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Original Research

Decompression as conservative treatment of multiple odontogenic keratocysts in a patient with Gorlin-Goltz syndrome: 6-year follow-up

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Odontogenic keratocyst is an intraosseous lesion with aggressive potential and infiltrative behavior, presenting high recurrence rates. The occurrence of multiple odontogenic keratocysts in the same patient may be associated with Gorlin–Goltz syndrome, a condition characterized by a predisposition to neoplasms and other developmental abnormalities. There is no consensus regarding the best treatment modality. In cases of multiple or large lesions, surgical management may be associated with significant morbidity, resulting in aesthetic and functional impairment. Conservative approaches, such as decompression, represent an effective treatment option due to their simplicity and low morbidity. **Aim:** This study aims to describe the case of a 13-year-old female patient diagnosed with Gorlin–Goltz syndrome and multiple odontogenic keratocysts in the mandible. **Methods:** The patient was treated using surgical decompression techniques and posterior enucleation. Decompression devices were fabricated from surgical suction tips or needle cap tips and were placed under local anesthesia in five mandibular lesions. After 17–21 months following the decompression procedures, a significant volumetric reduction was observed in four out of five lesions. Subsequently, the patient underwent enucleation surgery. The patient has been under long-term postoperative follow-up for six years after decompression and approximately five years after enucleation, demonstrating satisfactory postoperative recovery, bone neoformation, and absence of aesthetic or functional sequelae. **Conclusion:** Conservative techniques such as surgical decompression for the treatment of odontogenic keratocysts are simple, low-cost, and effective options, reducing surgical morbidity and ensuring favorable aesthetic and functional outcomes. However, continuous follow-up and patient compliance are essential for optimal results.

Keywords: Odontogenic cysts. Basal cell nevus syndrome. Decompression, surgical.

Introduction

The term odontogenic keratocyst (OKC) was first introduced by Philipsen in 1956 to describe mandibular cysts exhibiting keratinized epithelial lining[1]. However, the defining biological and histopathological characteristics of the OKC were only fully described later by Pindborg and Hansen in 1963[2].

The odontogenic keratocyst was initially classified as a cystic lesion[3]. Nevertheless, due to its aggressive growth potential, high postoperative recurrence rates, and the identification of genetic mutations associated with its pathogenesis, the World Health Organization (WHO) reclassified the lesion in 2005 as a benign neoplasm, naming it keratocystic odontogenic tumor (KCOT)[4]. Subsequently, after further scientific evaluation, this classification was reconsidered, and in 2017 the WHO reinstated the term odontogenic keratocyst, defining it as a benign odontogenic cystic lesion[5].

The odontogenic keratocyst remains one of the most controversial pathological entities in the maxillofacial region. Accounting for approximately 12–14% of all odontogenic cysts and tumors, OKC is an intraosseous lesion characterized by aggressive behavior, infiltrative growth, and a high tendency for recurrence[6-8]. Although it may occur at any age, it predominantly affects individuals in the second and third decades of life[9]. The mandible is more frequently involved than the maxilla, particularly the posterior region and ascending ramus, and the lesion is commonly associated with impacted teeth[3,9]. The OKC is considered a developmental epithelial cyst arising from remnants of the dental lamina or basal cells of the oral epithelium[3,9,10].

The occurrence of multiple odontogenic keratocysts in a single patient is frequently associated with Gorlin-Goltz syndrome (GGS), also known as nevoid basal cell carcinoma syndrome, which may also present clinical features such as bifid ribs and multiple basal cell nevi. Therefore, histopathological examination is essential for definitive diagnosis[3,10,11]. GGS is a rare, hereditary, autosomal dominant disorder with well-defined clinical criteria, first described by Gorlin and Goltz in 1960, and is characterized by a predisposition to neoplasms and other developmental abnormalities[12,13]. OKC treatment in patients with Gorlin–Goltz syndrome can be more challenging due to higher recurrence rates and the diversity of clinical manifestations associated with this syndrome, necessitating a multidisciplinary approach[14-16].

