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CASE REPORT

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Comprehensive treatment of non-syndromic multiple odontogenic keratocyst and impacted teeth in a pediatric patient—a case report and literature review

Yuhan Xu[1,†], Yanjia Hu[1,†], Zheng Li[2], Fang Yi[1], Yanhao Chu[1,*]

[1]Xiangya Stomatological Hospital and Xiangya School of Stomatology, Central South University; Hunan Key Laboratory of Oral Health Research, Hunan Engineering Research Center for Oral Digital Intelligence and Personalized Medicine, 410008 Changsha, Hunan, China
[2]The Affiliated Stomatological Hospital of Soochow University, Suzhou Stomatological Hospital, 215000 Suzhou, Jiangsu, China

***Correspondence**
chuyh5@mail3.sysu.edu.cn
(Yanhao Chu)

† These authors contributed equally.

Abstract

Background: The odontogenic keratocyst (OKC) is a developmental cyst that is considered benign but displays localized aggressiveness. Most patients with multiple OKCs present with syndromes, and non-syndromic patients with multiple OKCs are less common. Furthermore, there is a lack of information on the management of non-systemic multiple OKCs and the associated problems. Further investigation is needed to explore the efficacy of comprehensive treatment for OKC patients with large cysts and impacted teeth. **Case**: In this case, we presented a case of a 10-year-old patient with non-syndromic multiple OKCs and impacted teeth. A panoramic radiograph revealed two radiolucent areas in the mandible. To remove the lesions and promote the eruption of impacted teeth, a comprehensive therapy involving marsupialization, enucleation, obturator and orthodontic alignment was employed. Asymmetry of the mandible, alveolar bone and occlusal plane, in addition to the presence of a deep overbite, were observed during the course of treatment. **Conclusions**: This case underscores the effectiveness of comprehensive OKC treatment and highlights the potential impact of cysts and impacted teeth on jaw and dentition symmetry.

Keywords

Odontogenic keratocyst; Keratocystic odontogenic tumor; Impacted tooth; Marsupialization; Orthodontic treatment

1. Introduction

The odontogenic keratocyst (OKC), was previously renamed keratocystic odontogenic tumor (KCOT) from 2005 to 2017 [1, 2]. In the 4th edition of the World Health Organization (WHO) classification of head and neck tumors, this disease was reclassified as odontogenic keratocyst [3–5]. However, controversy remains regarding the reclassification, as some scholars maintain that OKC should be considered tumors consistent with the previous classification [6].

OKC is an intraosseous cystic lesion that originates from the dental lamina or primordial odontogenic epithelium and has a characteristic uniform lining of parakeratinised squamous epithelium [7–9]. Based on the number of lesions in the jaws, OKC can be categorized into single and multiple types. OKC can also be classified as systemic or non-systemic depending on the presence of other systemic pathologies. Most patients with multiple OKC have other symptoms, including cutaneous palmar or plantar pits, calcifications of the falx cerebri, and fused or markedly splayed ribs [10], which are also known as syndromic odontogenic keratocyst, Nevoid basal-cell carcinoma syndrome (NBCCS), or Gorlin-Goltz syndrome. Conversely, non-syndromic multiple types are relatively rare compared to syndromic multiple types [11].

Controversies persist regarding the optimal treatment strategies for odontogenic keratocysts, with both aggressive and conservative treatments being used in clinical practice. Given the recurrent nature of the disease, aggressive treatments including surgical enucleation and curettage are employed for some patients. However, conservative treatment options can be effective in reducing morbidity and minimizing the psychosocial impact on young patients. Marsupialization is a conservative and effective technique for reducing lesion size and promoting bone regeneration. Multiple and systemic features are important traits that indicate a higher risk of OKC recurrence, and in such cases, more aggressive surgical resection may be necessary. In contrast, for non-systemic OKC patients, comprehensive conservative treatment is recommended to minimize surgical damage and limit the impact on the patient's jaws, dentition, occlusion and esthetics. Moreover, few papers have focused on young, non-syndromic OKC patients with multiple jaw lesions, highlighting the need for further investigation into the effectiveness of such treatments.

Impacted teeth are frequently involved in odontogenic tumor lesions, which are usually extracted during surgery [12]. In conservative treatments, impacted teeth may be retained, but their spontaneous eruption may be hindered due to a lack of sufficient space or abnormal orientation, leading to crowding

in the dentition. This aspect is often overlooked in conservative treatments. In this report, we presented a rare case of non-systemic multiple OKC with impacted teeth, and describe a comprehensive and conservative treatment approach that includes marsupialization, enucleation, obturator and orthodontic alignment. During the course of treatment, we meticulously documented the changes in the patient's jaw lesions, as well as the alterations in the impacted teeth after orthodontic traction, and the challenges encountered during the treatment.

