown in the right maxillary sinus with defined borders suggestive of a cystic lesion, extending from the first and second right superior molars root's very tip to the ipsilateral floor of the orbit (02mm from the lesion capsule to orbital cavity). Its measurements were 35mm x 20mm in its largest dimensions.

Figure 1A. Axial Section of CT scan showing resorption of posterior wall of the right sinus. 1B - Axial Section showing ectopic teeth. 1C - Sagital Section showing proximity between lesion capsule and orbital cavity.

An aspiration puncture was initially performed, with the output of cheesy material, leading to the diagnostic hypothesis of odontogenic keratocyst. Therefore, enucleation along with removal of 18 under general anesthesia was performed. Through Caldwell-Luc approach the maxillary sinus was accessed, followed by the lesion enucleation – its capsule was rigid as lesion seemed to be infected – and third molar extraction. For closure of bone window, a polypropylene mesh was sutured onto the sinus wall. Cystic material and its content were sent for histopathological analysis.

Microscopic examination revealed a virtual cystic cavity lined by parakeratinized stratified squamous epithelium with superficial corrugation, presenting hyperchromatic and palisaded basal layer cells. In some regions, hyperplasia and disorganization of the epithelial lining were observed. There were also areas of disjunction between the epithelial lining and connective tissue. Peripherally, fibrous connective tissue with a capsular aspect is evident, showing areas with intense inflammatory infiltrate, predominantly mononuclear, foam cells and bone trabeculae. Therefore, together, the findings were suggestive of Odontogenic Keratocyst.

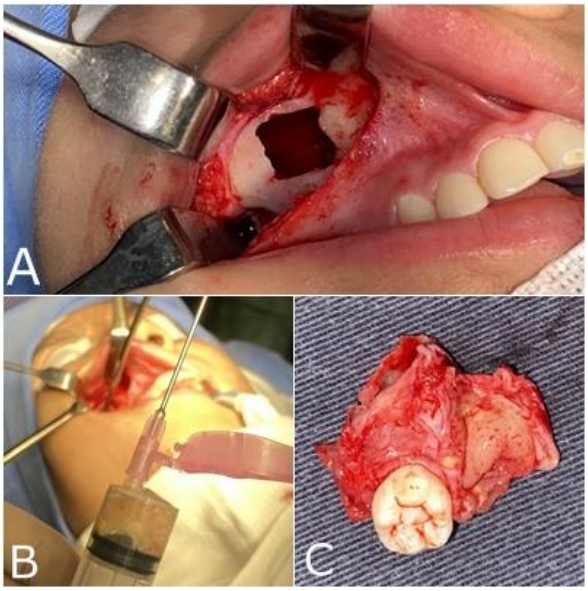

Figure 2A. Caldwell-Luc Access. 2B - Cheesy Material from lesion. 2C - Lesion and ectopic teeth.

It was not reported any postoperative complication. The patient has now been followed up for one year, with no recurrence of the lesion so far. On follow-up images, it was possible to identify bone neoformation in all the sinus walls, as well as complete removal of the lesion.

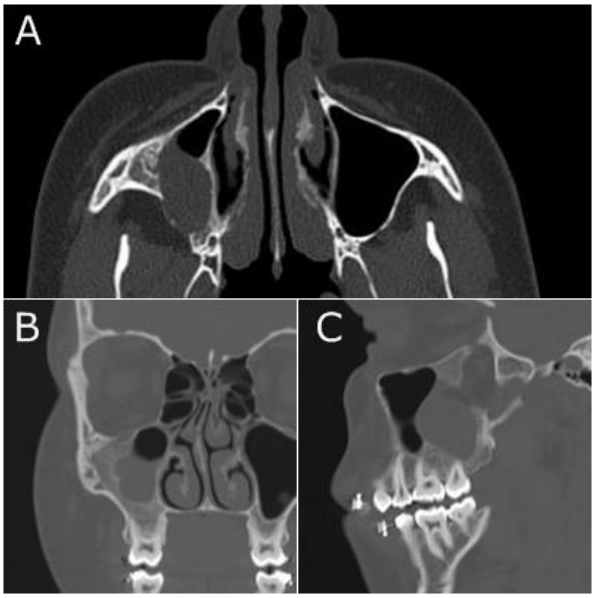

Figure 3A, B and C. Respectively Axial, Coronal and Sagital Sections of CT scan showing bone neoformation on sinus walls.