There is currently no consensus regarding a single optimal surgical approach for the treatment of odontogenic keratocysts, given their aggressive behavior and significant recurrence rates. Treatment modalities vary across institutions, ranging from conservative approaches (marsupialization, decompression, simple enucleation, enucleation following decompression, and enucleation combined with adjunctive therapies) to more aggressive options, such as bone resection. Several factors must be considered when selecting the treatment modality, including lesion size and location, proximity to adjacent anatomical structures, radiographic evidence of cortical bone perforation, histopathological features, recurrence risk, as well as the patient's age and systemic health status[17]. Conservative treatment

strategies are generally preferred, particularly in pediatric and adolescent patients, in order to avoid invasive procedures that may result in extensive bone loss, facial deformities, and significant psychosocial impact[18,19]. Literature have demonstrated that the combination of enucleation and adjunctive therapies may offer improved therapy in the management of OKC[20,21].

This study presents a case report of a 13-year-old female patient diagnosed with Gorlin–Goltz syndrome and multiple odontogenic keratocysts affecting the mandible. Decompression was selected as the initial treatment modality for the cystic lesions, followed by enucleation and adjunctive local cryotherapy when indicated. Therefore, this report highlights the main aspects of decompression as a conservative treatment approach, a technique widely supported in the literature for extensive odontogenic keratocysts and associated with high success rates[22-24].

Ethical Aspects

This case was approved by the Research Ethics Committee of the Federal University of Rio Grande do Sul (CAAE: 41796721.6.0000.5347). The patient and her legal guardian were informed about the risks and benefits of the procedure and of participation in the study. After reading the informed documents and having their questions clarified, both the patient and her legal guardian signed the Informed Consent Form, the Assent Form, the Post-Informed Consent, and the Authorization for Use of Images.

Case Report

A 13-year-old female patient, in good general health, sought care at the Lutheran University of Brazil (ULBRA) in 2018, complaining of pain and mild facial swelling. The patient's legal guardian had noticed facial "swelling" in the mandibular region. Panoramic radiography revealed multiple extensive radiolucent lesions affecting the mandible. An incisional biopsy was performed in October 2018, and the histopathological diagnosis was odontogenic keratocysts. The patient was subsequently referred to the Oral and Maxillofacial Surgery Unit of the Hospital de Clínicas de Porto Alegre (HCPA) for continuation of treatment.

In May 2019, the patient attended her first consultation with the Porto Alegre team, during which a clinical examination and evaluation of imaging studies were performed. A facial computed tomography scan and a new panoramic radiograph were requested. Concurrently, the patient was referred to the hospital's Medical Genetics team for evaluation of a possible diagnosis of Gorlin–Goltz syndrome.

The geneticist evaluation revealed long fingers, a hypopigmented abdominal macule, thoracic asymmetry, and palmar pits. Both the patient and her legal guardian denied any family history of basal cell carcinoma or macrocephaly. Complementary examinations demonstrated morphostructural abnormalities affecting the rib cage bilaterally, including rib hypoplasia and fusion of the fifth and sixth ribs, as well as mild right-convex thoracic scoliosis, left-convex lumbar scoliosis, and pelvic tilt, thereby confirming the diagnosis of Gorlin–Goltz syndrome. The patient and her legal guardian were informed of the diagnosis and received specific guid-

ance, particularly regarding sun exposure precautions and a scheduled 6-month follow-up. Referrals for dermatological evaluation and spinal orthopedic assessment were also provided.

The patient returned in June 2019 for follow-up with the Oral and Maxillofacial Surgery team to review the requested imaging studies. Five expansive lesions involving large areas of the mandible were identified. Based on the clinical findings and imaging evaluation (Figures 1-3), as well as the number of lesions, their size and location, and the patient's age, a conservative treatment approach was selected. The proposed management consisted of decompression of the lesions followed by enucleation and adjunctive local cryotherapy when indicated. The patient and her legal guardian were informed about the chosen treatment strategy and the importance of patient compliance throughout the treatment period.

