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CASE REPORT

Surgical treatment of orthokeratinized odontogenic cyst in maxillary sinus: Case report in pediatric patient

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Abstract:

The orthokeratinized odontogenic cyst is a rare developmental cyst that affects the jaws. Since 2017 it has been classified as an independent entity of odontogenic keratocyst, since it presents differences in its biological behavior, histopathological aspects, as well as less aggressiveness and potential for recurrence. It affects young individuals, with predominance for the masculine gender. Their behavior is not aggressive, but can reach large proportions. The present case reported, in a male patient, 9 years old, with an expansive osteolytic process of large proportions was observed, taking all the extension of the left maxillary sinus. The patient was submitted to enucleation of the lesion under general anesthesia, is being followed up without signs of relapse.

Keywords: Bone Cysts; Pathology, Oral; Maxillary Sinus.

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INTRODUCTION

The orthokeratinized odontogenic cyst (COO) is a rare developmental cyst that affects the jaws, representing about 0.4% of maxillofacial cysts. It was first described by Schultz in 1927. The term odontogenic keratocyst (OKC) was later used by Philipsen in 1956 to define all cysts with keratin formation. COO was first described as a variant of odontogenic keratocyst by Wright in 1981. This lesion presented minor histopathological differences, aggressiveness and relapse rate. In 2017, the COO classification was accepted as a separate entity from OKC[1-5].

The pathogenesis of the lesion is still controversial, but it is believed to be related to remnants of the dental blade. It is a change that mainly affects young individuals, of the masculine sort and leucodermas. Usually indolent, slow-growing, unilocular radiolucent lesions with sclerotic and well corticalized margins of low aggressive behavior, however, may reach large proportions (1-7cm). Generally associated with the teeth included in the posterior region of the mandible[1,2,6,7].

The cystic capsule is composed of stratified squamous epithelium, which shows a surface of orthokeratin with varying thickness. Basal palisade and prominent layer that is characteristic of OKC is not present. As well as the absence of satellite cysts and thinner epithelial lining[1,4,8].

The treatment instituted for COO depends on some factors, such as the location, age, and size of the lesion. And it is usually more conservative, due to its reduced risk of recurrence (around 4%). The treatment of choice is based on enucleation and curettage, but decompression/marsupialization obtains good results as adjunctive or even definitive treatment. Carnoy solution, peripheral ostectomy and cryotherapy may be chosen[3,4,7,8].

This manuscript aims to report an exuberant case of orthokeratinized odontogenic cyst in the maxillary sinus. As well as conducting a brief review of the literature, in order to update the classification and distinction between COO and OKC, which has undergone recent changes according to WHO.

CASE REPORT

A 9-year-old male patient sought the Buccomaxillofacial Surgery and Traumatology service with complaints of dental absences and an asymptomatic increase in the posterior maxilla (Fig. 1).

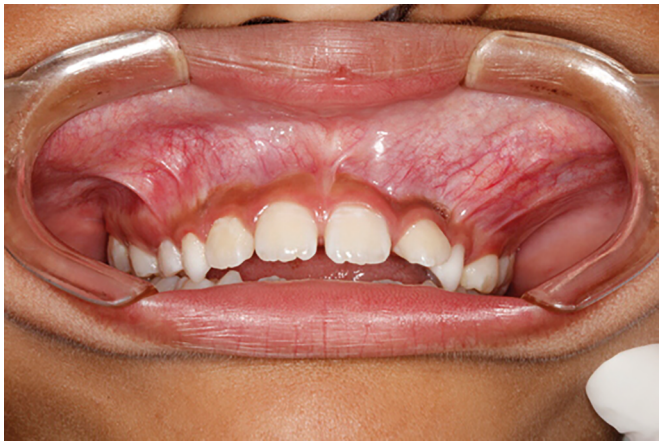

Figure 1. Intraoral image showing expansion in the left maxillary vestibule fundus region.

Orthopantomography and computed tomography, which showed a hypodense image in a region of the left maxillary sinus with a dental element included in the roof region of the maxillary sinus/orbital floor, root resorption of some elements and great expansion of the maxillary sinus walls with partial obstruction of the cavity left nasal. (Figs. 2-4)

Under local anesthesia with 2% lidocaine + 1: 100,000 epinephrine, it was aspiration puncture with straw staining contente (Fig. 5), and incisional biopsy was performed, concomitant with the drain installation to attempt decompression (Fig. 6). At anatomopathological examination, fragments of cystic capsule partially covered by orthokeratinized squamous epithelium with four to six layers of cells and showing a flat interface with the cystic capsule, composed of dense connective tissue, showed moderate lymphoplasmacytic inflammatory infiltration in some areas. In several areas, detachment of capsule lining epithelium and large numbers of keratin lamellae are noted. In some areas, the epithelial lining is thickened and exhibits twelve to fifteen layers of cells. With this, the diagnosis was closed in an orthokeratinized odontogenic cyst (Fig. 7).

