Submitted: 13 February, 2023 Accepted: 20 March, 2023 Published: 03 November, 2024 DOI:10.22514/jocpd.2024.142

CASE REPORT

Journal of Clinical Pediatric Dentistry

Large mandibular odontogenic keratocyst treated by decompression and secondary enucleation: a case report

Jianfeng He[1], Huijing Wang[1], Jun Zeng[1], Libin Zhou[1,*]

[1]Department of Oral and Maxillofacial Surgery, School and Hospital of Stomatology, Guangdong Engineering Research Center of Oral Restoration and Reconstruction & Guangzhou Key Laboratory of Basic and Applied Research of Oral Regenerative Medicine, Guangzhou Medical University, 510182 Guangzhou, Guangdong, China

***Correspondence**
fmmumario@126.com
(Libin Zhou)

Abstract

Odontogenic keratocyst (OKC) is a common developmental odontogenic cyst in clinic patients. Odontogenic cysts are often treated by enucleation, curettage, marsupialization and decompression. With apparent advantages, marsupialization and decompression are often the preferred option for adolescents with large jaw cysts. This article aimed to report a case of large OKC involving the left mandibular second molar and third molar in a 13-year-old adolescent. Decompression was performed to preserve the second molar and inferior alveolar nerve, and the third molar was extracted. After ten months of follow-up, cyst volume decreased significantly, and the impacted second molar erupted spontaneously through the bony window. Subsequently, the residual cyst was removed by enucleation. After 18 months, the second molar erupted to the occlusal plane and played a normal function. No evidence of recurrence was found during the entire follow-up period.

Keywords

Odontogenic keratocyst; Decompression; Impacted tooth; Tooth eruption

1. Introduction

Odontogenic keratocyst (OKC) is a developmental odontogenic cystic lesion with aggressive growth potential and a high recurrence rate, which is derived from odontogenic epithelium [1]. OKC was first described by Phillipsen in 1956 [2]. Until 2005, OKC was classified by World Health Organization (WHO) as Odontogenic keratocyst tumor (KCOT) [3]. However, it was later changed to odontogenic keratocyst again in 2017 [4]. In addition, OKC has a certain malignant potential, so the nomenclature and treatment of OKC remain still controversial [5]. Odontogenic cysts have similar characteristics in clinical and radiological examination. Because the envelopmental type of Odontogenic keratocyst (OKC) contains adjacent unerupted teeth and radiographs may reveal a radiolucent cystic lesion around the crown, it is often misdiagnosed as a dentigerous cyst [6]. Histological examination is the gold standard for diagnosis [7].

The postoperative recurrence rate of odontogenic keratocysts ranges from 14.8%–21.1% [8–10]. Cystectomy and decompression are both effective for the treatment of jaw cysts. Compared to cystectomy and decompression separately, the combination of cystectomy after decompression has a significantly lower recurrence rate [9, 10]. Younger patients treated with decompression for longer would get a better treatment effect [11]. This paper reports on a large odontogenic keratocyst in the left mandible that was treated by decompression and secondary enucleation.

2. Case report

A 13-year-old male patient came to the oral and maxillofacial surgery clinic with a chief complaint of progressive swelling in the left face for more than a week. The patient denied systemic disease and a history of drug allergies.

Extraoral examination revealed an asymmetric face with a bulging left mandible. The color and texture of facial skin are normal (Fig. 1). The left lower lip was hypoesthesia, but pain can still be perceived.

Intraoral examination demonstrated mandible expansion from the vestibular sulcus of the left mandibular second premolar to the buccal side of the retromolar pad (Fig. 2). The overlying mucosa was hard and tender on palpation. Teeth 37 (left mandibular second molar) and 38 (left mandibular third molar) were absent in the dentition.

Cone beam computerized tomography (CBCT) examination showed a unilocular radiolucent area with a clear boundary at the angle of left mandible and the mandibular ramus (Fig. 3). Tooth 37 is affected in a lower position close to the apex of tooth 36. Moreover, ectopic tooth 38 impacted near the mandibular notch. The shaded area contained teeth 37 and 38, measured approximately 70 $\times$ 50 $\times$ 27 mm. The mandibular outline was signally expanded, and the bone cortical was partially defective at the anterior lesion edge. There was no obvious resorption of the adjacent tooth root, and the mandibular canal was invisible in the CT image (Fig. 4).