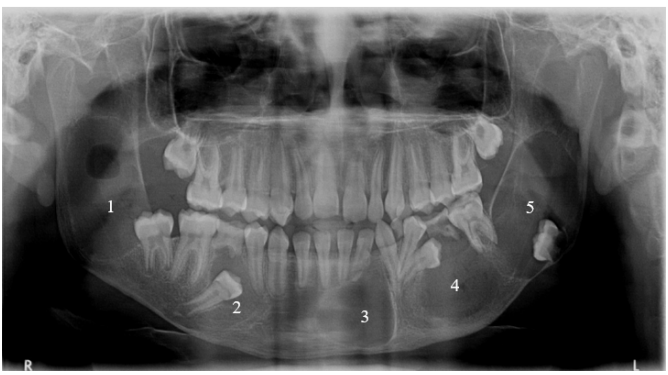

Figure 1. Panoramic radiograph showing five mandibular cystic lesions, well delineated by radiopaque margins. 1) Extensive lesion located in the right mandibular ramus involving tooth 47. 2) Cystic lesion in the right mandibular body involving the unerupted tooth 45. 3) Cystic lesion in the anterior mandibular region. 4) Lesion located in the left mandibular body involving the unerupted teeth 35 and 75. 5) Extensive lesion in the left mandibular ramus involving the unerupted tooth 37.

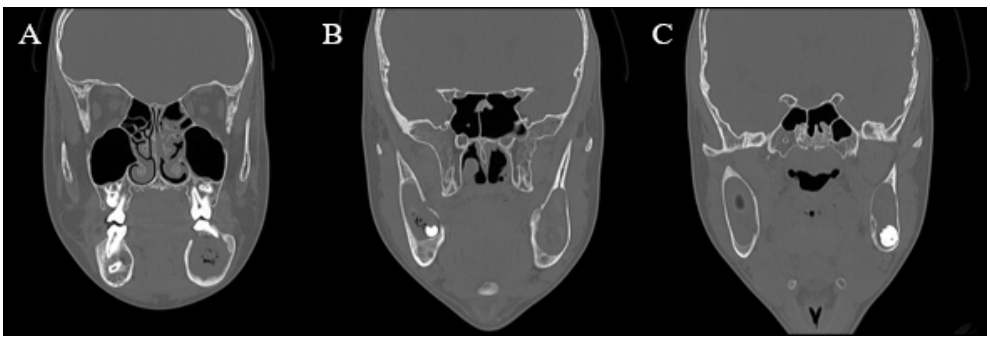

Figure 2. Coronal section in preoperative Computed Tomography. A) Lesions 2 and 4. B) Lesions 1 and 5. C) Lesions 1 and 5.

