

Conservative surgical approach for mandibular odontogenic keratocyst associated with guided tissue regeneration: a case report

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Abstract:

Odontogenic keratocyst (OKC) is a cystic lesion with locally aggressive behavior and high recurrence rates, making treatment decisions complex. Conservative approaches have been proposed to preserve oral structures; however, few cases have reported their association with bone reconstruction. This report describes a pediatric case of extensive mandibular OKC managed conservatively with associated bone regeneration. An 11-year-old female patient presented with a radiographic finding of an extensive asymptomatic intraosseous lesion in the right mandible. Incisional biopsy and decompression device placement were performed, with initial diagnosis of dentigerous cyst. After six months, the device was replaced by a smaller one to promote bone neof ormation. Nine months later, a second surgery was performed with device removal, extraction of teeth 47 and 48, and lesion enucleation. Histopathology confirmed OKC. At 28 months postoperatively, recurrence adjacent to the right first molar was detected, and a new conservative procedure was carried out, incorporating bone grafting with sticky bone. The patient was followed up for six months, showing absence of recurrence and graft healing. Despite initial misdiagnosis and multiple surgeries, conservative management preserved the stomatognathic system and avoided ablative procedures. Postoperative follow-up enabled early detection of recurrence and timely intervention associated with bone reconstruction.

Keywords: Decompression; Keratocyst; Pediatric; Guided tissue regeneration; Case report.

INTRODUCTION

The first reported diagnosis of odontogenic keratocyst (OKC) was in 1956 by Philipsen, who described it as any cyst of the jaws showing keratin formation¹. Since then, the World Health Organization has varied in the classification of OKC either as a cyst or as an odontogenic tumor. Pylkkö et al., described the fifth WHO classification, which reclassified OKC from a tumor to a developmental odontogenic cystic lesion as it accounts for 5% to 15% of all odontogenic cysts². OKC has a higher incidence in the age range from 10 to 29 years, with a male predilection at a ratio of 2:1.5 compared to females¹⁻³. The lesion predominantly affects the mandible, but can also be observed in the maxilla^{1,2}.

For most cases, the prognosis is favorable^{2,4,5}. However, its infiltrative characteristics reveal a higher propensity for recurrence after surgical treatment,

Statement of Clinical Significance

Conservative surgical management of extensive mandibular odontogenic keratocyst associated with guided tissue regeneration can preserve mandibular integrity, promote bone regeneration, and reduce morbidity, offering a functional and esthetic alternative to ablative procedures, especially in pediatric patients.

with recurrence rates varying between 5% and 62.5%². Surgical management of OKC may range from more conservative approaches characterized by marsupialization/decompression, enucleation by dissection and/or curettage, with or without adjuvant therapies (e.g. local application of Carnoy's solution and 5-fluorouracil) to more ablative procedures, such as marginal or segmental resection⁶. Although resective surgical treatments generally tend to present lower recurrence rates (i.e.

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4.39%), careful planning of immediate or postoperative bone reconstruction approaches is required^{1,6,7}.

Based on these data, more conservative surgical treatments, with or without adjuvant therapies, are currently considered the most promising strategies for managing OKC. However, it is important to highlight that due to the aggressive nature of the lesion, OKC can cause alterations in the bone morphological pattern of the maxilla and mandible, leading in some cases to loss of teeth adjacent to the lesion. This situation raises concerns regarding the viability of using the remaining alveolar process for future prosthetic rehabilitation with dental implants, as bone reconstruction of defects caused by OKC does not seem to be a routine practice, as observed in the literature. In this context, the objective of the present study is to report a clinical case of an extensive mandibular OKC in a pediatric patient treated with a conservative surgical approach combined with bone reconstruction of the alveolar process region for functional rehabilitation.

CASE REPORT

An 11-year-old melanodermic female patient, with no systemic alterations or relevant family history of hereditary pathologies related to maxillary and mandibular bones, attended the outpatient clinic of the Oral and Maxillofacial Surgery and Traumatology Service at the University Hospital of the Federal University of Juiz de Fora (HU-UFJF/EBSERH). The patient's mother reported that the intraosseous lesion was incidentally observed on a radiographic examination performed for orthodontic treatment purposes three months prior to the first appointment (Figure 1A).

On extraoral examination, no abnormalities were observed in the facial or cervical regions. Intraoral examination revealed the absence of the lower second molars, consistent with the expected eruption pattern for the patient's age.

On panoramic radiography (Figure 1A), a multilocular radiolucent lesion was identified on the right side of the mandible, extending from the apical to the distal region of the first molar (tooth 46). The lesion involved the mandibular body, angle, and ramus, reaching the subcondylar region. Teeth 47 and 48 were impacted and encompassed by the lesion, the former slightly displaced posteriorly and mesially, and the latter displaced posteriorly and positioned close to the ipsilateral mandibular condyle. Cone beam computed tomography (CBCT) was subsequently performed. Image segmentation and

multiplanar reconstruction revealed a well-defined multilocular hypodense lesion measuring 80.5 mm anteroposteriorly, 22.2 mm lateromedially, and 53.7 mm in height (Figures 1B, 1C).

In the retromolar region, a mucoperiosteal flap was raised to create a bony access window to the intraosseous lesion, followed by an incisional biopsy to remove adherent and adjacent fragments from the bony walls. Next, the presence of a clear, slightly viscous fluid was observed. Concurrently, a decompression device consisting of a sterile flexible rubber Penrose drain was placed through the bony access window into the cystic cavity and secured to the adjacent oral mucosa with non-absorbable sutures to maintain continuous drainage and reduce intralesional pressure. The differential diagnoses included dentigerous cyst, OKC and ameloblastoma. Histopathological examination confirmed the diagnosis of dentigerous cyst.

