

Unusual odontogenic keratocyst with impacted maxillary central incisor: A diagnosis challenge

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ABSTRACT

Background: Odontogenic keratocyst (OKC) is considered an odontogenic cyst, with the unique clinical characteristics of this entity being local destruction, aggressive behavior, a high recurrence rate, specific histopathologic features, and the tendency for multiple lesions when associated with syndromes. Odontogenic keratocyst accounts for 12–14% of all cysts in the jaws. **Purpose:** The aim of this case report is to present an unusual odontogenic keratocyst associated with impacted maxillary central incisor, case management of anterior maxilla odontogenic keratocyst involving an adjacent tooth, and its postoperative six-month follow-up. **Case:** A 13-year-old female patient was referred to the Universitas Airlangga Hospital with a complaint that her upper front teeth had not erupted. The parents revealed that the patient had suffered trauma from falling down the stairs at the age of four, i.e., eight years ago. **Case Management:** The cyst was enucleated together with the associated permanent upper right central incisor, retained deciduous upper right central incisor, the permanent upper right lateral incisor, and upper right canine. Post surgical treatment, the patient used a partial denture to increase her self-confidence. **Conclusion:** Enucleation of the cyst with extraction of the permanent teeth should be performed to avoid recurrence of the OKC. Periodic radiographic evaluation is required with long-term follow-up.

Keywords: dental trauma; dentigerous cyst; enucleation; impacted teeth; odontogenic keratocyst

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INTRODUCTION

The term “odontogenic keratocyst” (OKC) refers to a benign intraosseous cystic lesion of the gnathic bones that is most likely caused by diminished enamel epithelium of the dental follicle, down growth of the surface epithelium’s basal cell layer, or traumatized implantation.^{1,2} The World Health Organization (WHO) defined this disease as a keratocyst odontogenic tumor in 2005 when it was shown to be an odontogenic epithelium-derived neoplasm.^{2,3} Compared to other odontogenic cysts, odontogenic keratocysts are the third most frequent, accounting for around 12–14% of all odontogenic cysts in the jaws.⁴

OKC generally affects the mandibular molar area and ascending ramus and is more common in males (M/F 2:1). It usually manifests during the 2nd and 3rd decades of life.⁵ OKC is typically asymptomatic but can, however, present with swelling, pain, purulent discharge, paresthesia, nasal obstruction, and mobility of teeth. In addition, it exhibits a high recurrence rate ranging from 2,5 to 62%, which is frequently related to the chosen treatment.⁶ OKC has been linked to impacted teeth and has the potential to displace neighboring teeth, resorb neighboring tooth roots, and extrude erupted teeth.⁷ Malignant transformation of OKC has frequently been associated with a painless benign expansile lesion of the jaws, presenting a radiographic

picture similar to the cystic lesions from which they arise.⁸ Untreated OKC has the potential to grow significantly and cause damage to adjacent structures by being compressed, resulting in the teeth shifting, loosening, or tilting.⁹

The management of OKC aims to reduce the risk of recurrence while minimizing, at the same time, the morbidity for the patient. Size, position, patient's age, and association with critical structures all play a role in OKC's surgical treatment. Several treatment options are reported for OKC.¹⁰ In this case presented, lesion of the cyst involved several teeth. The aim of this case report is to present an unusual case of odontogenic keratocyst in a 13-year-old girl, associated with an impacted maxillary central incisor and involving an adjacent tooth, with six-month follow up.

CASE

A 13-year-old female patient was referred to the Universitas Airlangga Hospital with a complaint that the upper front teeth had not erupted. The parents revealed that the patient had suffered trauma from falling down the stairs at the age of four, i.e., eight years ago. Clinical examination revealed no signs of discomfort, systemic disease, or lymphadenopathy. The extraoral examination revealed no facial asymmetry, edema, hyperemia, or fistulas (Figure 1A–B). On intraoral examination, #11 and #13 were clinically not erupted. No swelling, edema, hyperemia, or fistulas were diagnosed. Bulging of the maxillary right canine buccal region was palpable, caused by mass (Figure 1C–E), and followed with pain. Tooth #51 was non-vital with mobility grade 2 (Miller Classification), while #12, and #21 were vital.