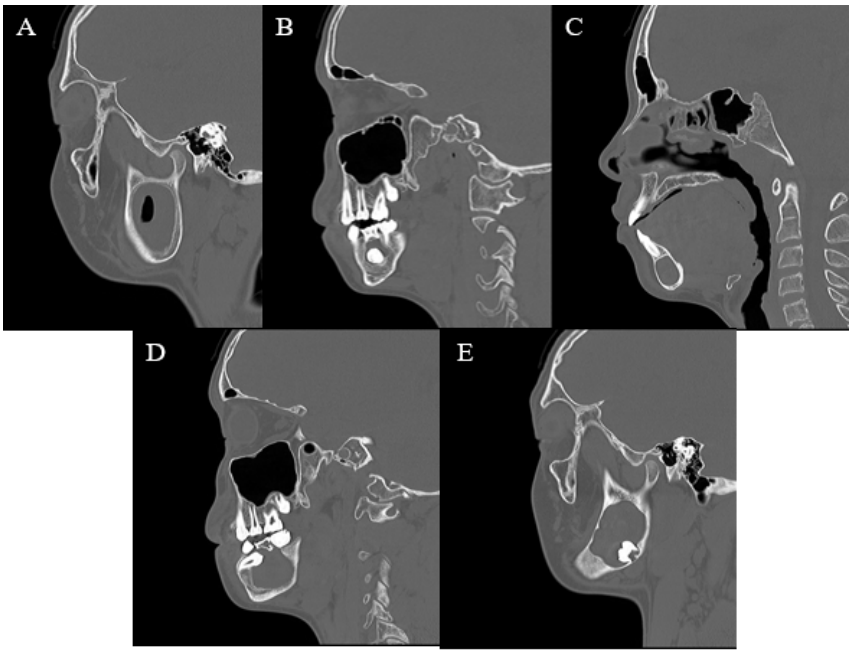

Figure 3. Sagittal section in preoperative Computed Tomography. A) Lesion 1. B) Lesion 2. C) Lesion 3. D) Lesion 4. E) Lesion 5.

Treatment

As previously reported, the treatment of choice consisted of surgical access to the lesions followed by decompression using custom-made devices. These devices establish a communication between the interior of the lesion and the oral cavity, resulting in a progressive reduction of the cystic cavity. The decompression devices were manually fabricated using surgical suction tips and the plastic hub of needle caps. Given that the patient was preadolescent, the surgical interventions were performed under local anesthesia in three separate sessions. The anesthetic solution used was 2% lidocaine with 1:100,000 epinephrine. The principle of all procedures was the same: access to the cystic cavity through osteotomy using burs, followed by placement of the device to maintain communication between the cystic cavity and the oral environment. The first procedure was carried out in July 2019 and involved the placement of decompression devices in the left-sided lesions, surgical extraction of teeth 73 and 75, and fixation of both the device and the surgical wound using 5-0 mononylon sutures. The second procedure was performed in September 2019 and consisted of surgical extraction of tooth 85 and decompression of the lesions located on the right side. The final procedure was carried out in November of the same year, addressing the lesion located in the anterior region of the mandible (Figure 4).