3. DISCUSSION

The OKC has a high recurrence rate due to its tendency to invade adjacent tissues (satellite cysts), it also figures as one of the most aggressive lesions of

odontogenic origin. The recurrence rate may depend on lesion size, the treatment modality employed and invasion of satellite cysts into the surrounding tissues[7].

Only 30% of OKC cases are associated to impacted teeth[8]. The occurrence of OKC in maxilla is lower when compared to the mandible, about 65% occur in the mandible, having a predilection for the posterior region. In maxilla it involves about 13%[3]. The involvement of the maxillary sinus, as occurred in the present case, is even rarer, occurring in less than 1% of cases[9,10].

The maxillary sinus is close to the developing teeth and root apices of the premolars and molars. It is vulnerable to odontogenic infections, cysts, and tumors of odontogenic origin. The diagnosis of this lesion is challenging, as maxillary radiographs do not offer characteristic features due to the overlap of several structures[11].

Maxillary sinus pathology can occur when the Schneiderian membrane is ruptured by conditions such as odontogenic pathology of the maxillary bone. Odontogenic infections and pathologies are responsible for 11% to 12% maxillary sinusitis cases[12].

Differently of when presented in mandible, OKC often shows an expansion of the alveolar bone with remodeling in maxila[13], and because its cortical bone is thin it can be easily perforated by the progression of the lesion and access places of noble structures, such as the maxillary sinus, nasal and/or orbital cavities[14].

Though literature shows teeth in unusual locations, ectopic eruption of teeth is rare. Nasal cavity, maxillary sinus, mandibular condyle, coronoid process, and palate are some of the documented sites. Some of possible etiologies of ectopic teeth are trauma, infection, developmental anomalies, and pathological conditions, such as odontogenic cysts[15]. When related to odontogenic cysts, it is believed that their growth in the maxillary sinus ends up displacing teeth buds, causing their ectopy.

OKC presents most frequently in second and third decades of life with the male predilection.4 In the present case, the lady affected was 17 years old.

Treatment of OKC has always been a debatable subject. The present study followed the most advocated treatment option in literature: the complete removal of the lesion and its margins as well as the associated impacted tooth/teeth.

Recently, marsupialisation or decompression followed by secondary enucleation have been discussed by some authors as the first line of treatment option for OKC. It has been stated that marsupialisation decompression leads to significant reduction in size and thickening of the lining of the lesion after which a secondary enucleation can be done easily with decreased morbidity.

Nonethless, there are few limitations of decompression since it needs patient compliance, might fail to completely eradicate lesion or there may be some malignant changes in the cavity which may go undiagnosed. Furthermore, very often a secondary enucleation is required. So, patient must go under two surgical procedures since there is consensus that the lesion needs to be removed in its entirety, given its high recurrence rate[16].

In this present case, location, presence of impacted third molar tooth and possibility of the lesion being infected made us decide the enucleation. Hence it is important for the clinician to consider OKC in the differential diagnosis for such lesions when they occur in a younger patient. Therefore, it is necessary to include OKC as a differential diagnosis for lesions with the presentations as seen in this case, even when they occur in younger patients.

4. CONCLUSION

OKC in the maxillary sinus is very rare and usually is discovered on periodic tests. The difference between OKC and other jaw cysts is its potential aggressive behavior and recurrence. To add to the literature, we emphasize the presence of OKC in the maxillary sinus and the importance of effectives treatments and long-term follow-up to detect any recurrence associated with the lesion when it occurs in the maxillary sinus.

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