2. Case report

A 10-year-old child was referred to the Xiangya Stomatological Hospital in November 2015 with a chief complaint of mandibular cysts found in another hospital. On facial examination, a slight asymmetry was noted in the lower region of the patient's face. No other abnormalities were detected, including cutaneous and rib anomalies. Clinical examination revealed no obvious jaw swelling in the vestibular area of the mandible. Panoramic radiograph revealed two large radiolucent lesions in the right mandibular ramus and mandibular bone extending from teeth 83 to 37, with six impacted permanent teeth within the cysts, including 33, 34, 35, 43, 47 and 48 (Fig. 1). The lesions showed well-defined translucent regions with rounded or scalloped margins that are well-demarcated. Additionally, no root or inferior dental canal resorption was observed in the mandibular lesion area.

Based on the results of clinical physical and radiographic examinations, the patient was diagnosed with multiple cysts in the mandible with suspected odontogenic keratocysts. Due to the young age of the patient and the extent of the cysts in the mandible, a combination of treatments was used to minimize the impact on the patient's jaw, occlusion and appearance. Marsupialization was the initial choice to reduce the jaw lesion, and protect the integrity of the mandible, in preparation for the complete removal of the lesions by subsequent surgery. To achieve this, two gingival flaps were elevated in the buccal area of teeth 75 and 47. An opening window was created in the buccal alveolar bone to expose the cysts, which were then irrigated with saline solution. Biopsy samples were obtained for histopathological examination. Alginate impressions were taken one week after the surgery to fabricate an acrylic resin obturator. The use of obturators was recommended to the patient to maintain the surgical opening during the healing process, prevent food debris from entering the lesion, maintain oral hygiene in the operative area, and serve as a space retainer. Subsequently, the patient was instructed to rinse the cavities with the saline solution daily, which can help to relieve the pressure and promote bone formation.

The histopathological diagnosis of both lesions was confirmed as odontogenic keratocyst, as demonstrated in Fig. 1. Hematoxylin and eosin (HE) staining sections showed a fibrous tissue cyst wall with a thin stratified squamous epithelium lining in the cysts without inflammation. As shown in Fig. 1C,E, the epithelium lining displayed even thickness and parakeratinisation. Moreover, there was a lack of rete ridges in the epithelium lining. The basal layer of the epithelium lining was composed of columnar cells arranged in a palisaded fashion.

Postoperative clinical evaluations were performed at monthly intervals for the first half year and every two months thereafter, while panoramic imaging reviews were conducted every three to six months. The obturators' size was progressively reduced beginning in the second month after the marsupialization as the opening windows began to narrow. During follow-up visits, the obturators were adjusted to achieve optimal contact and ensure that treatment was efficacious.

The radiographs revealed a significant decrease in the size of the mandibular bone cysts, along with progressive new bone formation in the two lesions of the mandible (Fig. 2). Three months post-surgery, the panoramic radiograph displayed a marked decrease in the size of the two translucent areas (Fig. 2A). The patient was advised to continue wearing the obturators and performing saline flushing to further decrease the cyst size. As treatment progressed, the size of two OKCs continued to decrease at 6 months post-surgery (Fig. 2B). Nine months after the surgery, due to the reduction in cyst pressure, the primary teeth 83 were lost, and the permanent teeth 43 erupted spontaneously (Fig. 2C). After fifteen months, with the decompression treatment, the cysts' size decreased significantly, and primary teeth in the left mandible, 73, 74 and 75 were lost, while permanent tooth 45 erupted automatically, but 43 and 44 remained impacted (Fig. 2D).

After a thorough examination, it was observed that impacted teeth 33 and 34 were malpositioned and oriented towards abnormal eruption pathways, with inadequate space in the dentition (Fig. 3A). In addition, tooth 34 was in close proximity to the root of tooth 35. Hence, the decision was made to implement orthodontic treatment to facilitate the proper alignment and eruption of teeth 33 and 34. When the lesion at tooth 48 was small enough, the enucleation procedure and the extraction of tooth 48 were operated to remove the lesion completely (Fig. 3B). Orthodontic brackets and nickel-titanium wires were utilized to align other teeth first, followed by the placement of coil springs to erect tilted teeth and create space for the impacted teeth. When primary teeth 74 and 75 were lost, and the lesion in the left mandible was minimal enough, the use of obturators was discontinued, and enucleation was performed in the left mandible. An orthodontic button and chains were bonded to the two impacted teeth during the enucleation procedure (Fig. 3C). Subsequently, elastic traction was employed between the impacted teeth and the stainless steel lower archwire to promote their eruption (Fig. 3D,E). After the eruption of the impacted teeth, intermaxillary elastic traction was used to bring the teeth closer to the occlusal plane. Once the majority of the labial surface was exposed, orthodontic buttons were removed, brackets were bonded to the impacted teeth, and high-elasticity nickel-titanium archwires were employed to align the teeth. The impacted teeth 33, 34 were positioned properly into the dentition. During orthodontic treatment, the patient was noted to have a deep overbite of the anterior teeth, which proved challenging to correct (Fig. 3E). Efforts were made to correct the deep overbite and the tilt of the mandibular occlusal plane in orthodontic treatment. But it's a pity that the patient was unable to complete the full course of orthodontic treatment as he entered a distant college to study, and could not return for regular check-ups, with poor oral hygiene. Both the