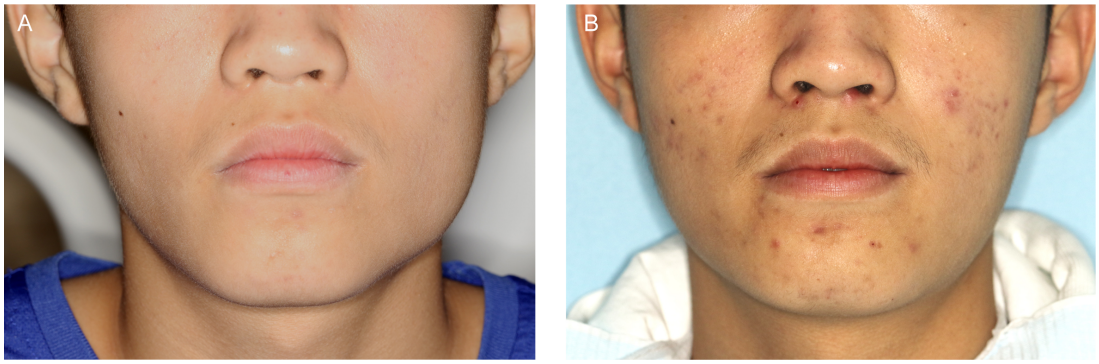

FIGURE 1. Extraoral photos of patient. (A) Extraoral photo showing an asymmetric face in first visit. (B) Extraoral photo eighteen months after enucleation.

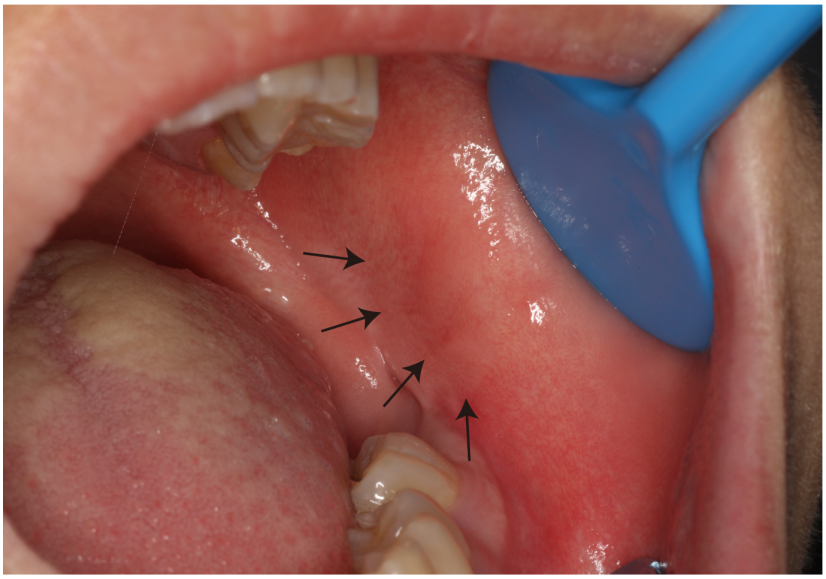

FIGURE 2. Intraoral photo in first visit showing the bulge at buccal side of retromolar pad (the area indicated by the black arrows).

3. Treatment

3.1 First stage: decompression

Decompression was performed under general anesthesia, and the impacted third molar was extracted. During the operation, we first made an incision to create a bony fenestration of about 20 × 20 mm at the distal of tooth 36. After the incision, the cholesterol crystal overflowed, and white keratin was found in the cyst cavity. The cyst tissue was thick and tough. However, teeth 37 and 38 were undetected in the cystic cavity because the crowns were covered by cyst tissue. The biopsy defined a diagnosis of odontogenic keratocyst (Fig. 5A,B). One week after the operation, an obturator was manufactured (Fig. 6A) and inserted into the fenestrae to maintain the surgical window during decompression (Fig. 6B). The patient was instructed to irrigate the cavity with a sterile saline solution twice a day to avoid infection.

Regular clinical and radiological examinations were arranged throughout the decompression period. Tooth 37 moved to bone fenestration at six weeks of follow-up (Fig. 7). A CT scan three months after the first stage operation indicated that cyst volume decreased significantly (Fig. 3). The mandibular canal could be clearly identified in a normal position (Fig. 4).

Ten months after decompression, a new CBCT examination showed that the radiolucent lesion size was reduced to 33 × 11 × 15 mm (Figs. 3,4). In the oral cavity, we found that the second molar spontaneously erupted with a lingual tilt and almost reached the occlusal plane (Fig. 8). Because the fenestration closed (Fig. 8) and the decompression achieved remarkable results, the enucleation was scheduled.

3.2 Second stage: enucleation

Ten months after the first stage operation, the second stage operation, the residual cyst was performed under general anesthesia. After enucleation, the residual bone cavity is filled with bone graft materials (Bio-Oss Collagen® 100 mg × 4, Geistlich Pharma AG, Wolhusen, Switzerland). Pathological examination confirmed the final diagnosis of OKC again. One and a half years after enucleation, a radiograph showed an ideal osteogenesis effect in the entire cavity (Figs. 3,4). The crown of tooth 37 moved about 11.6 mm compared to the preoperative CBCT. In clinical examination, the contour of mandible and face gradually changed into symmetry.