At six months postoperatively, a follow-up CBCT scan showed a reduction in the size of the osteolytic lesion involving the body and ramus of the right mandible. Tomographic images suggested the need to replace the decompression device to optimize the reduction of the lesion size, and a second surgical intervention was performed (Figures 2A, 2B). After a period of approximately three months, the decompression device was removed and enucleation of the cystic capsule was performed with curettage.

Removal of the impacted teeth and complete enucleation of the lesion were performed under general anesthesia with nasotracheal intubation in a hospital setting. The surgery involved creating a mucoperiosteal flap extending from the mandibular ramus to the body, combined with subperiosteal detachment on both buccal and lingual sides. Osteotomy was performed to enlarge the access to the cystic lesion, followed by enucleation by means of dissection (in areas adjacent to the inferior alveolar neurovascular bundle) and curettage to achieve total excision of the lesion components. Concurrently with the removal of the lesion, peripheral ostectomy of the remaining bone was performed and the second and third molars were extracted (Figure 3A). For haemostasis within the bone cavity, a collagen sponge (Gelfoam, Maquira, PR, Brazil) was inserted and closure was performed by using absorbable polyglecaprone sutures (Monocryl, Ethicon, USA).

The anatomopathological material obtained from the enucleation was submitted to histopathological analysis, resulting in a new diagnosis of OKC (Figure 3B). Histological examination revealed a fibrous connective

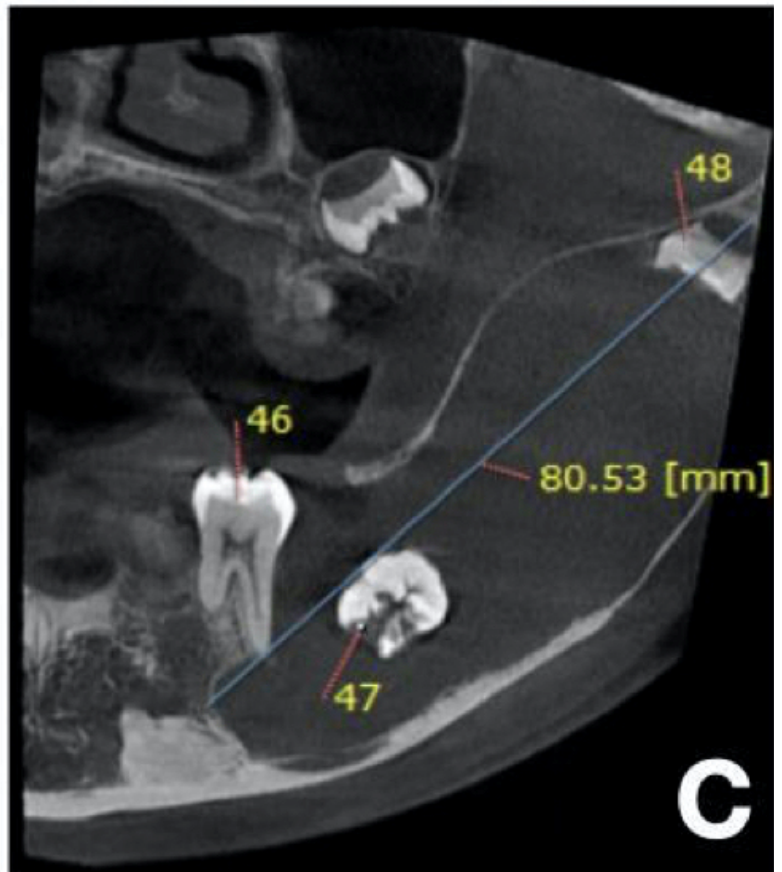
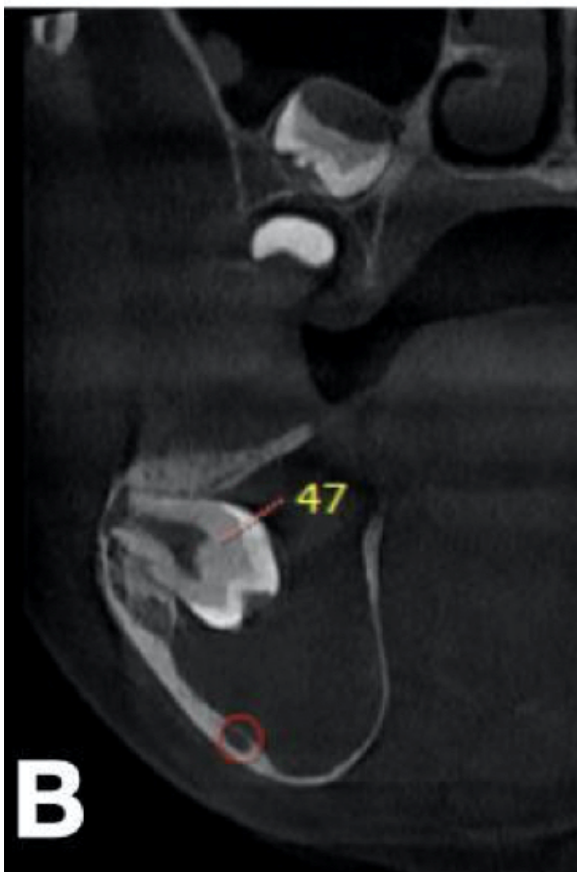
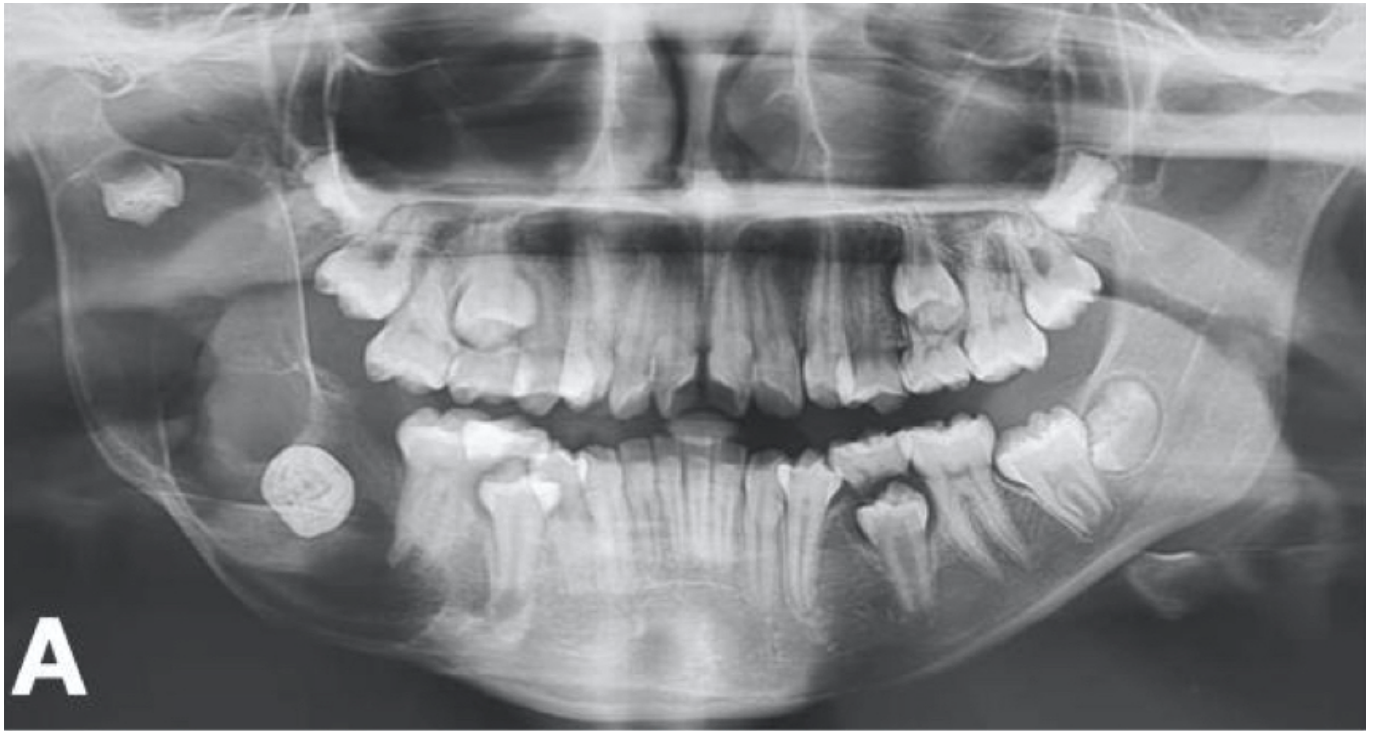


