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Odontogenic Keratocyst: Case Study of a Young Patient

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Abstract **Case Report**

Odontogenic keratocysts (OKCs) are benign cystic lesions of the jaws. They are aggressive with a relatively high recurrence rate. Most often they are discovered by chance during routine radiographic examination, they typically grow in an anteroposterior direction within the jawbone without causing significant expansion. OKCs generally occur in the second third, and fourth decades of life and are rare in pediatric patients. The management of these cysts is diverse and depends on several factors. The aim of the present study was to report an odontogenic keratocyst in a 13-year-old patient, associated with an impacted mandibular canine on the right side. The case was characterized by significant vestibulolingual expansion, a feature that differentiates it from typical presentations and underscores the importance of recognizing such variations in clinical practice. Management involved an initial decompression procedure, followed by subsequent enucleation of the lesion.

Keywords: Odontogenic cysts; Decompression; Case reports.

INTRODUCTION

The term 'odontogenic keratocyst' was first defined by the World Health Organization (WHO) in 2005 as a benign keratocystic odontogenic tumor with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behavior [1]. In 2017 and the latest edition of the WHO Classification of Head and Neck Tumours published in 2022, the condition was moved back to the cyst category [2].

There is currently no agreement in the literature about the best surgical technique(s) for treating OKCs. Nevertheless, it has been observed that conservative surgical management is not necessarily associated with recurrences characteristic of neoplastic disease [3].

The aim of this paper was to describe the clinical and histopathological management and the planning of conservative surgical treatment of a large right mandibular odontogenic keratocyst in a 13-year-old child associated with a retained tooth and its postoperative follow-up.

CASE REPORT

A 13-year-old child was referred by a colleague for a cystic lesion related to the impacted right mandibular permanent canine. The extraoral clinical examination did not reveal any significant asymmetry or neurosensory disorders, with no signs of inflammation or palpable lymph nodes. The intraoral clinical examination revealed a slight increase in the volume of the vestibular mucosa of the lower right region extended from 42 to 44 without color alterations (Fig 1).

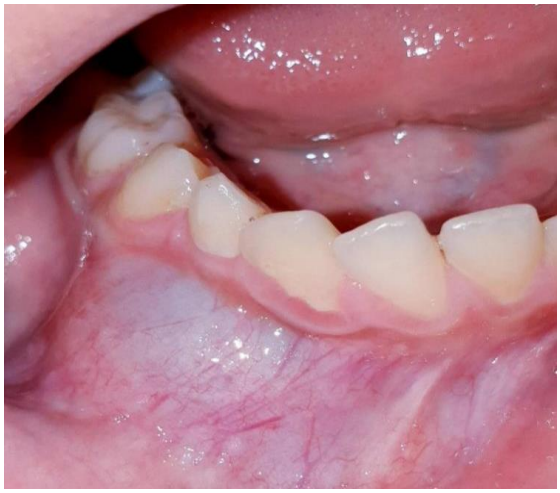

Figure 1: Intraoral view: Slight increase in the volume of the vestibular mucosa extended from 42 to 44

The patient consulted us with a CBCT that was requested by the colleague which showed an extensive radiolucent lesion, well-defined and corticated margins located between 42 and 44, involving tooth 43 impacted and pushed back to the basilar edge of the mandible (Fig 2).