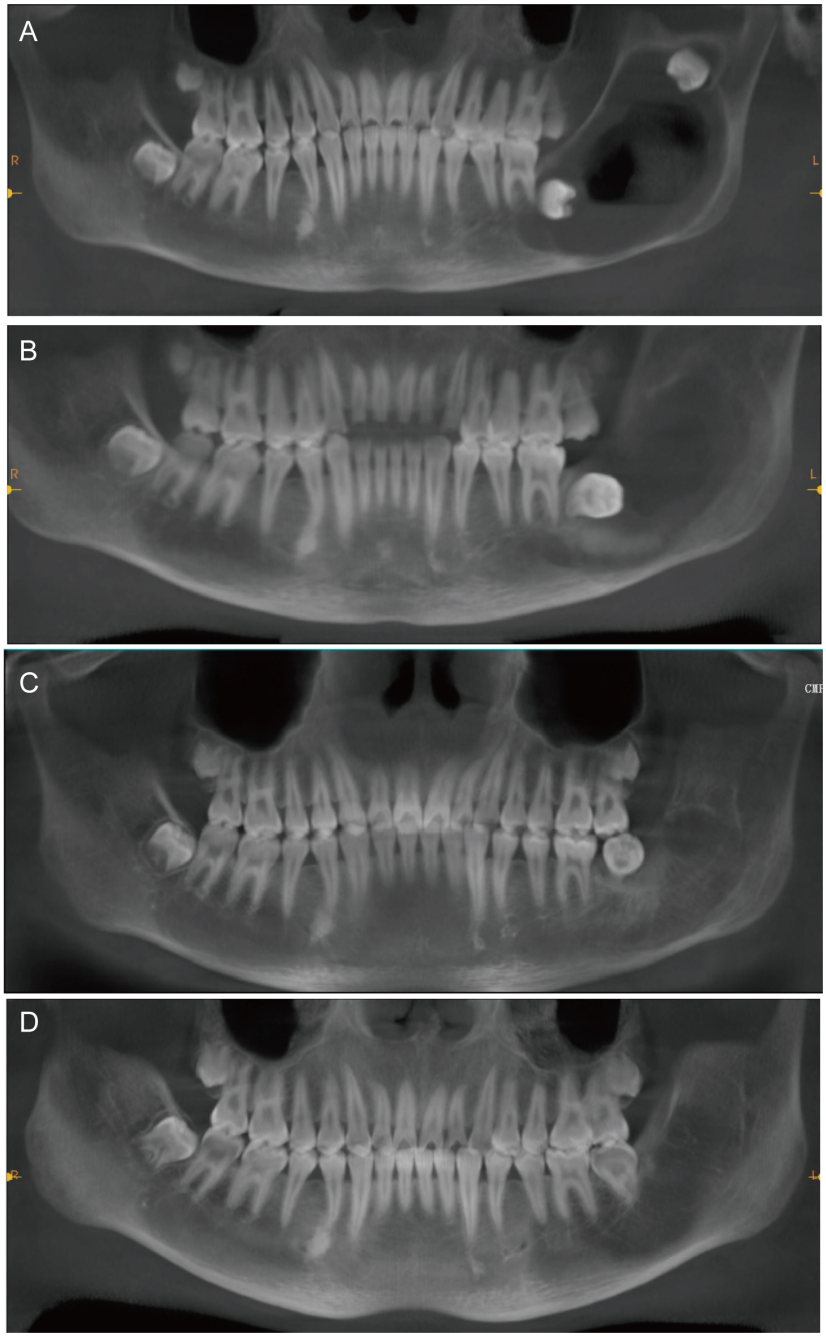

FIGURE 3. Reconstructed CBCT images in different periods. (A) Preoperative reconstructed CT images of the mandibular lesion. (B) Reconstructed CT images of the mandibular lesion three months after the decompression. (C) Reconstructed CT images of the mandibular lesion ten months after the decompression (before the secondary enucleation). (D) Reconstructed CT images of the mandibular lesion eighteen months after the enucleation.

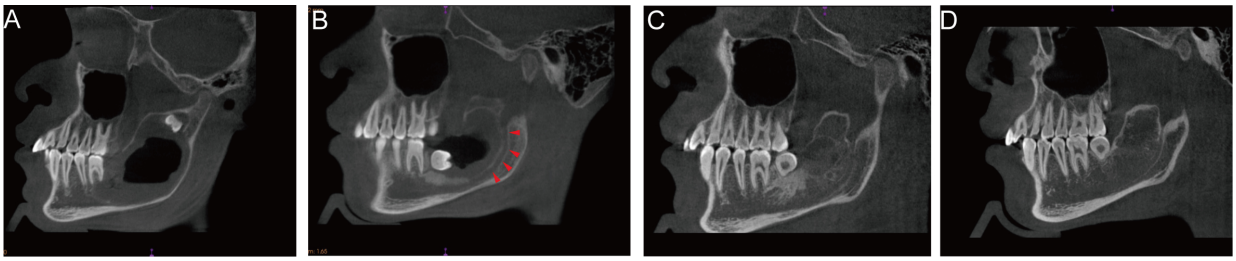

FIGURE 4. CBCT sagittal view of the left mandible. (A) Sagittal view of CT image at the first visit. (B) Sagittal view of CT image three months after the decompression showing the reappearance of mandibular canal. (C) Sagittal view of CT image ten months after the decompression (before the enucleation). (D) Sagittal view of CT image eighteen months after the enucleation.