Figure 1. (A) Panoramic radiograph showing an extensive multilocular lesion on the right side of the mandible, involving the body, angle, ramus and sub-condylar region. (B) Coronal multiplanar reconstruction image of the mandibular body region, showing tooth 47 encompassed and displaced by the lesion, with the mandibular canal deviated toward the mandibular base. (C) Sagittal multiplanar reconstruction image revealing tooth 48 displaced posteriorly, adjacent to the mandibular condyle.

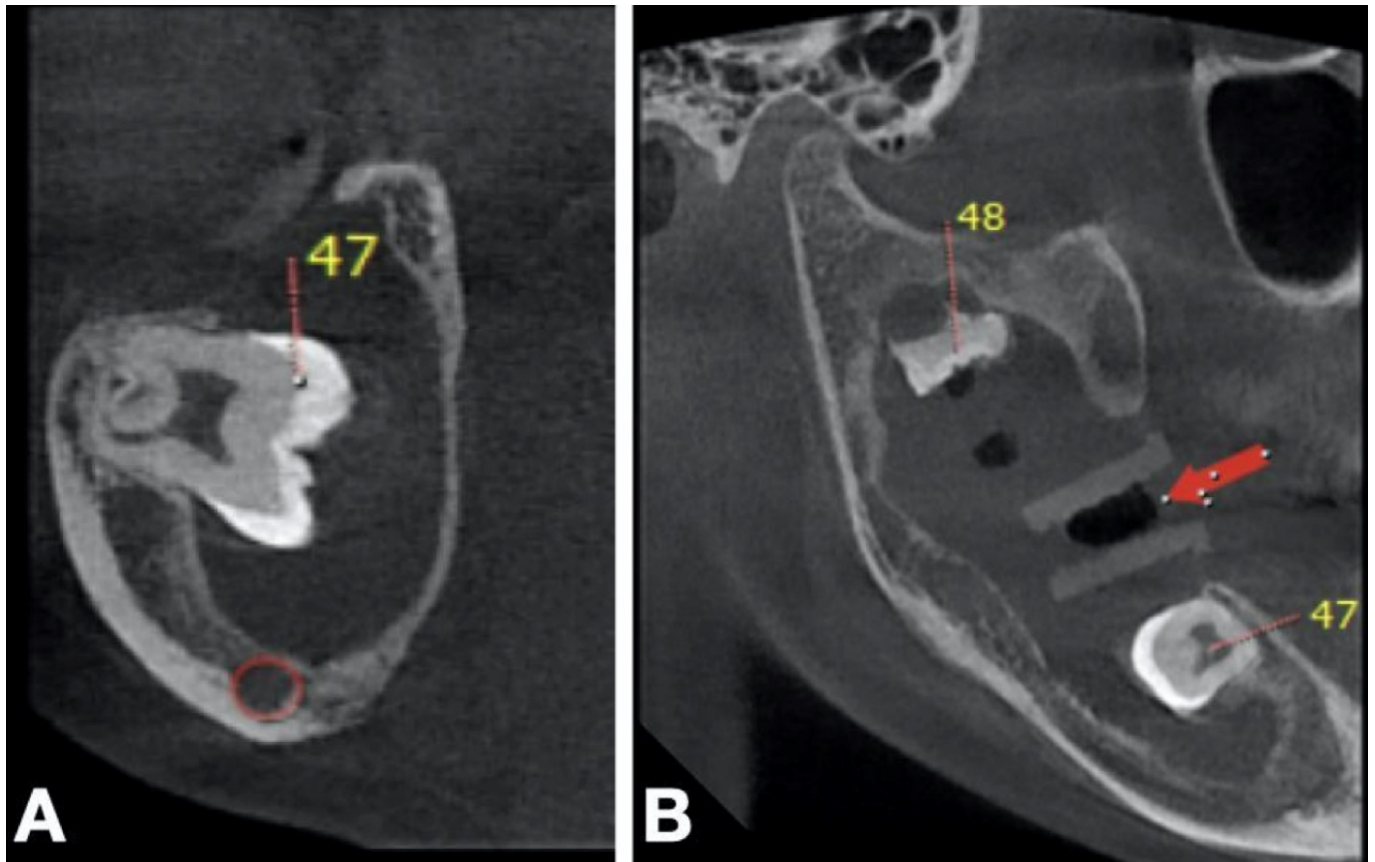


Figure 2. (A) Coronal multiplanar reconstruction image of the mandibular body region showing new bone formation adjacent to the lesion and displaced mandibular canal (red circle). (B) New bone formation surrounding teeth 47 and 48. Rubber drain (red arrow) positioned between the two teeth.

tissue capsule lined by a multi-layered squamous epithelium, approximately 5 to 8 cells thick, uniform and without epithelial rete ridges. The well-defined basal layer consisted of cuboidal or columnar cells arranged in a palisaded pattern. The spinous layer was thin and showed subtle differentiation, whereas the superficial keratinized layer showed incomplete keratinization with an undulated or corrugated epithelial surface (Figure 3C).

The patient was monitored by clinical and radiographic examinations. Follow-ups were weekly during first three postoperative weeks, which were later spaced to biannual visits. After one year and 10 months postoperatively, control tomographic images suggested the presence of a hypodense lesion in the distal region of the right first molar (tooth 46), measuring 15.52 x 12.2 x 15.29 mm. When comparing the CBCT images obtained at 10 months (Figure 4A) to those obtained at 22 months (Figure 4B) postoperatively, minimal new bone formation was observed. Thus, recurrence of smaller cystic lesion adjacent to tooth 46 was suspected, requiring planning for a new surgical approach to diagnose and treat it.

At two years postoperatively, the fourth surgical procedure for removal of the recurrent intraosseous lesion was performed under general anesthesia, following the same parameters used in previous surgeries. Additionally, 60 ml of peripheral blood was collected from the left brachial vein to produce autologous leukocyte- and platelet-rich fibrin membranes (L-PRF) and injectable platelet-rich fibrin (I-PRF). During the same surgical session, peripheral venous blood was collected and distributed into six 10-ml A-PRF tubes (Process for PRF[®], by Choukroun, Implantec, Brazil), vacuum-collected with silica particles and without anticoagulant, and centrifuged at 1300 rpm for 14 minutes in a PRF Duo Quattro centrifuge (Implantec, Brazil)⁸. Additionally, blood was placed into two white-capped vacuum plastic tubes and centrifuged at 700 rpm for 5 minutes (Figure 5A)⁹. After centrifugation, the blood in the silica-coated tubes was separated into red cell fraction and A-PRF clot, which was gently isolated and processed on the PRF Box[®] (Figure 5B) for compressive dehydration, thus generating fibrin membranes ready for use.