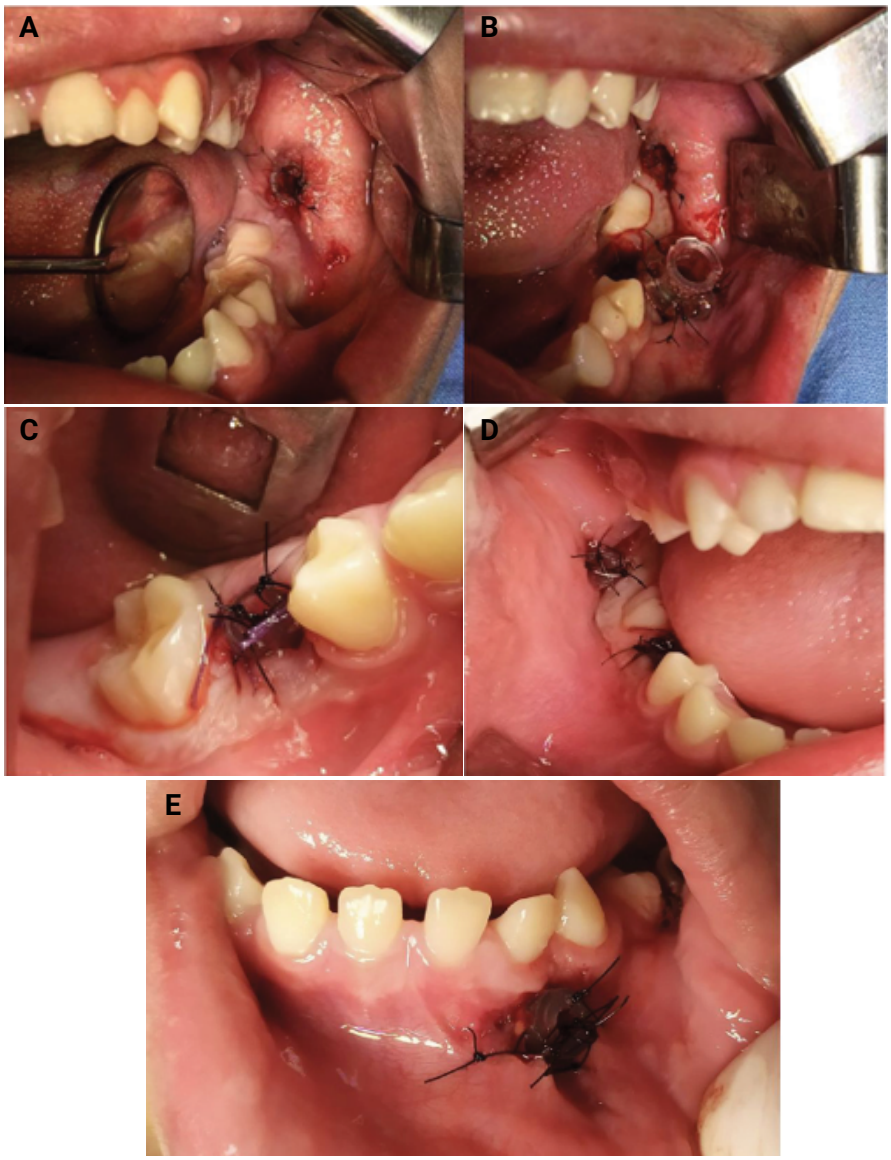

Figure 4. Intraoperative images. A and B) First surgical procedure performed. C and D) Second surgical procedure performed. E) Third surgical procedure performed.

Postoperative Follow-up

In addition to standard postoperative care applicable to any surgical intervention, the patient was instructed to perform intra-lesional irrigation five times daily using saline solution and 0.12% aqueous chlorhexidine with the aid of a 20-mL syringe. Furthermore, both the patient and her legal guardian were provided with direct contact information for the surgical team in case of questions or complications, such as device displacement or signs of local infection.

Following the procedures, the patient continued regular follow-up with the team through outpatient visits at the Hospital de Clínicas de Porto Alegre, with both clinical and radiographic monitoring until sufficient reduction of the lesions was achieved, allowing for subsequent surgical enucleation and adjunctive cryotherapy. The effectiveness of decompression of the odontogenic keratocysts was assessed by comparing pre and postoperative imaging studies. The expected outcome was local bone neoformation and a reduction in lesion size. Despite the patient's good overall compliance during the postoperative period, her legal guardians reported considerable difficulty in irrigating the lesion located in the anterior region of the mandible.

In April 2021, a new panoramic radiograph and tomography were requested to evaluate the lesions. Imaging revealed a marked reduction in the odontogenic keratocysts, with the exception of the lesion in the anterior mandibular region, where hygiene had been more challenging for the patient and her caregivers (Figures 5 and 6). Consequently, surgical enucleation of the cystic lesions was performed under general anesthesia, and cryotherapy was used as an adjunctive treatment (Figure 7).

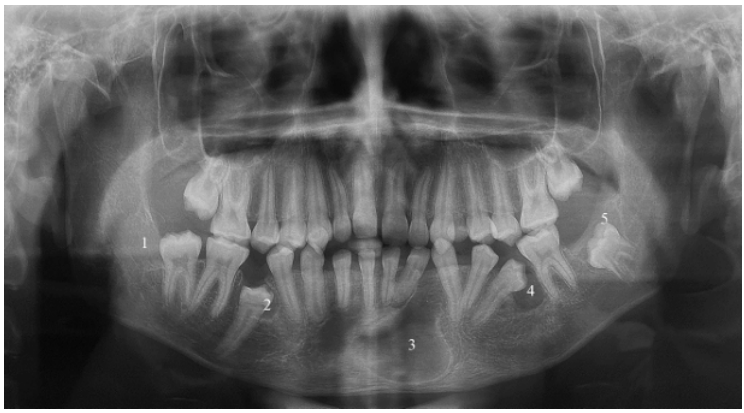

Figure 5. Panoramic radiograph showing five mandibular cystic lesions after decompression. 1 and 2) 19-month follow-up. 3) 17-month follow-up. 4 and 5) 21-month follow-up.