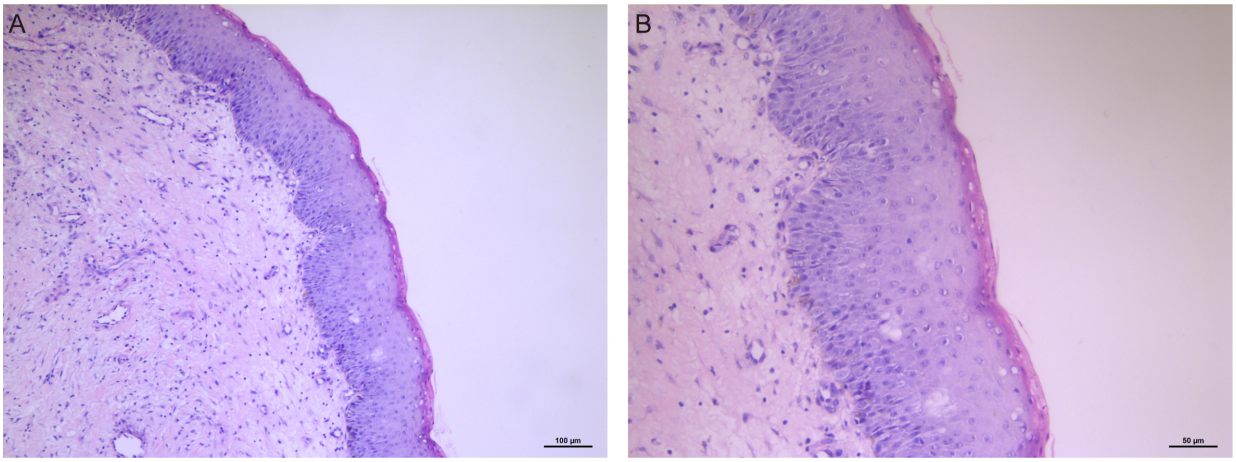

FIGURE 5. The histopathologic image of lesion. (A) The histopathologic image of the cystic wall (Hematoxylin and Eosin stain; scale bar, 100 μm). (B) The histopathologic image of the cystic wall (Hematoxylin and Eosin stain; scale bar, 50 μm).

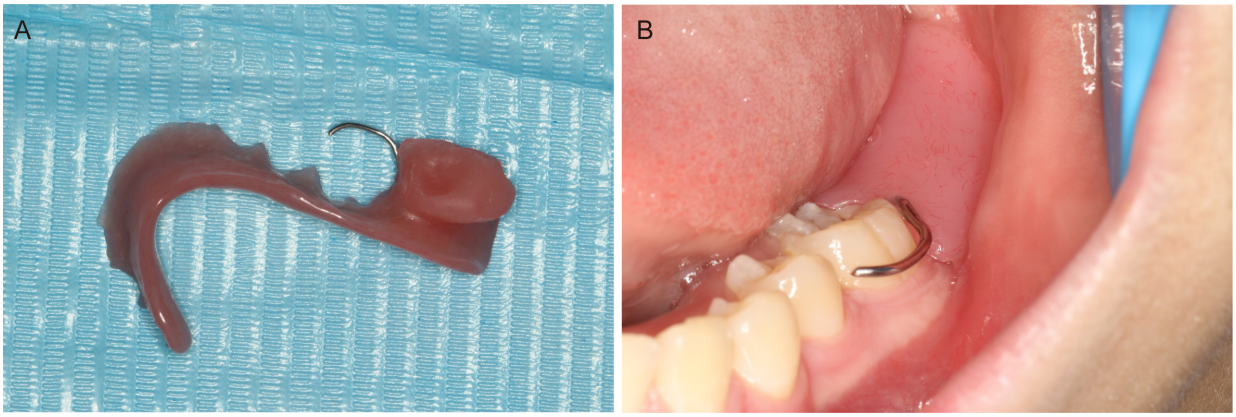

FIGURE 6. A resin obturator. (A) The obturator like a removable partial denture. (B) The obturator retained in lower dentition.