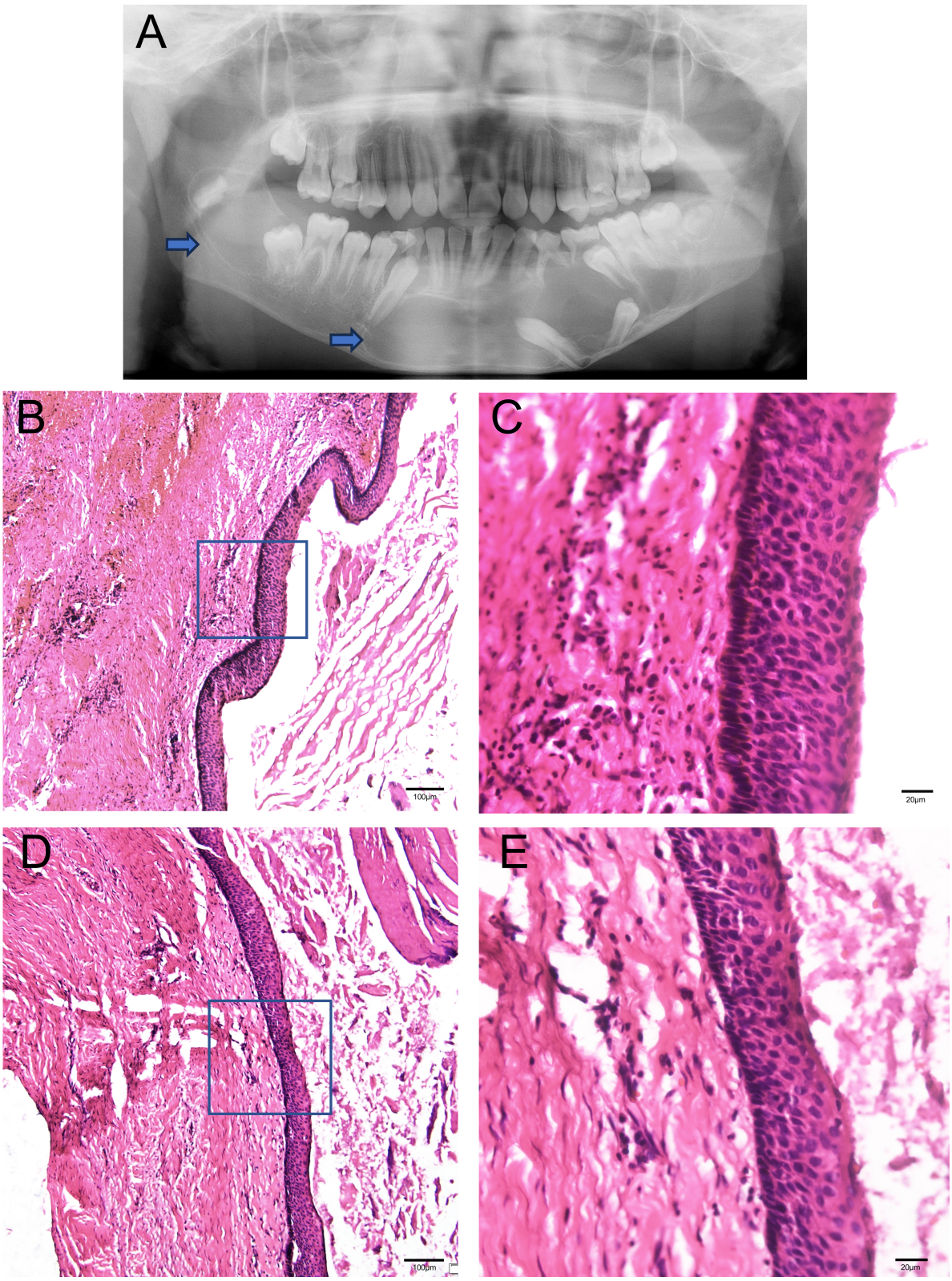

FIGURE 1. Initial panoramic film and hematoxylin and eosin (HE) stain images. (A) Two large radiolucent lesions at the first visit (one in the right mandibular ramus, another one from the tooth 83 to 37). (B) HE staining of the lesion in the left mandibular bone. (C) Higher magnification HE staining in the blue box in Fig. 1B. (D) HE staining of the lesion in the right mandibular bone. (E) Higher magnification HE staining in the blue box in Fig. 1D. Scale bar: 100 μm in Fig. 1B,D; 20 μm in Fig. 1C,E (Blue Arrows in Fig. 1A: the area of the lesions).