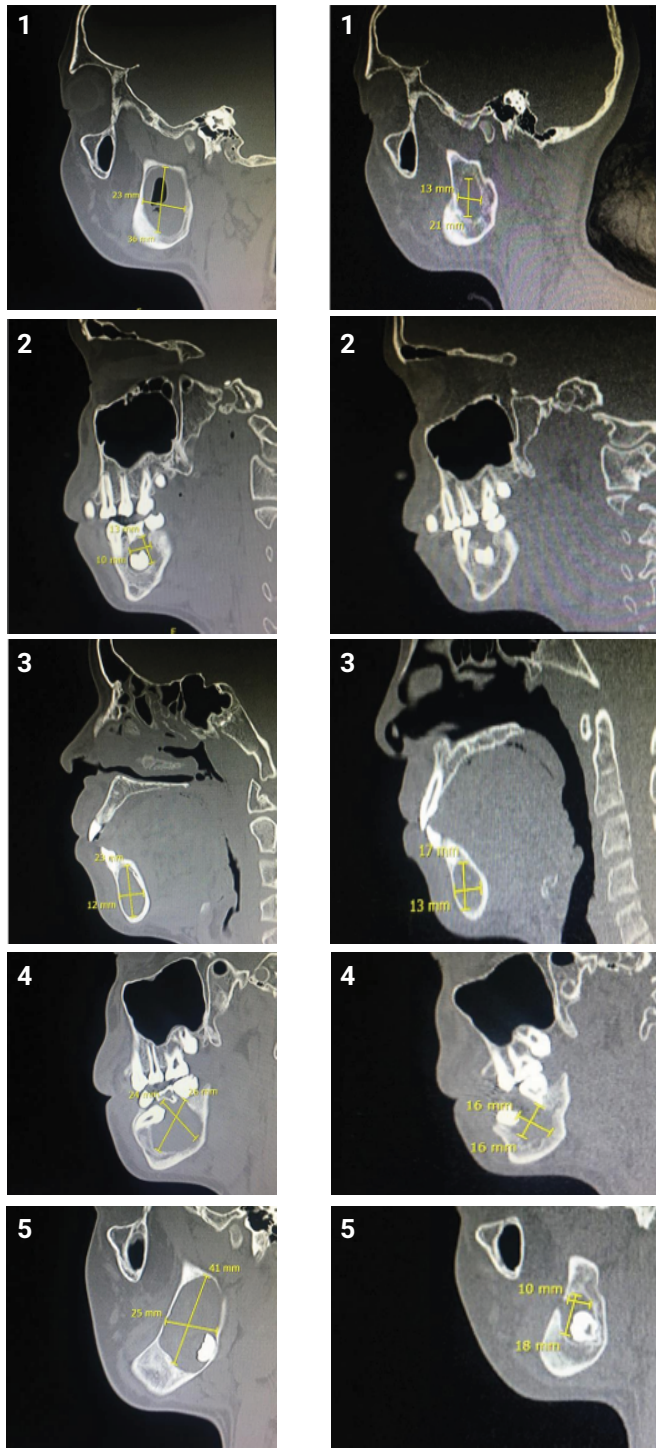

Figure 6. Tomography showing five mandibular cystic lesions before and after decompression in sagittal slice.