In the 180-day follow-up of the decompression, no changes were observed in the lesion limits, and the patient, despite the care, evolved with multiple episodes of local infection, thus enucleation and curettage of the cystic lesion under general anesthesia and extraction of dental element included in the orbital floor region (Fig. 8). Large bone destruction was observed in the region of the anterior wall of the left maxillary sinus. In order to minimize the risk of postoperative buccal and buccal communication, the pedicled ipsilateral buccal adipose body was sutured by planes.

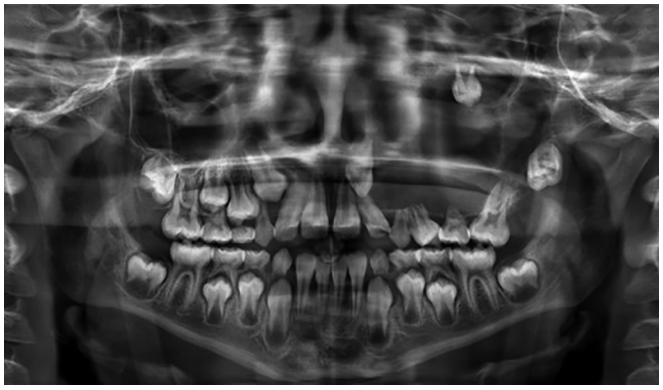

Figure 2. Panoramic radiographic image of the jaws showing radiolucent image inside the left maxillary sinus, with associated dental element.

In the post-surgical period antibiotic therapy was prescribed with amoxicillin 500mg + potassium clavulanate 125mg, 3x/day for 14 days, as well as analgesia with sodium dipyrone, mouthwashes with chlorhexidine 0.12% and irrigation of the nasal cavity with saline 0.9%. The patient took loratadine 10 mg (1x/day) for 5 days.

In the follow-up of 180 days after of surgery, no signs of relapse were observed. As complications, the patient evolved with transient infraorbital nerve paraesthesia, with remission within 90 days (Fig. 9).

DISCUSSION

In 2005, the WHO classified the variant with parakeratinized epithelial component in a keratocystic odontogenic tumor (TOQ), in contrast to the COO with orthokeratinized component and did not correspond to a neoplasia. However, in 2017, the original classification was reused, it was designated that they are odontogenic developmental cysts, the odontogenic keratocyst and orthokeratinized odontogenic cyst being different entities and both defined as cysts. In fact, OKC's classification as neoplasia in 2005 was not a consensus among oral pathologists[1,5,7,8].

In a systematic review by MacDonald-Jankowski[7], 192 cases found in the literature concerning COO were discussed. There was a ratio of approximately 2:1 between men and women. Higher prevalence of involvement in the mandible (2.4:1), particularly the posterior region. In the second decade of life has an increased occurrence in women, which suggests a relationship with menarche and hormonal factors. 48% of the cases were found in routine radiographic examinations, 41% referred to swelling and 24% associated pain. It is interesting to note that 68% of the reported cases presented

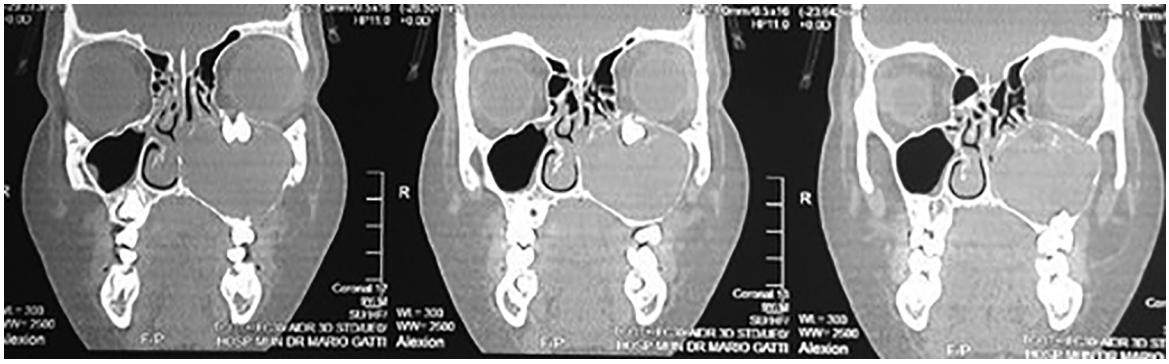

Figure 3. Computed tomography image in coronal section showing the lesion dimension, bulging of the lateral wall of the ipsilateral nasal cavity. Dental element impacted in the orbital region.