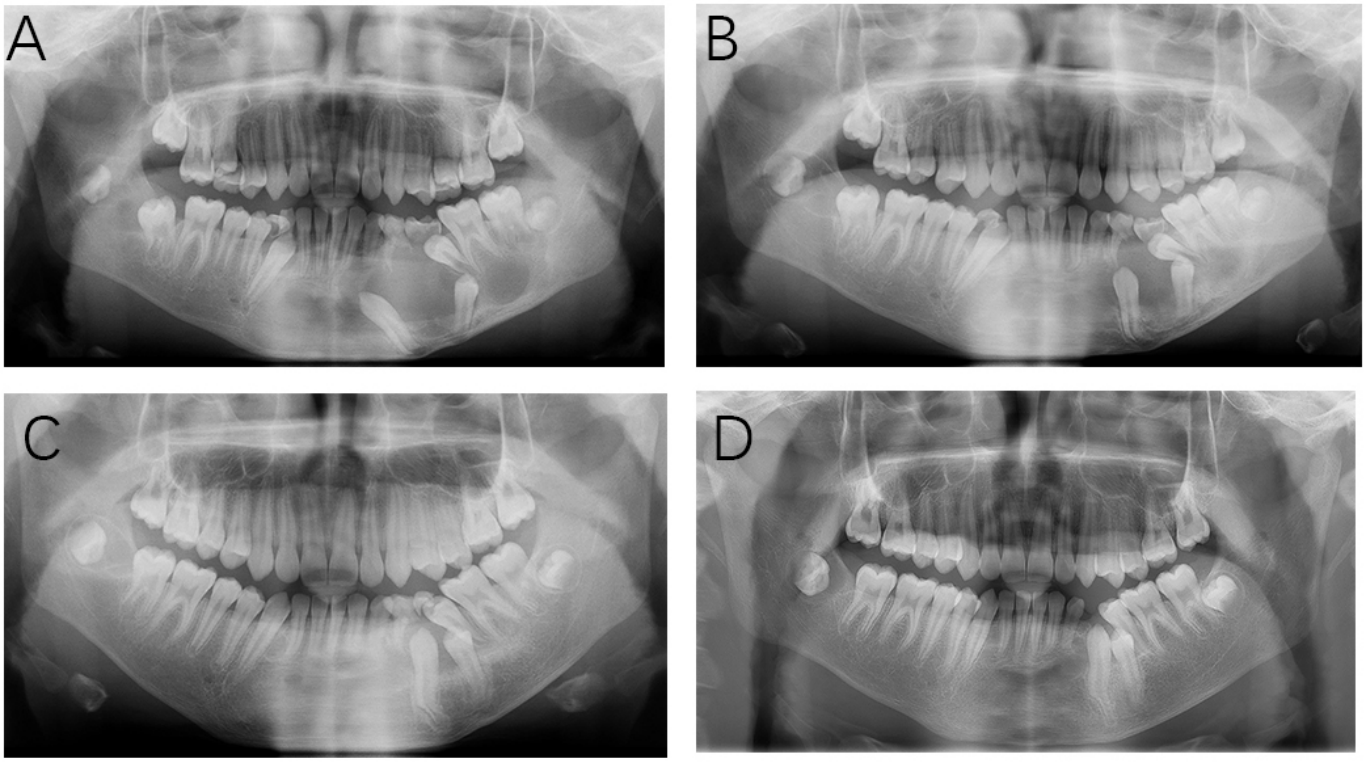

FIGURE 2. Panoramic radiographs during treatment. (A–D) After the marsupialization, the panoramic radiograph demonstrated the new bone formation and the reduction of lesions.