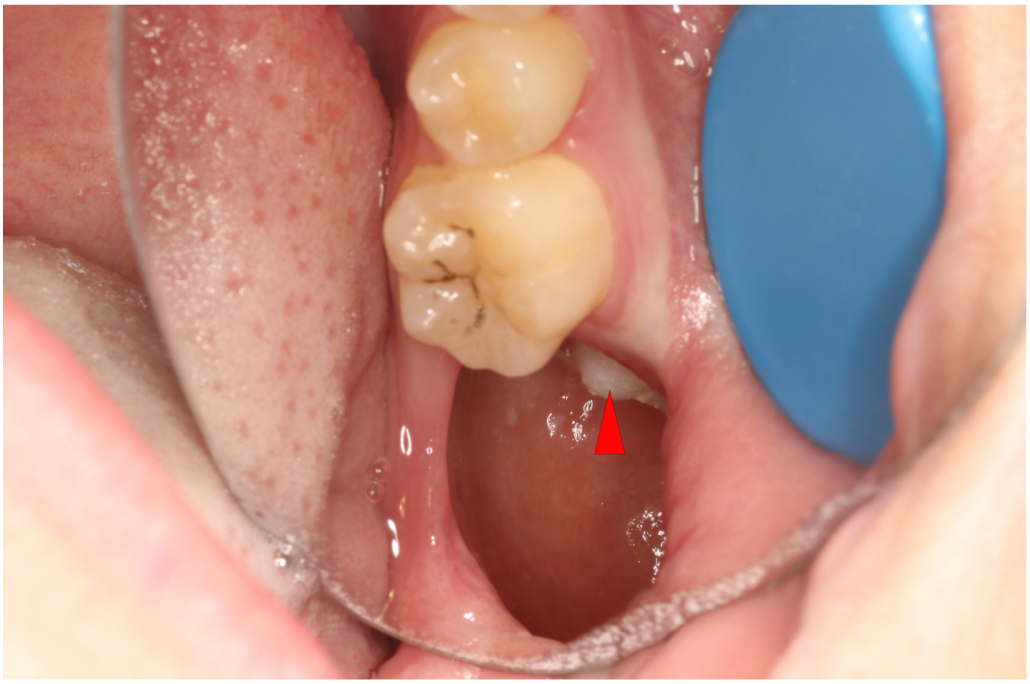

FIGURE 7. Intraoral photographs showing the tooth 37 (red arrow) at the surgical window.

Additionally, tooth 37 spontaneously erupted to the occlusal plane with a slight lingual inclination (Fig. 8), and the buccal cusp of tooth 37 contacted the central fossa of the second molar maxillary (Fig. 9). Extraoral examination showed that the facial appearance was symmetrical (Fig. 1) and the sensory function of inferior alveolar nerve recovered to normal. There are no signs of recurrence in clinical and radiographical examinations.

3.3 Analysis of tooth eruption

The patient underwent several CBCT scans during the entire treatment period. We exported the CBCT data (pre-operation, three and six months after decompression, before enucleation, and eighteen months after enucleation) in DICOM format. We imported them to Mimics Research 21.0 (Materialize, Leuven, Belgium) to reconstruct the 3D models of jawbone and teeth of the left lower jaw. Finally, the 3D models in STL format were imported in 3-Matic 11.0 (Materialize, Leuven, Belgium) to analyze the eruption of impacted second molar. After aligning multiple models in the 3-Matic, we can see the eruption pathway of impacted tooth 37 during the entire treatment process (Fig. 10A–D). After measurement, we found that the angle of impacted tooth 37 changed by approximately 70°.

4. Discussion

Enlargement of the jawbone cyst cavity is mainly affected by hyperplasia of the cyst lining epithelium, the increase of hydrostatic pressure and osmotic pressure in the cavity, and various factors related to bone resorption in cyst fluid [11, 12]. The bony wall and the involved teeth in odontogenic keratocysts are under pressure from the outward expansion of cyst, resulting in bone resorption, tooth impaction and distant displacement.

The postoperative recurrence rate of OKC is related to surgical methods. Aggressive surgical approaches might have a lower recurrence rate than conservative strategy [10]. But Mohanty *et al.* [13] suggested that conservative procedures are effective in most cases of OKC. Stanbouly *et al.* [14] concluded that 2-stage conservative treatment (decompression followed by enucleation) has a lower recurrence rate compared to single-stage conservative treatment. Hyun *et al.* [15]