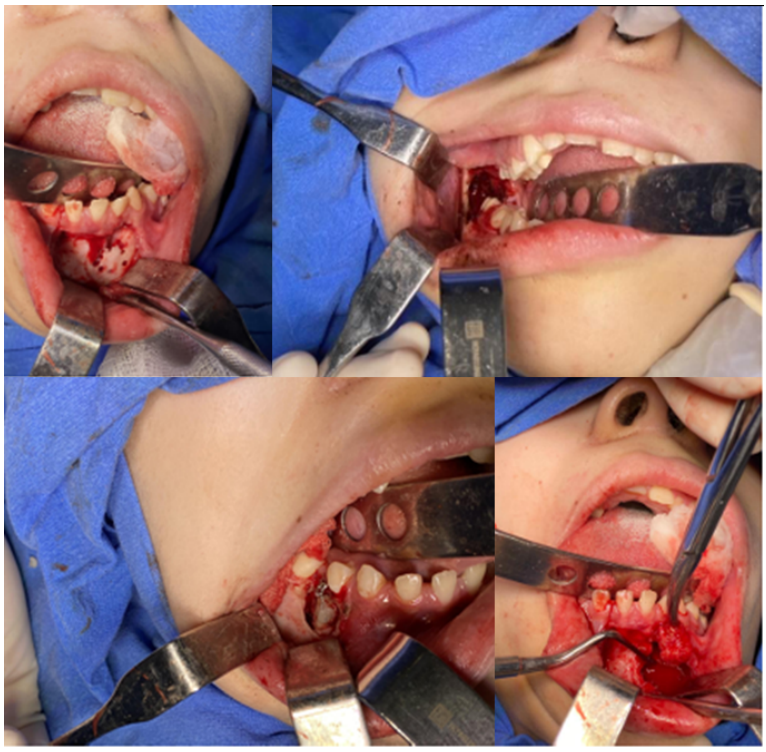

Figure 7. Intraoperative images of the odontogenic keratocysts enucleation surgery in mandible.

The patient demonstrated good postoperative recovery and has remained under regular follow-up with the clinical team. She is currently in the late postoperative period, six years after the initial procedure performed by the team. Physical examination revealed no aesthetic or functional limitations. Radiographic evaluation demonstrated satisfactory bone healing with no evidence of recurrence (Figure 8).

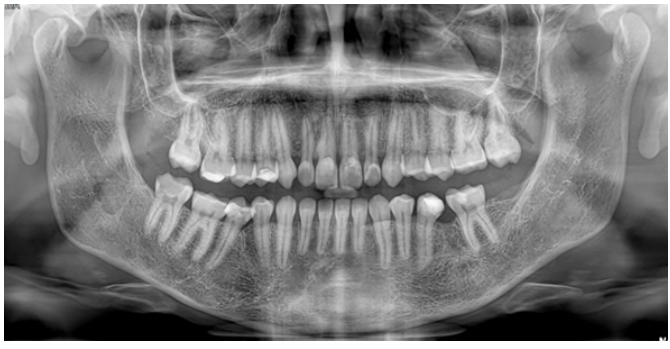

Figure 8. Postoperative panoramic radiograph showing great bone healing and no evidence of recurrence.

To facilitate understanding of the surgical stages, a timeline outlining the dates of each surgical procedure is provided (Figure 9)

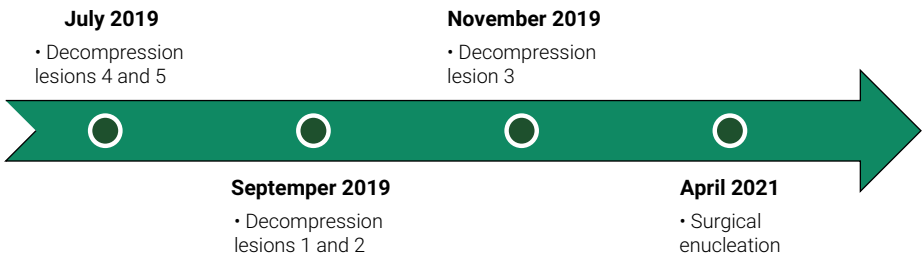

Figure 9. Timeline of each surgical procedure.

Discussion

OKC is an intraosseous lesion with aggressive potential and infiltrative behavior. This pathological condition presents slow growth and is often asymptomatic. Consequently, clinical identification in the early stages is not possible in most cases, and lesions are frequently detected unintentionally during routine imaging examinations. When symptomatic, patients may present pain and cortical bone expansion, as observed in the present case, in which the patient exhibited facial swelling. In cases of multiple odontogenic keratocysts occurring in the same patient, the condition may be associated with Gorlin–Goltz syndrome, a rare hereditary disorder. OKCs are present in up to 75% of patients with GGS and are often the first clinical manifestation of the syndrome[25].