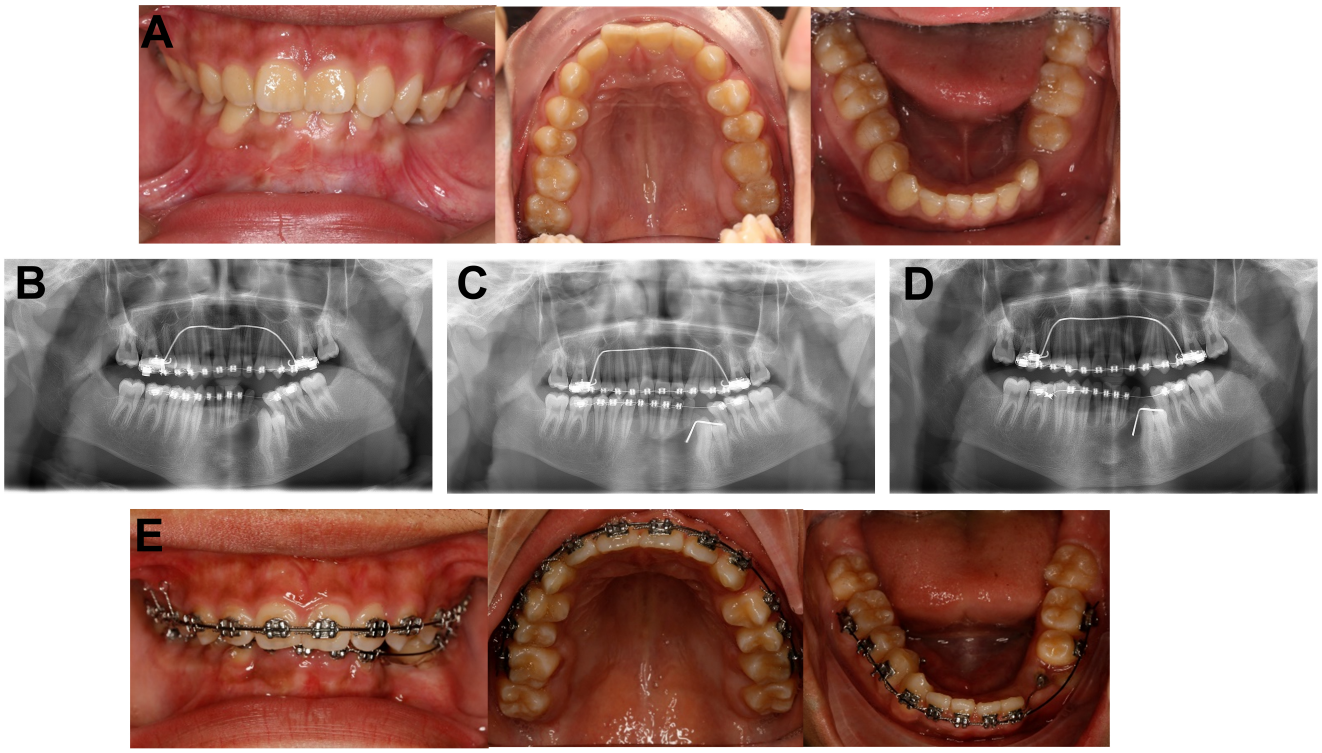

FIGURE 3. Intraoral photograph and panoramic radiographs during orthodontic treatment. (A) The photo before starting the orthodontic treatment. (B) After the enucleation of the lesion at tooth 48, the orthodontic treatment was used to open space. (C,D) Continuing decompression until the lesion in the left mandible was small enough to remove entirely by enucleation. (E) The photo during the orthodontic treatment.

patient and his family were satisfied with the outcome of the treatment and requested for the end of the treatment. As shown in Fig. 4, the impacted teeth were aligned, and the deep overbite was improved. The patient was debonded early and instructed to wear a Harley retainer throughout the day. No low-density lesions were observed on the panoramic radiograph one year after the orthodontic treatment was finished (Fig. 4).

3. Discussion

OKC has been recognized as one of the most controversial cysts in terms of both its diagnosis and treatment due to its unique peripheral aggressive features and high recurrence rate. Studies have shown that OKC expresses more proliferation markers, such as Ki67 and Proliferating Cell Nuclear Antigen (PCNA), and exhibits genetic alterations in tumor suppressor genes such as *TP53* and Patched 1 (*PTCH1*), which are considered to be associated with its aggressive features [13, 14]. Additionally, other developmental cysts have shown *PTCH* changes, such as loss of heterozygosity on the 9q22.3 region [15]. Despite this evidence, there is currently insufficient evidence to confirm the neoplastic origin of OKC. Therefore, the term keratocystic odontogenic tumor (KCOT) was removed from the WHO's new 4th edition classification.

The clinical diagnosis of OKC is typically based on radiology, but a histopathological examination is required for a definitive diagnosis. OKC accounts for approximately 10% of odontogenic cysts and is more frequently located in the mandible, with a prevalence that is twice as high in the mandible than in the maxilla [8, 16]. In particular, OKC tends to occurs in the posterior sextants, mandibular angle or mandibular ramus. On panoramic radiography, OKC appears as a well-defined unilocular or multilocular radiolucency bounded by corticated margins [17, 18]. Approximately 30% of OKCs involve at least one unerupted tooth, with the third molar being the most commonly affected tooth [18]. Compared with other odontogenic cysts, mandibular OKC exhibits proximodistal expansion along the long axis of the mandible with minimal buccolingual expansion. Additionally, root resorption in the involved teeth is uncommon in OKCs but frequently observed in aggressive odontogenic tumors such as ameloblastoma [18]. Dentigerous cysts, ameloblastoma and radicular cysts are commonly considered the most frequent odontogenic lesions in the differential diagnosis of OKCs on panoramic radiography. OKC can be classified into single and multiple types according to the number of lesions, with multiple OKCs usually considered a diagnostic criterion of nevoid basal cell carcinoma syndrome. Therefore, when multiple OKC are observed, attention should also be paid to the presence of other systemic symptoms. In rare cases, multiple OKCs can be present without any other systemic symptoms [19]. The presence of non-systemic multiple OKCs may be attributed to the multifocal nature of the lesion rather than a specific syndrome [20, 21].