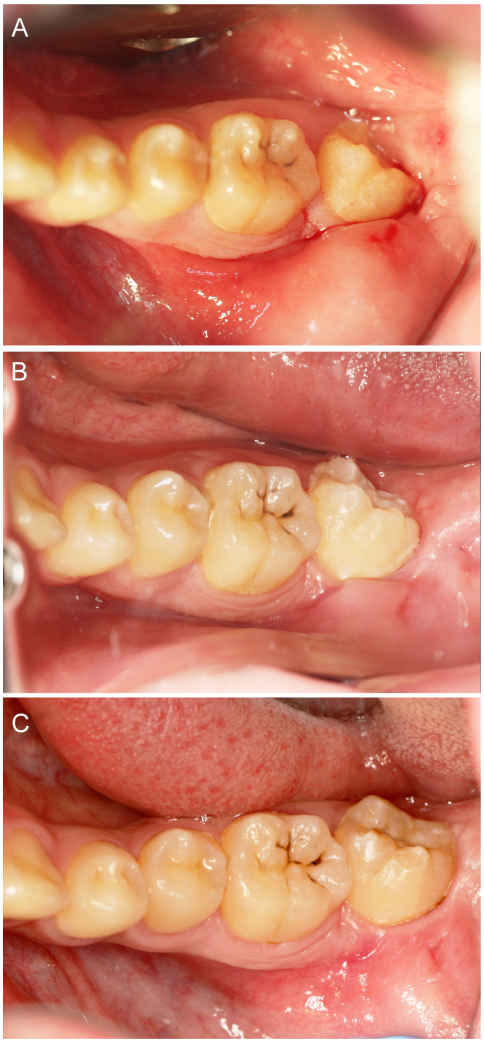

FIGURE 8. Intro-oral photography in different period. (A) Intro-oral photography ten months after the decompression (before the enucleation). (B) Intro-oral photography six months after the enucleation. (C) Intro-oral photography eighteen months after the enucleation.

FIGURE 9. Buccal view of occlusion showing the functional occlusal contact of the tooth 37.

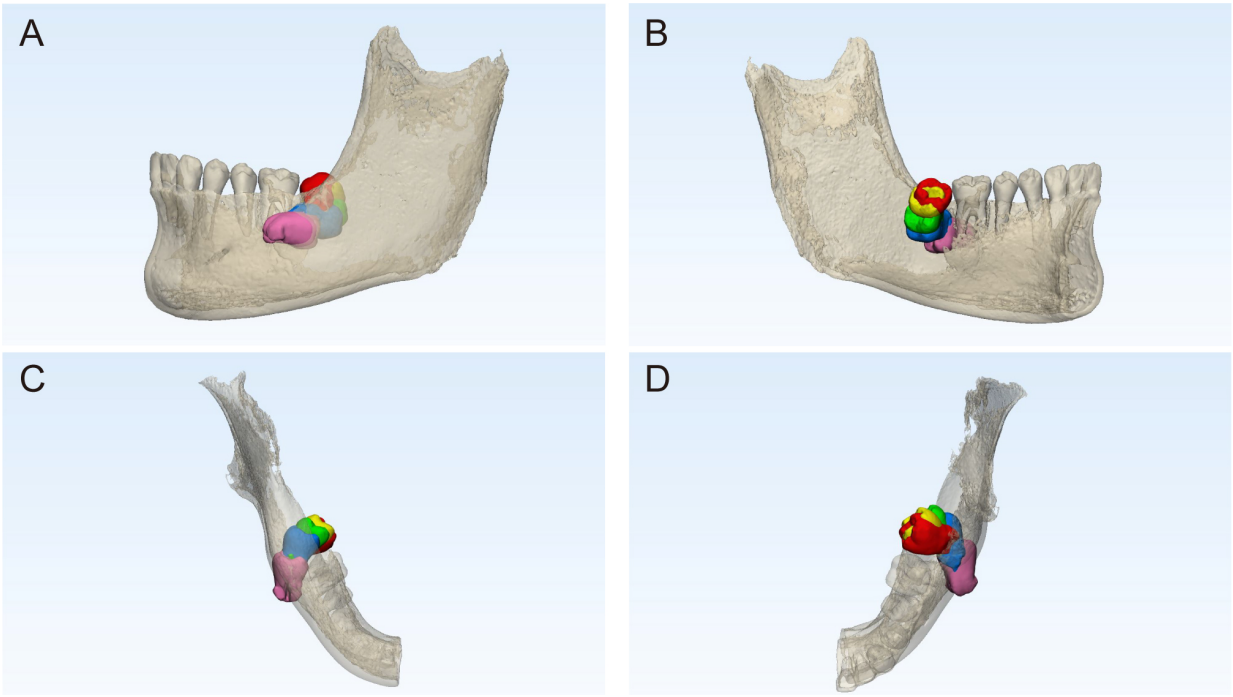

FIGURE 10. Views of the matched reconstructed models at different times showing the eruption pathway of the impacted second molar during the entire treatment period. (pink: first visit, blue: three months after decompression; green: six months after decompression; yellow: before secondary enucleation; red: eighteen months after enucleation). (A) Buccal view (B) Lingual view (C) Bottom view (D) Top view.

described the successful treatment of recurrent OKC involving erupting teeth in a 7-year-old boy. He concluded that conservative surgery should be considered first for children who suffer from OKC involves unerupted permanent teeth. Zhang *et al*. [16] reported the satisfactory effect of recompression in a case of recurrent OKC after decompression with adjunctive enucleation. ÖZKAN *et al*. [17] reported a 10-year-old boy with OKC treated by decompression, the cyst healed and the impacted teeth erupted spontaneously after treatment. In conclusion, conservative treatment (decompression and marsupialization) should be the first choice for children with OKC even if recurrent OKC.