The panoramic radiograph of the patient showed a well-defined radiolucent lesion on the right region of the central incisive maxilla that extends to the mesial of the canine and showed the impacted right permanent maxillary

central incisor and canine (Figure 2A). Due to the size of the lesion, a cone-beam computed tomography (CBCT) image was taken and revealed a lesion covering the coronal 11 with an area of 16.92 x 9.3 mm that extends to coincide with the base of the nasal fossa and the mesial tooth 13. The image showed a single lesion with well-defined corticated boundaries (Figure 2B–E).

The clinical and radiological findings led to the provisional diagnosis of dentigerous cysts. The patient was then referred to the Oral and Maxillofacial Surgery department. Enucleation of the cyst along with odontectomy of the unerupted tooth and followed with histological examination to ensure the definitive diagnosis of the lesion as performed.

CASE MANAGEMENT

The unerupted status of #11 and #13 in the 13-year-old patient was comprehensively managed intradisciplinary to achieve complete healing and increase the patient's quality of life. The procedure was performed under general anesthesia (GA) due to the patient's age and the cyst's proximity to a critical region. Preoperative examinations were carried out, and no systemic problems were noted that would prevent or complicate surgery. The parents were informed regarding the diagnosis, treatment options, prognosis with and without treatments, advantages, risks associated with the procedure, and potential adverse reactions. Once agreement was reached, the informed consent was signed. Patient then underwent thorax radiographic, laboratory and SARS-CoV-2 screening of nasopharyngeal and oropharyngeal swabs to ensure safety.

Enucleation of the cyst was done by the oral and maxillofacial surgeon. Extraction of #51, #11, #12, #13 were unintended administered considering the extension

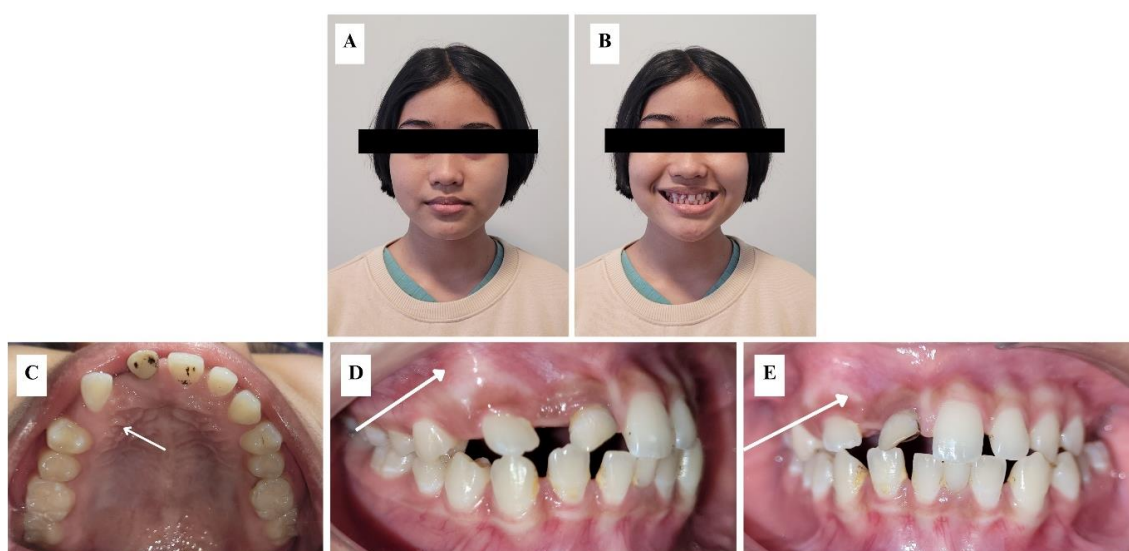


Figure 1. Pre-operative photo. Extra oral image of the patient (A–B). Intra oral image of the patient with bulging of the maxillary right canine buccal region (C–E).