The treatment protocols for OKC remain controversial. Various treatment options are available, including enucleation alone or in combination with adjunctive therapies such as peripheral ostectomy, Carnoy's solution, liquid nitrogen cryotherapy, marsupialization with or without subsequent surgery, and resection [22–24]. Enucleation alone has been shown to have a high recurrence rate, while resection, including segmental resection or marginal resection, has been reported to have the lowest recurrence rate at 1.85% [2]. A meta-analysis showed that currently for OKC, analysis of the recurrence rate of OKC does not allow to conclude which clinical treatment is the best option [25]. However,

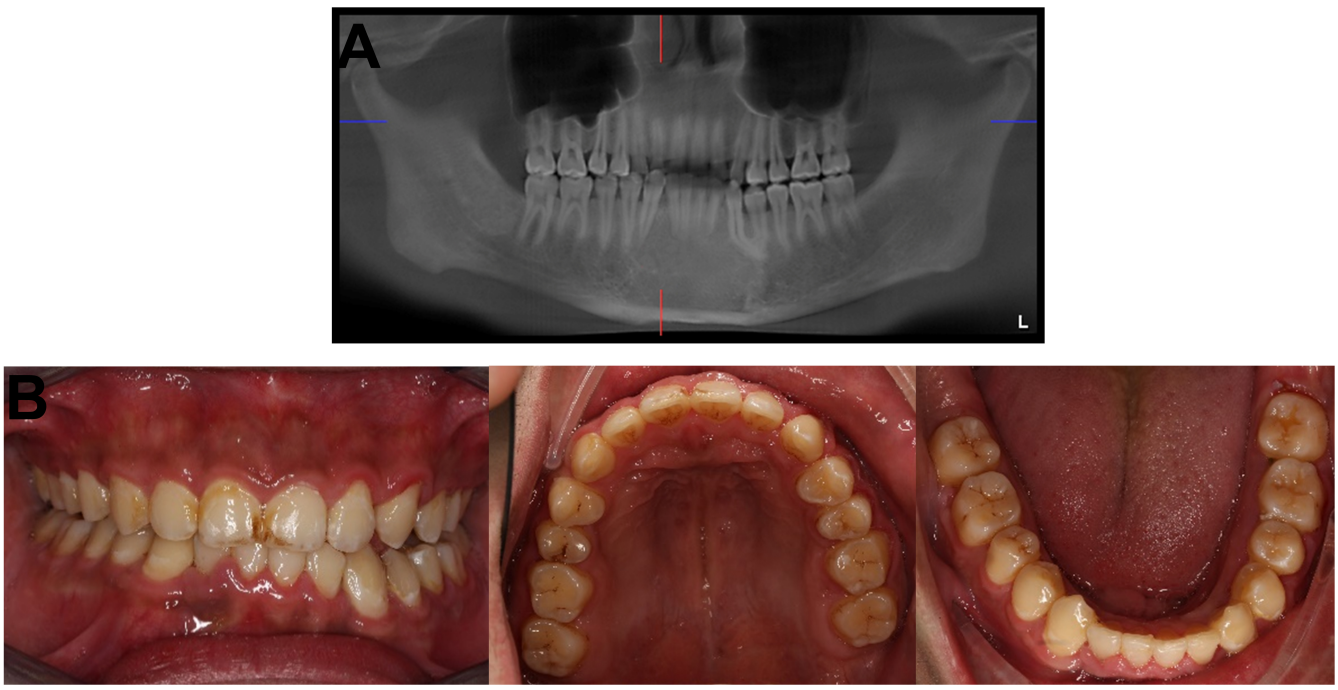

FIGURE 4. Intraoral photograph and panoramic radiographs after the treatment. (A) After the orthodontic treatment, the impacted teeth were aligned. (B) The photo after the comprehensive treatment.

resecting a large OKC lesion in a young patient may result in a series of complications, such as facial deformities, bone defects, bone fractures and tooth loss [26]. In this regard, marsupialization or decompression has been considered an optimal option to promote new bone formation, limit the invasiveness of secondary surgery and protect important anatomical structures [27]. However, marsupialization also has some drawbacks, such as long treatment duration, the need for patient cooperation, and an increased risk of infection due to the opening window [28]. Additionally, some researchers argued that marsupialization may leave small remnants behind due to the decrease in size of the lesion and the cyst lining being carried forward [20]. Therefore, marsupialization is often used as an initial treatment for extensive OKCs to reduce the extent of the cyst. An obturator treatment can be taken into consideration to prevent food accumulation or closure of the fistula. Subsequently, a second stage of enucleation is usually utilized to remove any residual cyst. However, there are some limitations associated with the use of obturators. The patient needs to be instructed properly, and a careful recall is recommended. The obturator may also need to be adjusted frequently to maintain an appropriate fit, which can be time-consuming and costly. Moreover, the obturator can be uncomfortable and difficult to wear. Therefore, it is essential to avoid interference with occlusion and mastication as much as possible.