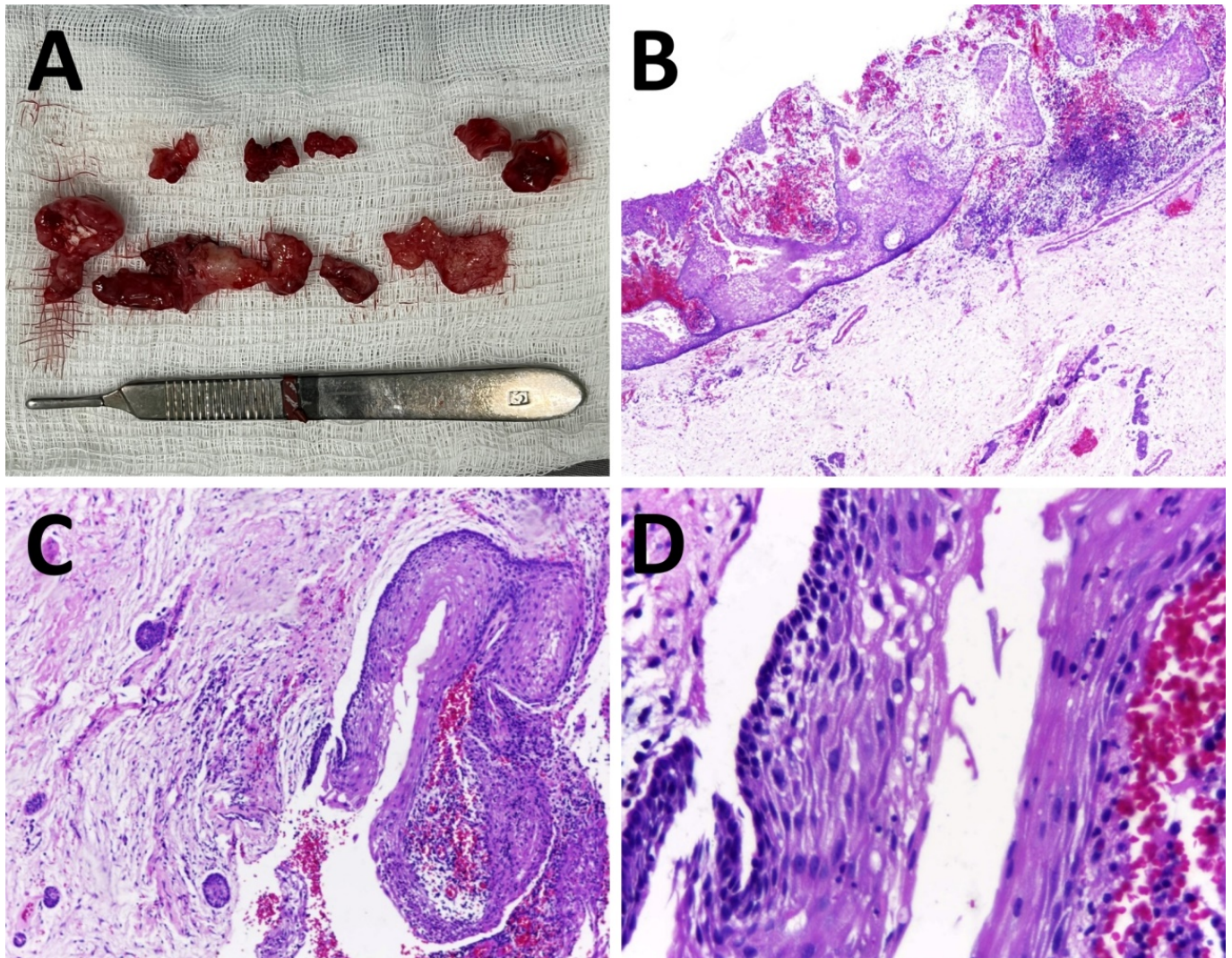


Figure 3. (A) Anatomopathological specimen from the enucleation sent for histopathological analysis. (B-D) Histopathological photomicrograph showing a cystic lesion lined by stratified epithelium with basal layer cells arranged in a palisaded pattern. The fibrous capsule is thin and contains mild to moderate inflammatory infiltrate, with focal hemorrhagic areas (H&E, $\times 40$, $\times 100$, $\times 200$).

As the recurrent lesion was smaller in size and associated with a bone defect from previous approaches, bone grafting was also performed by using autologous block bone graft harvested from external oblique line of the donor site on the contralateral side (Figure 5C). The graft was particulated by using a bone mill (Neodent, Belo Horizonte, Brazil) and mixed with L-PRF and I-PRF to form sticky bone (Figure 5D). The sticky bone was inserted into the bone defect area up to the upper margins of the alveolar process (Figure 5E). Moreover, two or three L-PRF membranes were placed on the bone graft and adjacent bone margin in contact with the overlying soft tissues. Next, the surgical site was sutured by using absorbable suture material. The biological material removed from the recurrent lesion was again sent to histopathological analysis, which confirmed

the diagnosis of OKC. The sutures were removed 10 days after surgery and favorable healing progression were observed. Postoperative follow-up was conducted following the same previously described timeline.

The patient has been currently under follow-up for 37 months since the first intervention. Nine months after the fourth procedure, CBCT images showed features suggestive of favourable scar tissue formation and new bone formation (Figures 6A, 6D). In the future, the patient is expected to receive osseointegrated implants for fixed prosthetic rehabilitation in the mandibular molar region.

DISCUSSION

Since the introduction of the term “keratocyst” by Philipsen in 1956, numerous studies have explored

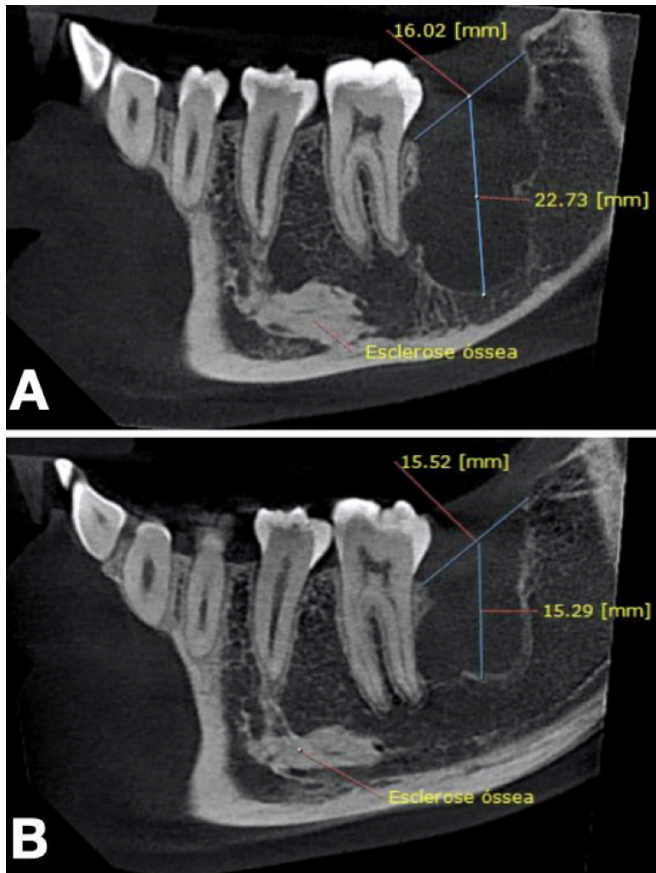


Figure 4. (A) Multiplanar sagittal reconstruction from CBCT imaging at 10 months after the third surgical approach. Bone sclerosis near the apical region of tooth 45. (B) Hypodense lesion in the distal region of tooth 46 at 22 months after the third surgical approach. Minimal bone neoformation.

its clinical, histopathological, and therapeutic characteristics^{2,4,6}. Despite the large volume of publications addressing etiology, pathogenesis and different surgical approaches, there is still a scarcity of studies discussing bone reconstruction strategies following lesion excision, especially in cases of large lesions treated with conservative surgical methods.