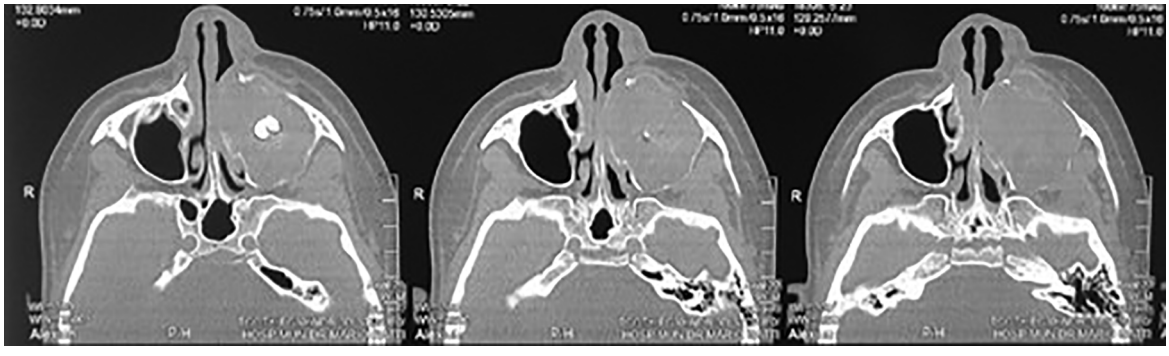

Figure 4. Image of computed tomography in axial section, obstruction of the left nasal cavity is observed. The cortex of the anterior wall of the ipsilateral maxillary sinus is thinned or ruptured.

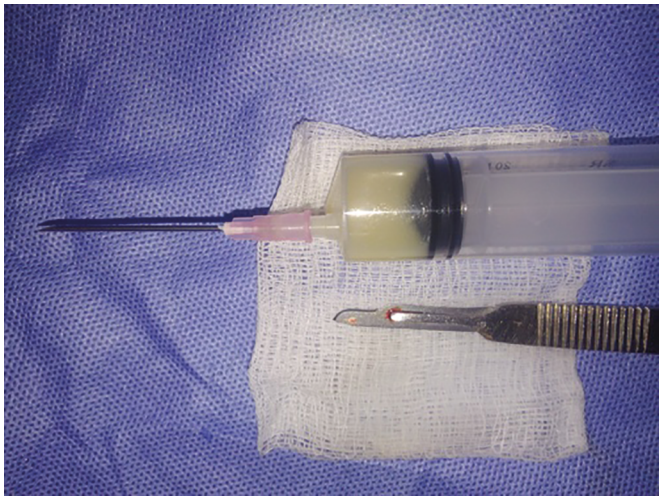

Figure 5. Aspiration puncture with straw staining contente.

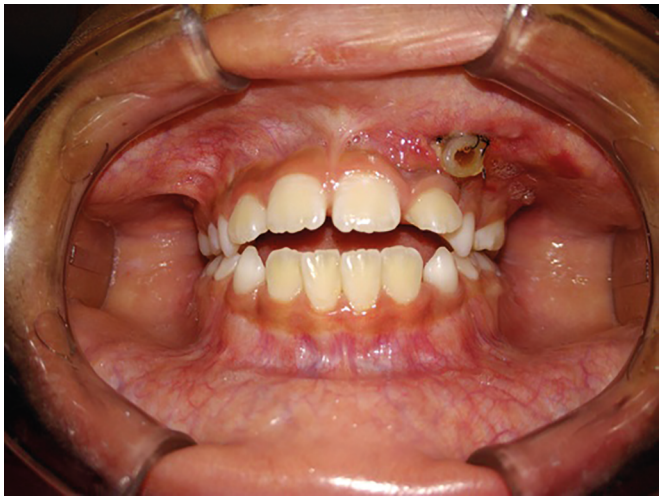

Figure 6. Device for decompression in position, aiming to reduce the size of the lesion.

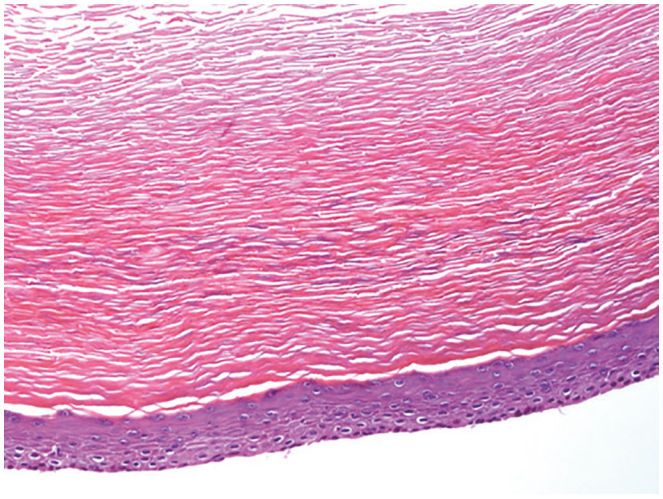

Figura 7. Surgical specimen after exeresis. There is a dental element (premolar) associated with the cystic capsule, probably the lesion originated from remnants of the dental blade of this tooth.