During the course of treatment for OKC, the patient exhibited features that were challenging to fully correct, including a tilted occlusal plane, asymmetrical alveolar bone, and deep overbite. These features were likely attributed to the lack of vertical growth of alveolar bone caused by the lesions, resulting in mandibular asymmetry and deep overbite. Enucleation or resection surgeries for OKC often involve the extraction of impacted teeth, leading to tooth loss in the dentition. Furthermore, tooth loss-induced occlusal hypofunction during the growth period can further impair vertical alveolar bone growth [29]. The combination of marsupialization and orthodontic treatment can be effective in preserving and aligning impacted teeth, reducing alveolar bone defects, and decreasing the need for implant surgery. Moreover, due to the eruption of the impacted teeth, there will be new bone formation in the eruption process, and the contact of erupting impacted teeth with opposing teeth to establish an occlusal relationship can promote the restoration of occlusal function and further promote the formation of new bone [30]. However, the patient in this case had insufficient mandibular alveolar bone height due to substantial bone loss in the left mandible prior to treatment, resulting in a tilted occlusal plane. Moreover, for this patient, due to the occlusal tilt, a series of compensatory changes including extrusion of the opposing maxillary posterior teeth, mandibular shift, and torque compensation of posterior teeth were formed to maintain masticatory function. As a result, it's difficult to improve the deep overbite during orthodontic treatment. In the late stages of treatment, attempts were made to correct the deep overbite and the tilt of the occlusal plane, but the patient urged to end the treatment early, leading to an imperfect overbite. This case highlights the importance of paying more attention to mandibular asymmetry, occlusal plane tilt, and the buccal-lingual torque of posterior teeth in the orthodontic treatment of OKC or other odontogenic cysts. Moreover, intermaxillary elastic traction for impacted teeth should be used with caution. If necessary, implant anchorage can be used for elastic traction. The findings of this case underscore the importance of identifying the potential impact of odontogenic cysts and jaw tumors on occlusal cant and abnormal occlusal relationships during orthodontic treatment. These findings may be particularly important for clinicians seeking to optimize treatment outcomes and prevent potential complications in this patient population.

The presence of satellite cysts, budding of cyst linings, or remnants of cyst epithelium remaining after enucleation can contribute to the recurrence of OKC [31]. OKC of multiple types and NBCCS have a high recurrence rate due to the development of new lesions, with some cases recurring even after 14–21 years of the first surgery [32]. Therefore, regular oral and maxillofacial physical examinations and panoramic radiographs should be recommended for patients with multiple OKC after initial therapy.

4. Conclusions

This report presents a rare case of non-systemic multiple OKC with impacted teeth, which was treated with a combination of marsupialization, obturator, enucleation and orthodontic alignment. The comprehensive treatment option was effective in reducing jaw damage and preserving the integrity of the dentition. Moreover, this case highlights the importance of early attention to the impact of cysts and impacted teeth on alveolar bone development and dentition symmetry during treatment.

ABBREVIATIONS

OKC, odontogenic keratocyst; KCOT, keratocystic odontogenic tumor; NBCCS, Nevoid basal-cell carcinoma syndrome.

AVAILABILITY OF DATA AND MATERIALS

The data are contained within this article.

AUTHOR CONTRIBUTIONS

YHX and YHC—drafted the initial manuscript. YJH and YHC—revised the manuscript. FY and ZL—collected the files and processed the pictures. All the authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was approved by the ethics committee of The Xiangya Stomatological Hospital, Central South University (No. 20160014). For all research involving human subjects, informed consent to participate in the study was obtained from the legal guardian of this patient.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

REFERENCES

[1] Pogrel MA. The keratocystic odontogenic tumor. Oral and Maxillofacial Surgery Clinics of North America. 2013; 25: 21–30.

[2] Johnson NR, Batstone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology. 2013; 116: e271–e276.

[3] Speight PM, Takata T. New tumour entities in the 4th edition of the World Health Organization classification of head and neck tumours: odontogenic and maxillofacial bone tumours. Virchows Archiv. 2018; 472: 331–339.

[4] Zhang R, Yang J, Zhang J, Hong Y, Xie X, Li T. Should the solid variant of odontogenic keratocyst and keratoameloblastoma be classified as the same entity? A clinicopathological analysis of nine cases and a review of the literature. Pathology. 2021; 53: 478–486.

[5] Slootweg PJ, El-Naggar AK. World Health Organization 4th edition of head and neck tumor classification: insight into the consequential modifications. Virchows Archiv. 2018; 472: 311–313.

[6] 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.

[7] Nayak M, Singh A, Singhvi A, Sharma R. Odontogenic keratocyst: what is in the name? Journal of Natural Science, Biology and Medicine. 2013; 4: 282.

[8] Bilodeau EA, Collins BM. Odontogenic cysts and neoplasms. Surgical Pathology Clinics. 2017; 10: 177–222.

[9] Kolář Z, Geierová M, Bouchal J, Pazdera J, Zbořil V, Tvrdý P. Immunohistochemical analysis of the biological potential of odontogenic keratocysts. Journal of Oral Pathology & Medicine. 2006; 35: 75–80.

[10] Manfredi M, Vescovi P, Bonanini M, Porter S. Nevoid basal cell carcinoma syndrome: a review of the literature. International Journal of Oral and Maxillofacial Surgery. 2004; 33: 117–124.

[11] Hammannavar R, Holikatti K, Bassappa S, Shinde N, Reddy M, Chidambaram YS. Multiple, multifocal odontogenic keratocysts in non-syndrome patient: a case-report. Oral Health and Dental Management. 2014; 13: 189–193.