When choosing a treatment strategy, many factors should be considered, such as cyst size, relation to important anatomical structures, and the position of involved teeth [18]. Retention of impacted teeth involved in cysts is a key point. It has been pointed out that the extraction indications for cysts involving impacted teeth are the following: impacted third molars; inverted impacted teeth; supernumerary teeth; deformed teeth [19]. However, tooth extraction should be suspended if the impacted teeth are adjacent to an important anatomical structure, such as the mandibular canal and maxillary sinus. Furthermore, premolars, canines, and other teeth that may affect the integrity of the dental arch and cause aesthetic and

psychological problems after extraction should not be removed [18, 19]. In this case, the inverted impacted third molar was suitable for extraction, and the second molar was preserved.

Eruption of normal teeth requires two conditions. First is the eruption pathway formed by bone resorption over the crown. The second is bone formation at the root. The dental follicle regulates these two procedures of bone remodeling [20]. A previous article believes that dental follicle tissue can be used as a stress sensor to mediate bone tissue remodeling, which plays a crucial role during intraosseous eruption [21]. Sarrafpour *et al*. [22] reported that cystic soft tissue may also have the same effect as dental follicle tissue. Additionally, during the extra-bony eruption stage, the position of the teeth will be affected by bone remodeling and muscle force [21].

In the present case, the pressure in the cystic cavity is released after decompression. And the eruption channel of impacted second molar was created. At the same time, concentric osteogenesis occurred in the bone wall around the cyst due to the outflow of internal fluid and the release of pressure. Therefore, impacted teeth can spontaneously erupt after decompression. Some studies show that nearly 70% of impacted teeth treated by fenestration and decompression can spontaneously erupt. Most remaining impacted teeth that do not completely erupt can also be treated with orthodontic traction to erupt in a normal position and establish stabile occlusion [19].

According to the literature, the angle of impacted teeth might change the fastest during the first three months after decompression [23, 24]. Surgically, it has been reported that if the impacted teeth cannot erupt in three months, they will eventually not spontaneously erupt to the occlusal plane [24]. However, Farshidfar *et al*. [25] reported a case of a 9-year-old patient whose right lower first premolar fully erupted spontaneously one year after decompression. In our case, a first-time radiological examination indicated that the second molar horizontally impacted the buccal side of first molar apex. Eighteen months after enucleation, tooth 37 had to stabilize occlusal function. Due to the outcomes of this patient, we believe that although the bone remodeling and tooth eruption are active within three months after decompression, the period of postoperative observation can also be appropriately extended.

5. Conclusion

We showed this case with a large odontogenic keratocyst in the left mandible. In this young patient, decompression and secondary enucleation were performed because the lesion affected the left mandibular second molar, third molar, and even the mandibular canal. These two combined surgical techniques can effectively reduce the risk of injury to the inferior alveolar nerve and seek more autogenous repair of bone defects. Eighteen months after the operation, the second molar erupted on the lingual tilt in the dentition, and it has played stabilized occlusal function. However, orthodontic treatment remains considered a necessary way for molar uprighting. The treatment effect of this case showed that decompression is an effective therapy for large cystic lesions in adolescents.

AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

AUTHOR CONTRIBUTIONS

LBZ and HJW—designed and performed the research study. JZ and JFH—analyzed the data. JFH—wrote the manuscript. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was approved and exempted by the Medical Ethics Committee of the Hospital (LCYJ2022017). Informed consent of this report was obtained from the patient and family.

ACKNOWLEDGMENT

None.

FUNDING

This work was supported by Special Fund for High-Level University Construction Talents of Guangzhou Medical University (Guangzhou, Guangdong Province, China. B185006003006).

CONFLICT OF INTEREST

The authors declare no conflict of interests.

REFERENCES

[1] Chen P, Liu B, Wei B, Yu S. The clinicopathological features and treatments of odontogenic keratocysts. American Journal of Cancer Research. 2022; 12: 3479–3485.

[2] Philipsen H. On keratosists in the jaws. Tandlaegeblader. 1956; 60: 963–980.

[3] Thompson L. World Health Organization classification of tumours: pathology and genetics of head and neck tumours. Ear, Nose & Throat Journal. 2006; 85: 74.

[4] Wright JM, Vered M. Update from the 4th edition of the World Health Organization classification of head and neck tumours: odontogenic and maxillofacial bone tumors. Head and Neck Pathology. 2017; 11: 68–77.

[5] Li TJ. The odontogenic keratocyst: a cyst, or a cystic neoplasm? Journal of Dental Research. 2011; 90: 133–142.

[6] Rajendra Santosh AB. Odontogenic cysts. Dental Clinics of North America. 2020; 64: 105–119.