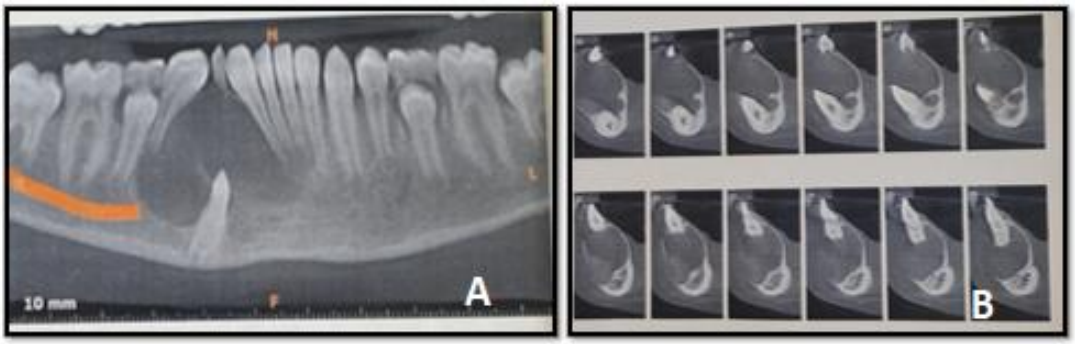

Figure 2: Cone Beam Computed Tomography; Fig 2A: Panoramic reconstruction, hypodense, and extensive lesion with defined borders surrounding the crown of right permanent canine. Fig 2B: Cross-sectional reconstruction, cortical expansion, thinning with disruption of the buccal cortex

After a thorough clinical and radiological examination, a presumptive diagnosis of a dentigerous cyst or odontogenic keratocyst was made.

Due to the size of the lesion, the presence of the permanent tooth, and the patient's age, surgical decompression was performed with extraction of the temporary canine. A sample of the lesion was removed and sent for histopathological study (Fig 3).

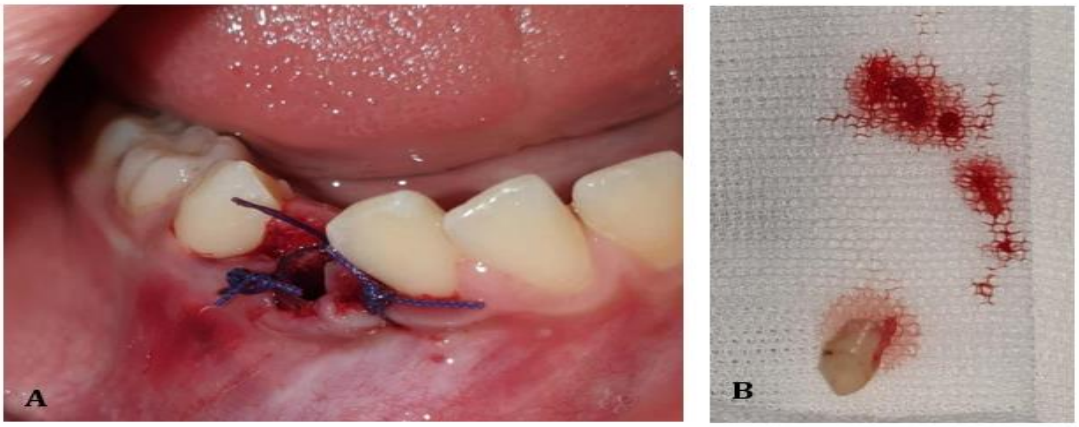

Figure 3: Decompression and incisional biopsy. Fig 3A: Placement of a drainage tube sutured to the mucosa. Fig 3B: Temporary canine and a sample of the lesion

A panoramic X-ray, was taken one month after the decompression, showed a significant reduction in the size of the cyst (Fig 4).

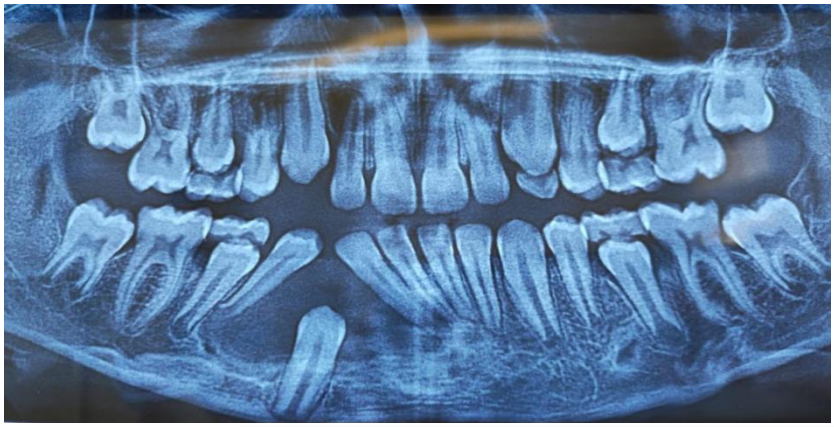

Figure 4: Panoramic radiograph: Reduction in the size of the lesion one month after decompression