[12] Erdur EA, Ileri Z, Ugurluoglu C, Cakir M, Dolanmaz D. Eruption of an impacted canine in an adenomatid odontogenic tumor treated with combined orthodontic and surgical therapy. American Journal of Orthodontics and Dentofacial Orthopedics. 2016; 149: 923–927.

[13] Gomes CC, Diniz MG, Gomez RS. Review of the molecular pathogenesis of the odontogenic keratocyst. Oral Oncology. 2009; 45: 1011–1014.

[14] Geleţu GL, Burlacu A, Baciu ER, Diaconu-Popa D, Murariu A, Foia LG, *et al*. Various surgical interventions in treating odontogenic keratocyst: a radiological case report. Healthcare. 2023; 11: 416.

[15] Levanat S, Pavelić B, Crnić I, Orešković S, Manojlović S. Involvement of PTCH gene in various noninflammatory cysts. Journal of Molecular Medicine. 2000; 78: 140–146.

[16] Harmon M, Arrigan M, Toner M, O'Keeffe SA. A radiological approach to benign and malignant lesions of the mandible. Clinical Radiology. 2015; 70: 335–350.

[17] Pogrel MA. The keratocystic odontogenic tumour (KCOT)—an odyssey. International Journal of Oral and Maxillofacial Surgery. 2015; 44: 1565–1568.

[18] Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, *et al*. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights into Imaging. 2018; 9: 883–897.

[19] Kargahi N, Kalantari M. Non-syndromic multiple odontogenic keratocyst: a case report. Journal of Dentistry. 2013; 14: 151–154.

[20] Al-Moraissi EA, Pogrel MA, Ellis E. Enucleation with or without adjuvant therapy versus marsupialization with or without secondary enucleation in the treatment of keratocystic odontogenic tumors: a systematic review and meta-analysis. Journal of Cranio-Maxillofacial Surgery. 2016; 44: 1395–1403.

[21] Sundaragiri K, Saxena S, Sankhla B, Bhargava A. Non syndromic synchronous multiple odontogenic keratocysts in a western Indian population: a series of four cases. Journal of Clinical and Experimental Dentistry. 2018; 10: e831–e836.

[22] Kim S, Oh M, Seo Y, Kim J, Nam S, Lim S. Conservative treatment of multiple keratocystic odontogenic tumors in a young patient with nevoid basal cell carcinoma syndrome by decompression: a 7-year follow-up study. Journal of Clinical Pediatric Dentistry. 2017; 41: 300–304.

[23] Demirkol M, Keskinruzgar A, Ege B, Oksayan R, Erkilic S, Aras MH. Complete eruption of a deeply impacted tooth in a recurrent keratocystic odontogenic tumor with orthodontic occlusal alignment. Journal of Craniofacial Surgery. 2015; 26: 944–945.

[24] Al-Moraissi EA, Dahan AA, Alwadeai MS, Oginni FO, Al-Jamali JM, Alkhutari AS, *et al*. What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: a large systematic review and meta-analysis. Journal of Cranio-Maxillofacial Surgery. 2017; 45: 131–144.

[25] Antonoglou GN, Sándor GK, Koidou VP, Papageorgiou SN. Non-syndromic and syndromic keratocystic odontogenic tumors: systematic review and meta-analysis of recurrences. Journal of Cranio-Maxillofacial Surgery. 2014; 42: e364–e371.

[26] Berretta LM, Melo G, Mello FW, Lizio G, Rivero ERC. Effectiveness of marsupialisation and decompression on the reduction of cystic jaw lesions: a systematic review. British Journal of Oral and Maxillofacial Surgery. 2021; 59: E17–E42.

[27] Pei J, Zhao S, Chen H, Wang J. Management of radicular cyst associated with primary teeth using decompression: a retrospective study. BMC Oral Health. 2022; 22: 560.

[28] Nakamura N, Mitsuyasu T, Mitsuyasu Y, Taketomi T, Higuchi Y, Ohishi M. Marsupialization for odontogenic keratocysts: long-term follow-up analysis of the effects and changes in growth characteristics. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2002; 94: 543–553.

[29] Hsieh YD, Devlin H, Roberts C. Early alveolar ridge osteogenesis following tooth extraction in the rat. Archives of Oral Biology. 1994; 39: 425–428.

[30] Shimomoto Y, Chung CJ, Iwasaki-Hayashi Y, Muramoto T, Soma K. Effects of occlusal stimuli on alveolar/jaw bone formation. Journal of Dental Research. 2007; 86: 47–51.

[31] Miyamoto S, Goto T, Shirakawa J, Kawano T, Murahashi M, Ide K, *et al*. Odontogenic keratocyst in the mandibular condyle base region: a case report. Experimental and Therapeutic Medicine. 2023; 25: 141.

[32] Oikarinen VJ. Keratocyst recurrences at intervals of more than 10 years: case reports. British Journal of Oral and Maxillofacial Surgery. 1990; 28: 47–49.

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