of the lesion. Preventive restorations using resin and sealant, as well as topical application fluoride for caries prevention, were performed on #16, #17, #26, #27, #36, #37, #46, #47. After ensuring that the patient was fully sedated, an extraoral asepsis technique was conducted using 70% alcohol and 10% povidone-iodine intraorally. A vasoconstrictor consisting of 2 ampoules of Lidocaine hydrochloride 2% with adrenaline 1:80,000 diluted with 6 ml of saline was injected with a 21-gauge syringe using a local infiltration technique, and a full-thickness trapezium flap was raised, extending from the distal surface of the left central incisor to the distal surface of the permanent

first. (Figure 3A). A round carbide burr was utilized to widen the bone defect to the buccal window, allowing access to the cyst and the affected tooth (Figure 3B).¹¹ A needle aspiration test revealed cyst fluid, which was then deflated. The cysts were enucleated together with the associated permanent upper right central incisor, retained deciduous upper right central incisor, the permanent upper right lateral incisor, and upper right canine (#51, #11, #12, #13). Curettes were subsequently utilized to remove the soft granulation tissue (Figure 3C–D). Spongostan™ was then applied to the defect region and sutured with silk Vicryl 4.0 (Figure 3E).

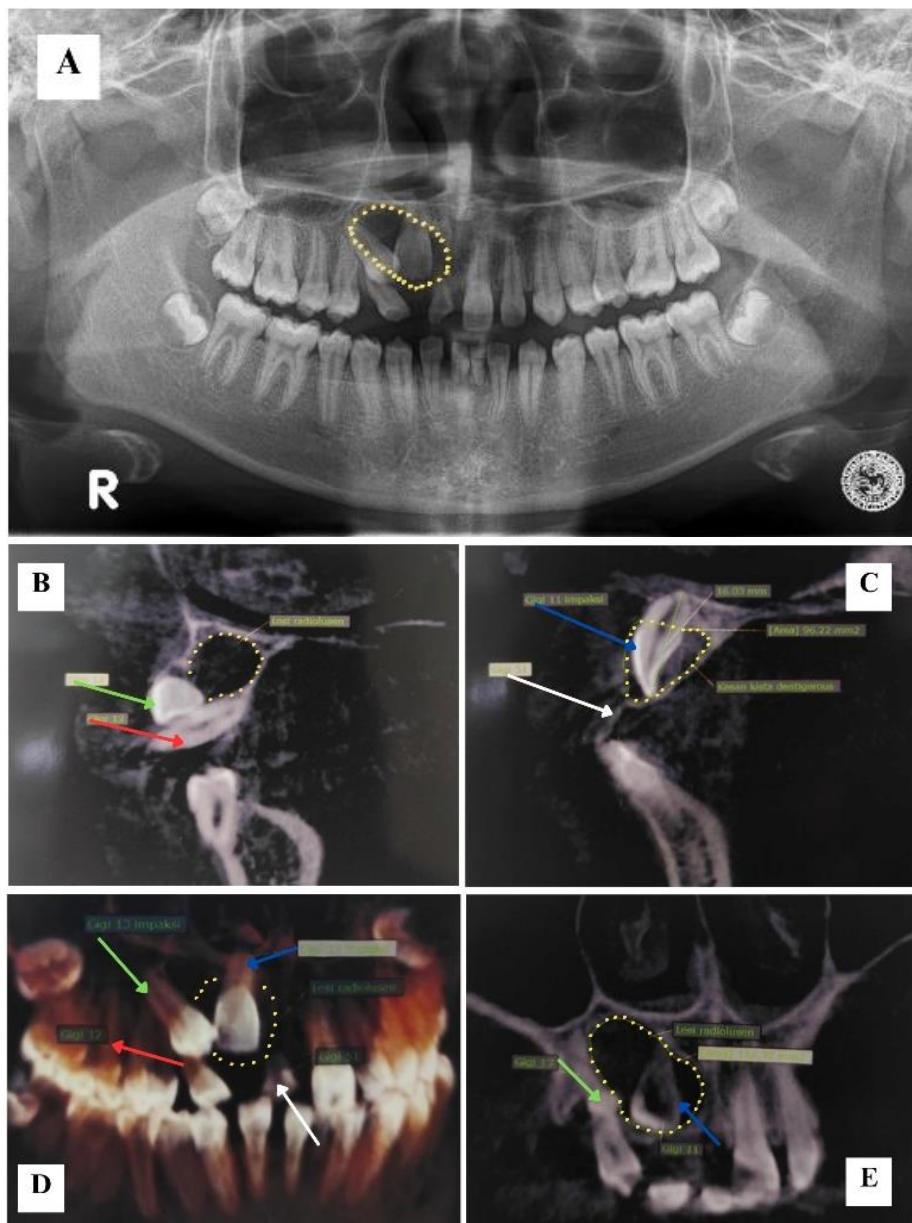


Figure 2. Preoperative radiograph views: (A) the panoramic view showed the well-defined radiolucent lesion around impacted maxillary right incisor; (B–C) the CBCT, sagittal view shows the lesion measuring 17,01 mm in length and 10,13 mm in width; (D) 3D and (E) coronal view shows the lesion measuring 16,92 mm in length and 9,3 mm in width. Yellow dots show the odontogenic keratocyst; green arrow is the impacted canines (#13); red arrow is the lateral incisor (#12); blue arrow is the impacted central incisor (#11); and white arrow is the deciduous central incisor (#51).