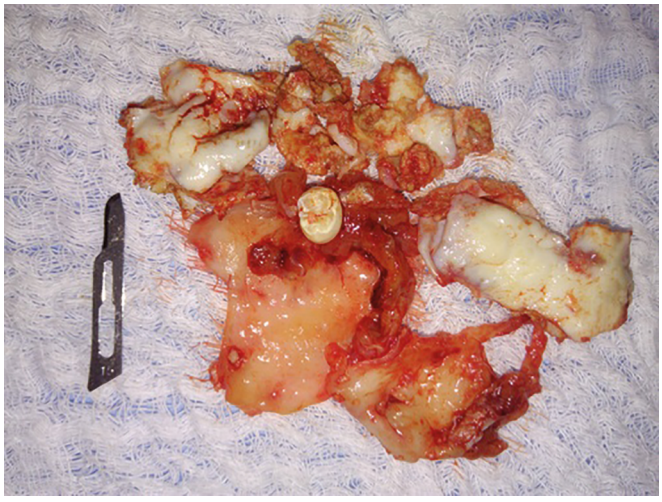

Figure 8. Photomicrograph that shows a cut of anatomopathological examination that exhibited de ragments of cystic capsule partially covered by orthokeratinized squamous epithelium with four to six layers of cells and showing a flat interface with the cystic capsule, composed of dense connective tissue, showed moderate lymphoplasmacytic inflammatory infiltration.

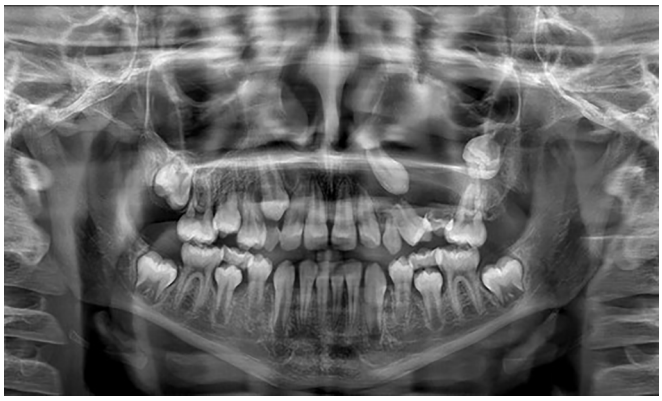

Figure 9. A 180-day post-operative panoramic image, which shows no radiographic signs of relapse, as well as showing the change in the position of the element 23 that is in an eruption path.

associated erupted tooth, which suggests a diagnostic hypothesis of a dentigerous cyst. The recurrence rate was 4.24%, all of which were reported in Asian patients, well below the 28.1% found in the OKC. A ratio of 1:9 was observed between the occurrence of COO and OKC[1,7,8].

The case reported corresponds to the literature, since it affected a young male patient and presented with an associated tooth. However, its location was unusual.

In 2017, the classification of this cyst was accepted as a separate entity for the first time. This differs both clinically and histopathologically from OKC. The importance of this differentiation lies in the relevance of the suitability of the treatment in relation to the biological behavior of each one. As well as, COO is not associated with any syndromes, does not have a high recurrence

rate and does not show aggressive clinical behavior, which indicates a better prognosis[2,5]. Histologically, basal COO cells are not well developed like those of OKC. Cells tend to be cuboidal and have little tendency for polarization. COO has a luminal surface coated with orthokeratin and a well-developed granular layer with different thicknesses[9,10].

Some rare COO presentations have been described in the literature, such as peripheral COO, mandibular condyle, multilocular condylar and COO with different associations, such as calcifying odontogenic cyst, ameloblastoma, heterotopic cartilage and squamous cell carcinoma. The case reported in this article is quite exuberant with regard to its size, location and the fact that the dental element is impacted in the orbit. It has been verified in the literature that this case is unprecedented, since there are no reports of COO of such proportions, taking the entire extension of the maxillary sinus[3,4,11,12].

CONCLUSION

It is interesting to note that most of the case reports described in the literature still consider the WHO classification of 2005, and there is a need for updating for didactic reasons with the publication of case reports and literature reviews. It is perceived that literature is constantly advancing and professionals must be aware of the changes. The correct diagnosis and treatment instituted compatible with the behavior of the lesion, corroborate to an adequate outcome. The presented case evolved without sequelae, in follow-up of 180 days postoperatively of simple enucleation.

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