As noted in these published studies^{6,9}, the approach considered most effective and least ablative was that using marsupialization/decompression followed by secondary enucleation, with or without peripheral ostectomy, showing a lower recurrence rate (15.5%) compared to isolated marsupialization (19.6%) or direct enucleation (22.1%). Beyond being a less ablative strategy, decompression prior to enucleation has been widely described as biologically favorable in the management of OKC. This approach promotes gradual reduction of intracystic pressure, leading to thickening of the cystic capsule and facilitating subsequent enucleation. Moreover,

decompression has been shown to induce morphological and metabolic changes in the cystic epithelial lining, rendering it more similar to normal oral mucosa and potentially reducing its aggressive behavior, which has been associated with improved prognosis and a lower risk of recurrence^{1,4,6,7,9}. Although this modality requires adequate oral hygiene and a second surgical stage, these biological advantages support its indication as a first-line approach for extensive lesions^{6,7}. Our surgical planning followed these recommendations, and special attention was given to patient instruction and maintenance of decompression device patency.

With regard to the size of an intraosseous lesion, pathological specimens from these cystic lesions must obligatorily be submitted to histopathological examination, which is generally characterized by parakeratinized epithelium with 6 to 10 cell layers, palisaded basal layer pattern, and polarized nuclei^{2,9}. According to Pylkkö et al.², the sensitivity of the histopathological examination may be compromised when the immune system reacts to the cyst formation, resulting in intense inflammatory process around the OKC and thus interfering with the lesion's histology, which may interfere a precise diagnosis. In the present case, the incisional biopsy was performed prior to placement of the decompression device. Therefore, the initial diagnostic limitation should be interpreted as inherent to the limited representativeness of incisional biopsy sampling in large lesions, rather than to the decompression strategy itself. This limitation may result in misinterpretation when small or fragmented tissue samples are obtained. It is likely that the evaluation of a larger and more representative specimen could have allowed an earlier diagnosis of OKC at the time of the initial biopsy.

In this case report, the recurrence of the lesion occurred approximately 22 months (about 2 years) after the third surgical procedure, which highlights the need for long-term postoperative follow-up. This recurrence may be partially explained by the biological behavior of OKC, particularly their thin and friable epithelial lining, as well as the presence of satellite cysts and microcysts left in the overlying mucosa, which may hinder complete removal during enucleation and curettage, especially in extensive lesions. Residual epithelial remnants left within the surgical cavity are well recognized as a major risk factor for recurrence, even when meticulous surgical technique is employed^{6,7,9}. According to previous publications, the average time to lesion recurrence can vary from a minimum of 6 to 9 months up to a maximum of 16 to 21 years, making

it important to maintain close surveillance, particularly during the first five postoperative years, to enable early diagnosis and timely intervention^{4,6}. In this case report, the early detection of recurrence was consistent

with the postoperative time intervals reported for recurrence, which also allowed for a less invasive fourth surgical approach, as well as enabling reconstruction of the bone defect.