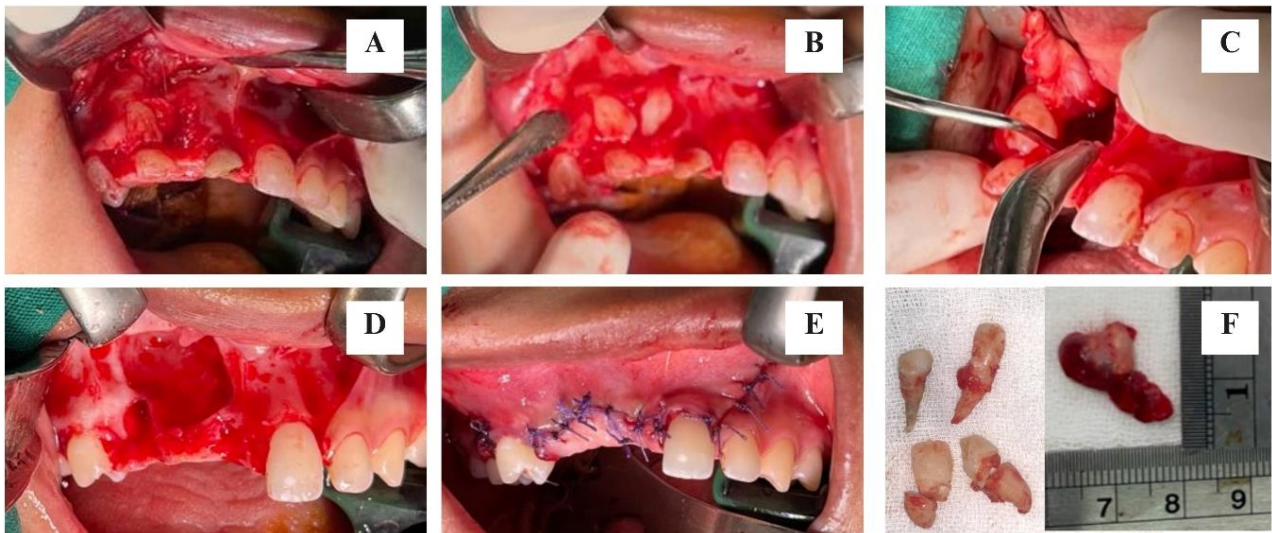


Figure 3. Surgical procedure: (A) full-thickness trapezium flap; (B) enlarge the bony defect to the buccal window to gain access to the cyst and the tooth with the defect; (C) enucleation of the cyst; (D) evaluation; (E) suturing; (F) specimen postoperative tooth 11,12,13, 51 and the cystic specimen after enucleation.

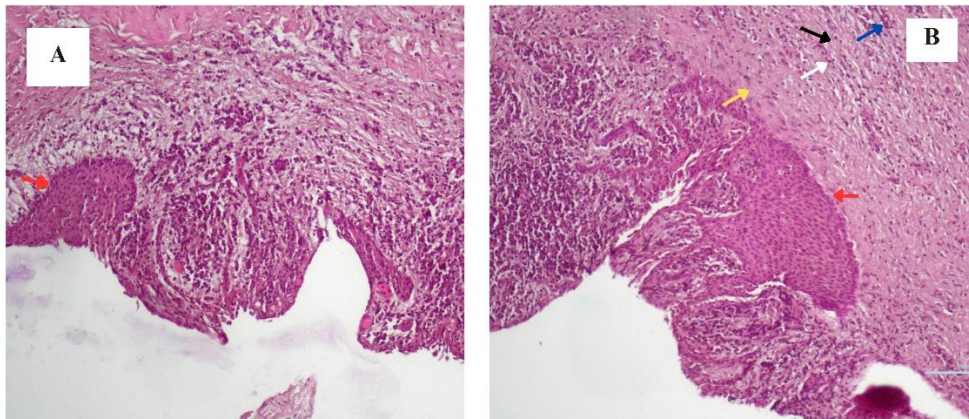


Figure 4. (A–B) Histopathologic features indicate an odontogenic keratocyst. Hematoxylin and eosin (H&E stain, x100) characteristics indicating a palisade arrangement of basal cells (red arrow); visible collagen fibers (yellow arrow) are arranged in parallel; there is a proliferation of blood vessels and chronic inflammation cells, including lymphocytes (white arrow); macrophages (blue arrow); and plasma cells (black arrow).

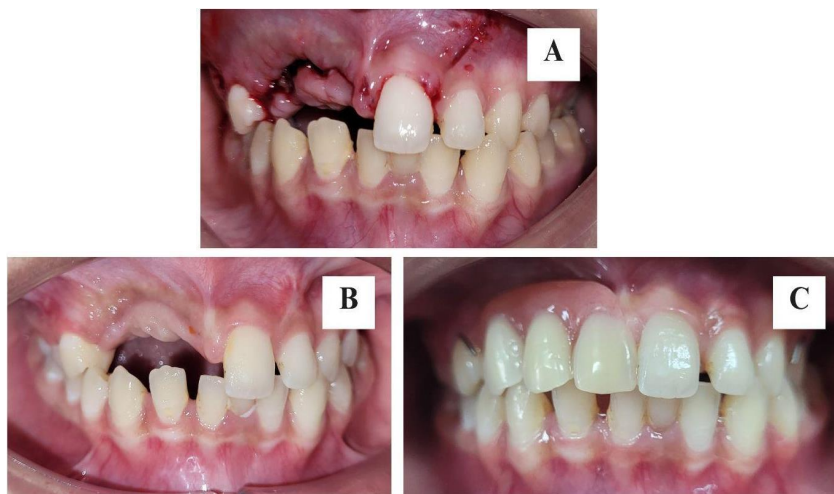


Figure 5. Postoperative intraoral view: (A) seven days postoperative; (B) one month postoperative; (C) partial denture insertion.