[7] Deboni MCZ, Brozoski MA, Traina AA, Acay RR, Naclério-Homem MDG. Surgical management of dentigerous cyst and keratocystic odontogenic tumor in children: a conservative approach and 7-year follow-up. Journal of Applied Oral Science. 2012; 20: 282–285.

[8] de Castro MS, Caixeta CA, de Carli ML, Ribeiro Júnior NV, Miyazawa M, Pereira AAC, *et al*. Conservative surgical treatments for nonsyndromic odontogenic keratocysts: a systematic review and meta-analysis. Clinical Oral Investigations. 2018; 22: 2089–2101.

[9] Chrcanovic BR, Gomez RS. Recurrence probability for keratocystic odontogenic tumors: an analysis of 6427 cases. Journal of Cranio-Maxillofacial Surgery. 2017; 45: 244–251.

[10] Karaca C, Dere K, Er N, Aktas A, Tosun E, Koseoglu O, *et al*. Recurrence rate of odontogenic keratocyst treated by enucleation and peripheral

ostectomy: retrospective case series with up to 12 years of follow-up. Medicina Oral Patologia Oral y Cirugia Bucal. 2018; 23: e443–e448.
[11] Kwon YJ, Ko KS, So BK, Kim DH, Jang HS, Kim SH, *et al.* Effect of decompression on jaw cystic lesions based on three-dimensional volumetric analysis. Medicina. 2020; 56: 602.
[12] Sá MC, de Matos FR, Conceição TS, Leitão AC, Freitas RA. Immunoexpression of tumour necrosis factor-α, interleukin-1α and interleukin-10 on odontogenic cysts and tumours. International Endodontic Journal. 2017; 50: 437–445.
[13] Mohanty S, Dabas J, Verma A, Gupta S, Urs AB, Hemavathy S. Surgical management of the odontogenic keratocyst: a 20-year experience. International Journal of Oral and Maxillofacial Surgery. 2021; 50: 1168–1176.
[14] Stanbouly D, Lee KC, Fazzolari JR, Philipone E. Decompression of odontogenic keratocysts before enucleation reduces the rate of recurrence. Journal of Craniofacial Surgery. 2022; 33: 1806–1808.
[15] Hyun H, Hong S, Kim J. Recurrent keratocystic odontogenic tumor in the mandible: a case report and literature review. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2009; 108: e7–e10.
[16] Zhang Q, Li W, Han F, Huang X, Yang X. Recurrent keratocystic odontogenic tumor after effective decompression. Journal of Craniofacial Surgery. 2016; 27: e490–e491.
[17] Özkan E, Özmen B. The treatment of different odontogenic cysts in children only using acrylic appliance-guided decompression: a long-term clinical study. Anatolian Clinic the Journal of Medical Sciences. 2023; 28: 15–23.
[18] Motamedi MHK, Talesh KT. Management of extensive dentigerous cysts. British Dental Journal. 2005; 198: 203–206.
[19] Zhao Y, Liu B, Zhao Y. Controversies regarding the management of teeth associated with cystic lesions of the jaws. The Chinese Journal of Dental Research. 2019; 22: 81–92.
[20] Wise G. Cellular and molecular basis of tooth eruption. Orthodontics & Craniofacial Research. 2009; 12: 67–73.
[21] Sarrafpour B, Swain M, Li Q, Zoellner H. Tooth eruption results from bone remodelling driven by bite forces sensed by soft tissue dental follicles: a finite element analysis. PLOS ONE. 2013; 8: e58803.
[22] Sarrafpour B, El-Bacha C, Li Q, Zoellner H. Roles of functional strain and capsule compression on mandibular cyst expansion and cortication. Archives of Oral Biology. 2019; 98: 1–8.
[23] Park JH, Kwak E, You KS, Jung Y, Jung H. Volume change pattern of decompression of mandibular odontogenic keratocyst. Maxillofacial Plastic and Reconstructive Surgery. 2019; 41: 2.
[24] Nahajowski M, Hnitecka S, Antoszewska-Smith J, Rumin K, Dubowik M, Sarul M. Factors influencing an eruption of teeth associated with a dentigerous cyst: a systematic review and meta-analysis. BMC Oral Health. 2021; 21: 180.
[25] Farshidfar N, Agharokh M, Daneste H. Eruption of the permanent first premolar associated with a mandibular keratocystic odontogenic tumor after marsupialization in a 9-year-old boy: a case report with 2 years of follow-up. Journal of Dentistry. 2021; 22: 144–148.

How to cite this article: Jianfeng He, Huijing Wang, Jun Zeng, Libin Zhou. Large mandibular odontogenic keratocyst treated by decompression and secondary enucleation: a case report. Journal of Clinical Pediatric Dentistry. 2024; 48(6): 213-220. doi: 10.22514/jocpd.2024.142.