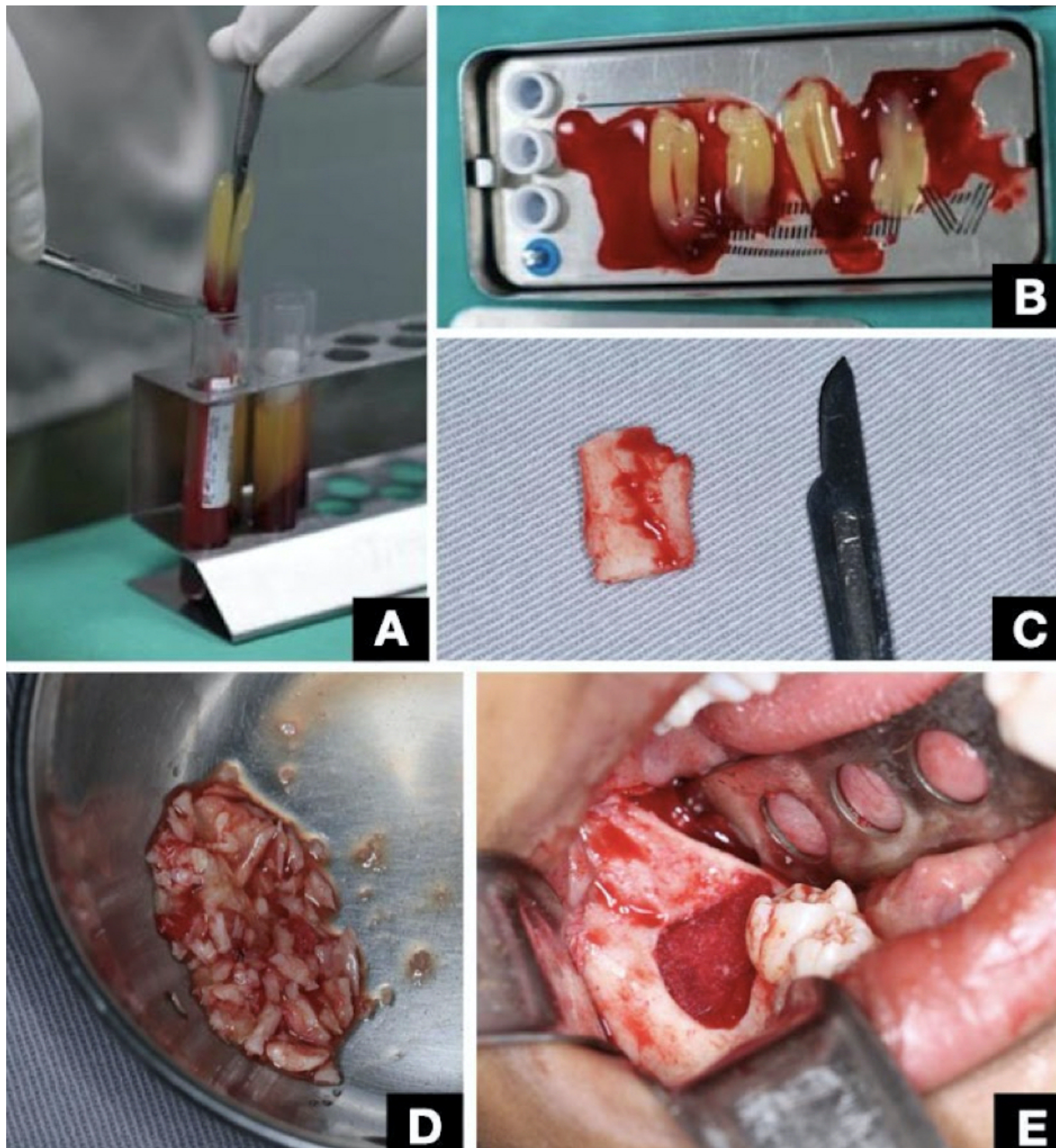


Figure 5. (A-B) L-PRF membranes produced after centrifugation. (C) Autogenous graft harvested from the external oblique ridge (contralateral side). (D) Particulate corticomedullary bone graft agglutinated with L-PRF and I-PRF to form sticky bone. (E) Sticky bone placed on the bone defect region up to the upper margins of the alveolar process.

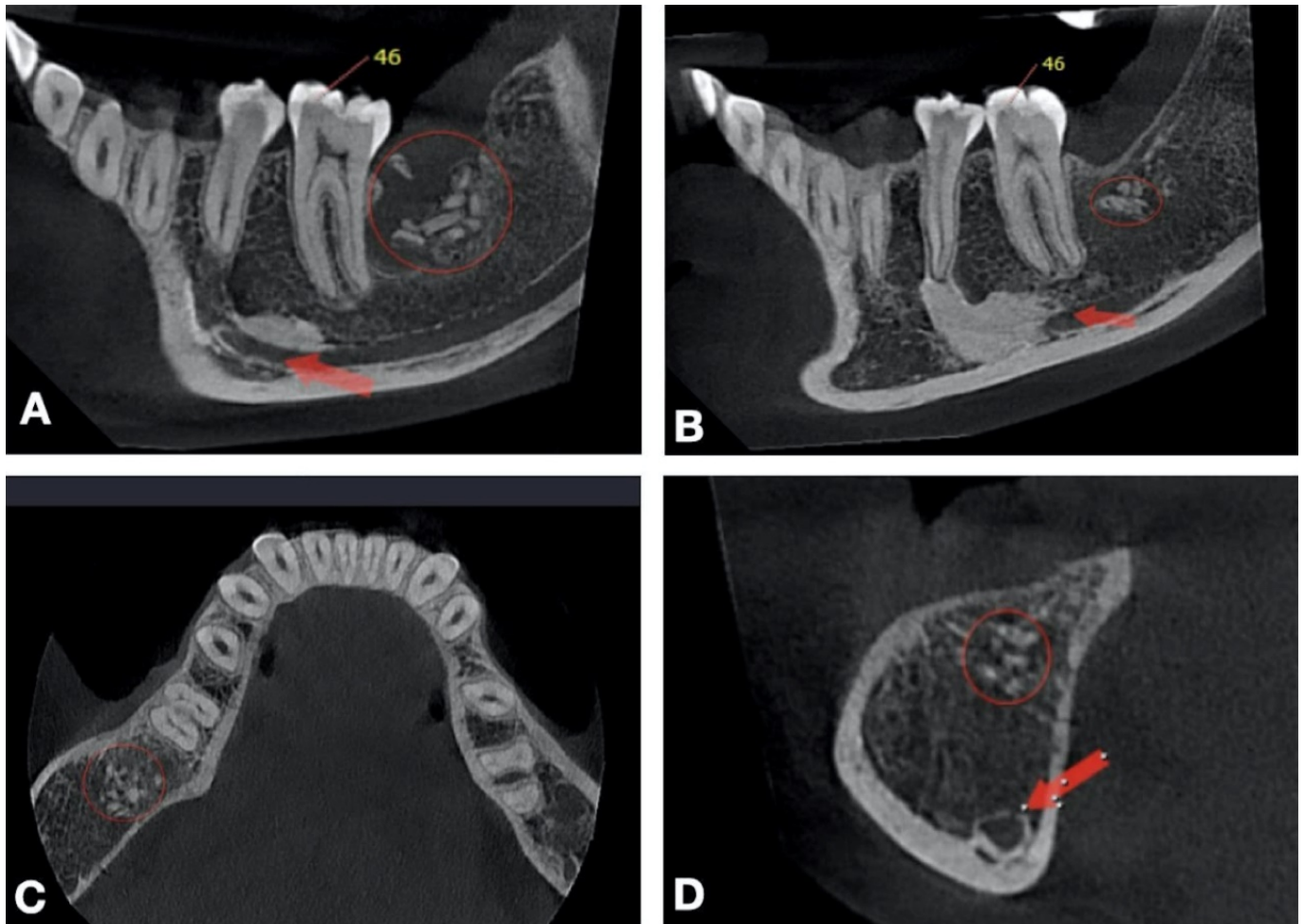


Figure 6. Multiplanar reconstructions: (A) Sagittal view at 3 months after the fourth surgical approach showing a still-visible hyperdense bone graft distal to tooth 46 (red circle); (B) Sagittal view at 6 months after the fourth surgical approach showing a still-visible hyperdense bone graft distal to tooth 46 (red circle); (C) Axial view at 6 months; (D) Coronal view at 6 months.