Histological examination showed a connective tissue wall lined with parakeratinized stratified squamous epithelium which confirmed the diagnosis of parakeratinized odontogenic keratocyst. Therefore, a reintervention under local anesthesia, with total enucleation followed by thorough curettage and lavage of the bone cavity with abundant sterile saline solution, was performed. Tooth 43 was extracted (Fig 5).

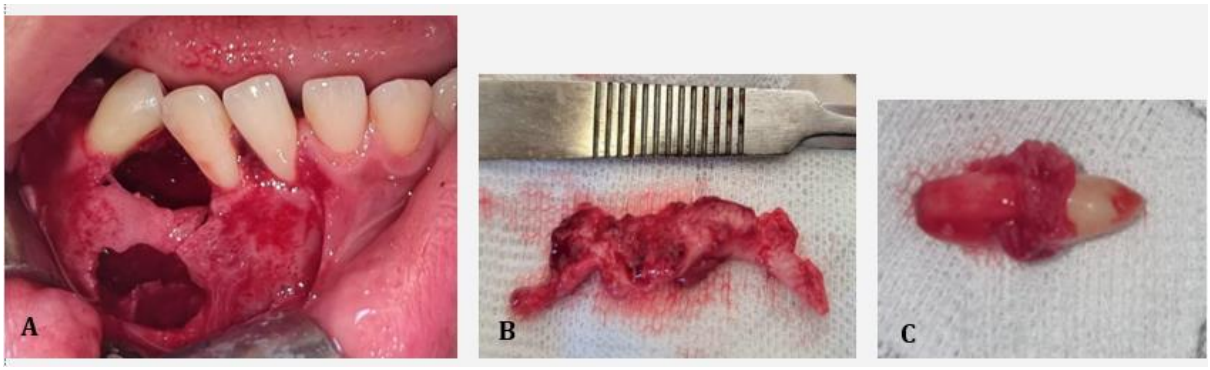

Figure 5: Surgical reintervention; 5A: Total lesion enucleation. Fig 5B: Surgical specimen. Fig 5C: Tooth extracted

Ten months later, the patient was seen for a clinical check-up and an evaluation with a panoramic radiograph. The radiograph showed excellent healing with bone neoformation in the affected region (Fig 6). Then, the patient was referred to an orthodontist for space closure and teeth alignment.

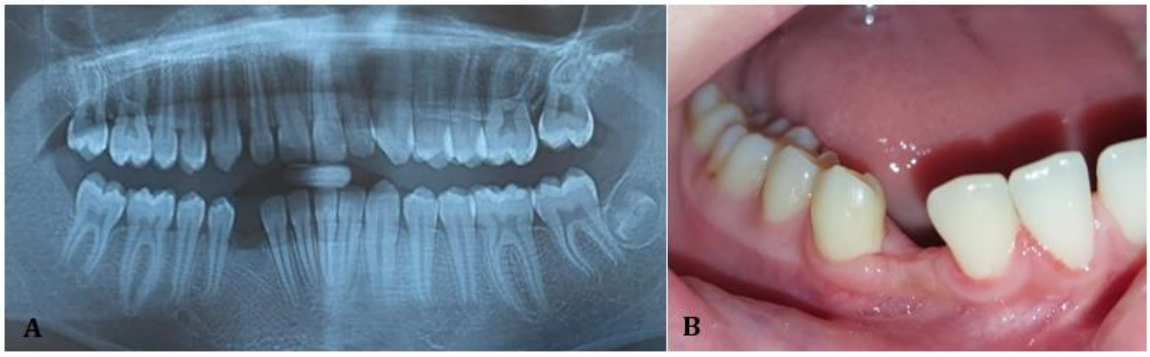

Figure 6 (A and B): Ten months postoperative follow-up

DISCUSSION

The odontogenic keratocyst (OKC) is one of the most studied cysts in oral pathology due to its aggressive behavior, high recurrence, and histopathological features. It is one of the most frequent odontogenic cystic lesions (approximately 10 to 15%) [4].