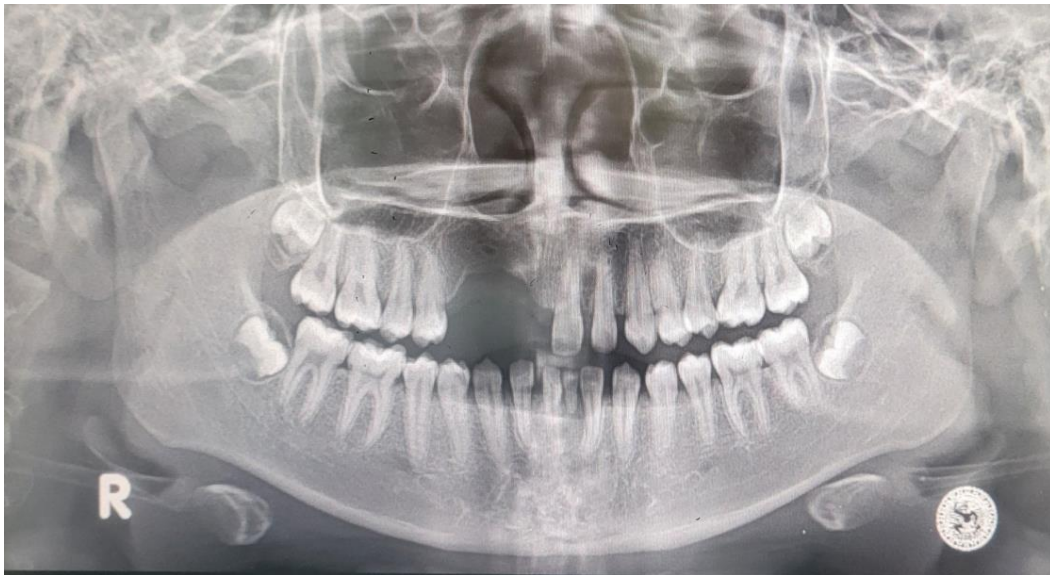


Figure 6. Postoperative panoramic radiograph, in the region of the right anterior maxilla.

The histological examination of the specimen showed a cystic lining along with stratified squamous epithelium without rete pegs, with a thickness of 6–8 layers and accompanied by para-keratinization on the surface of the epithelial layer. The basal layer consisted of cuboidal or columnar cells arranged in a palisade manner with hyperchromatic nuclei. In the connective tissue capsule, where visible collagen fibers are arranged in parallel, there was a proliferation of blood vessels and chronic inflammation cells, including lymphocytes, macrophages, and plasma cells. All these were suggestive of an OKC (Figure 4).

The patient was recalled after one week postoperatively and no sign of pain was found. Extraoral examination revealed no edema, hyperemia, or discomfort. On intraoral inspection, the surgical wounds on the edentulous area of 13, 12, and 11 were properly stitched, with hyperemia and minimum debris. The sutures were removed (Figure 5A). A month after the surgery, a partial denture was placed to increase the patient's self-confidence and comfort (Figure 5B–C). The patient was advised to eat with the partial denture in place and only remove it for cleaning. Follow-up consultations were planned every six months after surgery to ensure that there were no recurrences. A postsurgical orthopantomogram (OPG) performed 6 months after surgery indicated no radiolucent lesion of the maxilla right incisor (Figure 6).

DISCUSSION

Odontogenic keratocyst is classified as an odontogenic cyst, with differentiating clinical features including local destruction, aggressive behavior, a high recurrence rate, specific histopathologic features, and the tendency to multiply when associated with syndromes such as Nevroid

basal-cell carcinoma syndrome (NBCCS) or Gorlin-Goltz syndrome.^{12–13} OKC is uncommon in the anterior maxillary region, since it is more commonly found in the mandible or posterior/ramus area. In one series, over 50% of cases were associated with an impacted tooth; thus, clinically, they can mimic dentigerous cysts.¹⁴