In the fourth surgical procedure, bone reconstruction was performed concomitantly with enucleation of the recurrent lesion by using a combination of particulate autogenous graft and L-PRF the aim of correcting the residual defect in the mandibular body and alveolar process for future rehabilitation with implant-supported prostheses. According to Quirynen et al., L-PRF promotes tissue regeneration by concentrating platelets, leukocytes and other therapeutic blood components (e.g. fibrinogen/fibrin, growth factors, cytokines and circulating cells) at the surgical site^{10,11}. As demonstrated by Blanco et al.¹², the use of L-PRF may also contribute to the postoperative phase due to its anti-inflammatory, analgesic and antimicrobial properties.

In this case report, the combination of autologous compounds is referred to as sticky bone, which is considered a mixture of particulate autogenous and/or xenogenous bone graft, L-PRF, and I-PRF.

The corticomedullary bone is typically harvested from the donor site at the external oblique ridge due to its bone quality (i.e. high concentration of bone morphogenetic proteins), low morbidity, and minimal complaints of sensory loss on the side where the graft was harvested from^{13,14}. Once the graft was particulated, L-PRF and I-PRF were combined in order to agglutinate bone particles through a dense fibrin network, as recommended by Quirynen et al.^{10,11}.

The authors of this case report opted to graft the bone cavity defect caused by the lesion recurrence by using sticky bone, which eases the adaptation of the graft to the bone defect in the mandibular body and alveolar process, thus minimizing the possibility of graft movement during the maturation phase and preserving the grafted volume throughout healing. Furthermore, there was no need for the use of bone block fixed with titanium screws, titanium mesh, or collagen membranes

to promote guided bone regeneration (GBR). The findings of the systematic review by Sareen et al. support the bone reconstruction approach used in the present case, as fibrin network accelerates bone and soft tissue regeneration without requiring biochemical additives¹⁵. Moreover, fibrin interconnection prevents the ingrowth of these tissues, which renders the sticky bone suitable for the treatment of intraosseous defects^{16,17}.

The efficacy of sticky bone is well recognized to allow both vertical and horizontal bone regeneration, as highlighted by Soni et al., who emphasized the advantages of autografts such as osteogenicity, osteoinductivity and biocompatibility¹⁸. Concerns regarding bone volume and quality are justified in terms of future rehabilitation with dental implants. Accordingly, as analyzed in the network meta-analysis by Calciolari et al.¹⁹, adequate bone in the reconstruction area is essential not only to enable placement of dental implants and prosthetic rehabilitation, but also to maintain the soft tissue margin and interdental papillae over time following implant placement. Therefore, the use of sticky bone in this present case was fundamental to provide a suitable bone bed for the future insertion of osseointegrated dental implants.

The use of regenerative approaches after enucleation of OKC has been described in a limited number of clinical reports²⁰⁻²². Previous case reports have shown favorable healing and bone regeneration following the use of leukocyte-platelet rich fibrin or guided tissue regeneration techniques, with no evidence of recurrence during follow-up periods of 12²⁰, 15²¹, and 36 months²². Although these findings suggest that regenerative strategies may be safely applied after OKC removal, the available evidence is restricted to case reports and small case series. Therefore, considering the aggressive biological behavior and recurrence potential of OKC, the application of regenerative techniques should be accompanied by rigorous long-term clinical and radiographic follow-up.

Although we believe that the likelihood of recurrence in this case report is relatively low, it cannot be definitively excluded. This interpretation must be viewed with caution, considering the well-known biological behavior of OKC, the history of multiple surgical interventions, and the fact that complete capsular integrity could not be fully preserved throughout the treatment course. It can be hypothesized that the 22-month follow-up period may be considered short for evaluating bone reconstruction immediately after enucleation of the recurrent lesion (fourth surgery), which could imply

that a new recurrence might require removal of the lesion and consequent damage to the reconstructed bone tissue. Conversely, the patient's frequent postoperative visits allowed for early management, which highlights that a close follow-up helps maintain the morphological integrity of the mandible by preventing the lesion from reaching larger proportions.

Finally, we believe that simultaneously performing bone reconstruction will facilitate future implant placement, although we recognize that there is currently no consensus regarding the ideal age for dental implant placement in growing patients²³. Regardless of the lack of an established optimal timing, the results from a systematic review and meta-analysis indicate that implant success rates in adolescents around 16 years of age do not differ significantly from those in adults, provided that strict monitoring is ensured²³. It is important to emphasize that the studies included in this systematic review did not specifically address implant placement in sites previously affected by aggressive odontogenic lesions with a known potential for recurrence, such as OKC. Therefore, extrapolation of these findings to the present clinical scenario should be approached with caution. In the present case, implant-based rehabilitation is not planned in the short term and should only be considered in the future, after long-term clinical and radiographic follow-up, with acquisition of new imaging studies and confirmation of disease-free status for a minimum period of five years.

CONCLUSION

This case report demonstrates that the conservative management of an extensive mandibular OKC in a pediatric patient, combined with bone reconstruction using autologous graft and platelet concentrates, can achieve favorable clinical and radiographic outcomes while preserving mandibular integrity.

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AUTHORS' CONTRIBUTIONS

LVA: Conceptualization, Formal analysis, Investigation, Writing – original draft. YLM: Methodology, Formal analysis, Investigation, Validation,

Writing – original draft. LMS: Data curation, Formal analysis, Investigation, Visualization. KAMP: Data curation, Formal analysis, Investigation, Visualization. BABA: Investigation, Methodology, Software, Resources, Validation, Visualization, Writing – review & editing. DAAM: Conceptualization, Investigation, Methodology, Project administration, Resources, Supervision, Writing – review & editing.

CONFLICT OF INTEREST STATEMENT

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Competing interests: The authors have no relevant financial or non-financial interests to disclose.

Ethics approval: This case report was approved by the Human Research Ethics Committee of the Federal University of Juiz de Fora (Juiz de Fora, MG, Brazil), under protocol number 040018/2025 (CAAE 87869825.0.0000.5133). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

DATA AVAILABILITY STATEMENT

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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