Odontogenic keratocysts are benign intraosseous lesions. Histologically, they can be of primordial origin (60% of cases) when they arise from the remains of dental lamina and are not associated with teeth; and of dentigerous origin in 40% of cases, derived from a reduced enamel organ and associated with retained teeth [5].

The age distribution is considerably broad, with a peak incidence in the third decade of life and a slight male predominance [6]. The mandible is more commonly affected than the maxilla, with a frequency of up to 70% for the ramus [7]. On the contrary, our case reports a lesion in a male child involving the premolar region of the mandible.

The clinical examination is often not very suggestive because of the paucity of symptomatology, as in our case. But when the size of the lesion becomes more important, the keratocyst can become symptomatic and manifest itself by inflammation, pain, a feeling of discomfort, or a spontaneous discharge of cystic fluid by fistulization [8].

Radiographically, KOs often present as well-defined unilocular or multilocular radiolucent lesions delineated by cortical margins. Unilocular lesions are predominant, which is the case in our patient [9]. On panoramic radiography, unilocular mandibular KOs may show sparse and incomplete septa, arranged perpendicular to the long axis of the lesion, which may give the appearance of a bifocal lesion [6]. Therefore, CT is considered superior to conventional radiography in differentiating OKCs from other unilocular or multilocular osteolytic lesions and in the preoperative assessment [10].

Approximately 30% of OKCs are associated with at least one unerupted tooth, most commonly the third molars [9]. This association occurs particularly in younger patients [11].

Despite their aggressive behavior, the keratocyst is characterized by its lack of expansive character, due to their ability to grow along the length of the bone with minimal bucco-lingual expansion [12]. For this reason, thinning of the cortices is observed in the presented case.

The literature reported that the perforation of the cortical bone is not an unusual feature with an intraoperative incidence ranging from 39 to 51%. OKCs may show tooth displacement and root resorption.

The OK can take two histological forms depending on the type of keratinization: parakeratotic or orthokeratotic. In its parakeratotic form, it is referred to as a keratotic odontogenic tumor which is most often characterized by the presence of multiple OKs, usually associated with the nevoid basal cell carcinoma syndrome, also known as Gorlin-Goltz syndrome, an autosomal dominant multisystem disease [4].

The management of KCOTs remains a debatable topic due to their high recurrence rate.The treatments proposed in the literature are manifold and depend on the patient's age and compliance as well as the size and location of the lesion. However, the one with the lowest percentage of recurrence is total lesion enucleation, followed by curettage, abundant washing with saline solution, and extraction of compromised parts [13].

Conservative treatment, i.e., OKC decompression or marsupialization (transformation of the cyst into an open pouch, allowing continuous drainage) is not recommended due to very high recurrence rates (17–56%) [15]. Decompression should be followed by enucleation with curettage, as it was in the case of our patient. These procedures should be followed by a periodic radiographic monitoring of patients, at least for the first 5 years to report the success and healing of the lesion and avoid recurrence caused by multiple factors, including incomplete enucleation and curettage due to a weak cyst wall or invasion of adjacent soft tissues [14].

Recent studies have explored the association between OKC recurrence and clinicopathological factors such as large size, cortical perforation, tooth involvement within the cyst lumen, and the presence of daughter cysts [15, 16]. These findings suggest that these factors may serve as prognostic indicators of a high recurrence risk.

Based on these data, we conclude that the combination of clinical, radiological, and histological findings is useful in both confirming the diagnosis and guiding the choice of treatment modality for OKCs.

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Competing Interests: Authors have declared that no competing interests exist.

AUTHORS' CONTRIBUTIONS

All authors have read and agreed to the final version of this manuscript and have equally contributed to its content and to the management of the cases.

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