Dentigerous cysts are the most frequently developing odontogenic cysts linked with impacted teeth, affecting mostly middle-aged people. The cyst is still asymptomatic and was found by coincidence during a routine radiological scan. It is identified by its connection to the crown of an erupting tooth. The clinical examination and radiological results of this cyst are consistent with our case. However, histological studies are necessary to rule out dentigerous cysts.^{15,16} Based on our patient's radiological picture and clinical evaluation, we determined that no biopsy was required due to the particular characteristics of a dentigerous cyst. Furthermore, the inflammation caused by the biopsy might have altered the histological outcome of the postoperative specimen.¹⁷

A clinical examination, radiographic imaging, and, most importantly, the results of histopathologic examination are used to make the diagnosis. Radiography plays a crucial role in diagnosing, arranging treatments, and evaluating outcomes. The three-dimensional geometric accuracy of CBCT compared to conventional radiography has been identified as a significant advantage. Furthermore, as compared to typical computed tomography (CT) scans, this technique provides high isotropic spatial resolution of osseous structures with a low radiation dose.¹⁸

OKC has characteristic radiographic features such as a radiolucent lesion with a corticated and scalloped border, minimal expansion, especially toward the medial side, and growth along the internal aspect of the mandibular or maxilla bone. In most cases, the lesion is associated with impacted teeth and results in the extrusion of erupted

teeth, displacement of neighboring teeth, and resorption of neighboring tooth roots.¹⁹ This radiograph characteristic was shown in this case to be similar to another study reported by Berty et al.²⁰ OKC may occur due to traumatic implantation, down growth of the basal cell layer of the surface epithelium, or reduced enamel epithelium of the dental follicle. Previous research found two examples of OKCs caused by both trauma and non-trauma. In this study, our patient had a history of trauma eight years earlier, which might be one of the triggering variables that induce epithelial rests to proliferate, shutting off the inflammatory or traumatized region from the surrounding healthy tissues.^{21,22} Chronic inflammation's histopathologic traits include the presence of macrophages and lymphocytes, as well as the growth of tiny blood vessels.

The literature describes several OKC therapies, ranging from conservative procedures to more invasive surgery.²³ Conservative methods such as decompression preserve tissue and cause minimal aesthetic and functional damage, while marsupialization and enucleation are considered as standard treatment methods but have a high recurrence rate. Radical surgical treatment has lower recurrence rates, causes more aesthetic-functional damage, and affects the patient's quality of life. To lower the recurrence rates, a conservative treatment combined with adjuvant therapies, such as marsupialization or decompression procedures coupled with enucleation and peripheral osteotomy, cryotherapy, or Carnoy's solution, remains an option.^{24–26}

Enucleation and potential peripheral osteotomy can be used to treat smaller OKCs and achieve good bone borders. Larger OKCs may require marsupialization or a resection.^{27,28} OKC are surgically removed, although further multidisciplinary operations such as surgery, pediatric dentistry, and, in certain cases, prostheses are frequently necessary. Removal of the whole cyst completely, with extraction of the teeth or tooth buds involved by the cyst, is a severe surgical treatment that gives the lowest recurrence rate, similar to previous studies reported by Rączkiewicz et al.²⁹ In this case, decompression procedures coupled with enucleation was not performed because we were punctured to deflate it, and in CBCT no detect cortical perforations.³⁰

Regardless of the treatment option, in patients treated at developmental age, close clinical and radiographic follow-up are scheduled more frequently in the first year following treatment, often every 3–6 months, then tapered off to once every 6–12 months, depending on clinical and radiographic findings. Radiographic evidence of reduction in lesion size associated with a clinically functional tooth may suggest a "healing" bone defect and should continue to be observed biannually. A limitation of our case report was the lack of patient monitoring after 6 months of follow-up. In conclusion, enucleation of the cyst with the extraction of the permanent teeth should be performed to avoid the recurrence of the OKC. Long-term follow-ups are essential, along with periodic radiographic examinations.

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