Due to its thin and friable capsule, the presence of cortical bone perforation, adhesion to adjacent soft tissues, and the occurrence of satellite cysts, OKC presents a high recurrence rate, as complete removal is not always achieved as a result of these factors. In syndromic OKCs, the likelihood of recurrence is even higher, since these lesions exhibit a greater number of satellite cysts, solid epithelial proliferation islands, odontogenic remnants within the capsule, increased parakeratinization, and mitotic figures in the epithelium[26]. Therefore, the comprehensive management of syndromic OKCs should involve a multidisciplinary team comprising dental, medical, and genetic specialists.

Despite advances in surgical techniques, controversy regarding the most effective management of odontogenic keratocysts persists. The inherent aggressive behavior and high recurrence rate of OKCs remain a significant challenge for oral and maxillofacial surgeons. Consequently, several treatment modalities have been proposed, however, none has proven to be completely infallible. Factors such as the number of lesions, size, location, proximity to critical anatomical structures, patient age, histological subtype, and association with syndromes must be carefully considered during treatment planning[17].

Decompression has been widely advocated in the literature, particularly in cases of extensive odontogenic keratocysts, such as the one reported herein. This technique reduces intracystic fluid pressure and induces bone deposition toward the

cystic wall by creating a communication between the cystic cavity and the oral environment through the use of a device. Furthermore, the device sutured to the lesion margin prevents spontaneous closure and allows internal sanitation of the cavity, avoiding bacterial proliferation and promoting morphological changes in the cystic capsule due to constant trauma generated by irrigation with saline solution or 0.12% chlorhexidine digluconate. Therefore, decompression is a treatment modality that preserves bone integrity, adjacent tissues, neuromuscular function, and psychological well-being.

The benefits of this approach are particularly significant in pediatric patients and include preservation of tooth buds, minimal interference with skeletal growth, and a lower risk of damage to adjacent anatomical structures such as the maxillary sinus, mandibular canal, and nasal and orbital cavities. Additionally, children and their parents tend to demonstrate better adherence to less invasive surgical approaches[27]. However, decompression also presents disadvantages, including prolonged treatment duration, discomfort, loss of the device, obstruction of the drainage opening, irrigation difficulties, infections, and, most importantly, dependence on patient compliance[28].

In the present case, the decompression tubes were sutured to the lesion margins using 5-0 mononylon sutures. Nevertheless, alternative fixation methods have been described in the literature using strailess steel wire and orthodontic brackets[29,30].

Regarding patient compliance, both the patient and her legal guardian adhered to postoperative instructions, which included regular irrigation with saline solution or 0.12% chlorhexidine digluconate. However, significant difficulty was reported in irrigating the lesion located in the anterior mandibular region. Consequently, this lesion was the only one that did not demonstrate a reduction in size. The authors believe that the lack of volumetric reduction in this lesion was due to insufficient irrigation of the cystic cavity, as it was the only cyst that did not receive consistent internal sanitation and also the only one without radiographic evidence of bone neoformation.

The literature reports that morphological changes at both macroscopic and microscopic levels can be observed in cysts treated by decompression as a result of constant trauma caused by exposure to irrigation solutions. These changes include capsule thickening and reduced friability, increased inflammatory infiltration in the connective tissue, and epithelial metaplasia, with transformation from a thin parakeratinized epithelium to a hyperplastic, non-keratinized squamous epithelium. Additionally, intraosseous pressure decreases, leading to reduced secretion of bone resorption factors, diminished osteoclastic activity, and inhibition of lesion growth. Consequently, bone formation begins concomitantly with cyst size reduction and may eventually result in complete regression[31]. The effectiveness of decompression was higher in younger patients, with longer decompression periods, and when lesions were located in the posterior maxilla. Among the evaluated parameters, age was considered statistically significant[32].

There is no consensus regarding the ideal duration of cyst decompression. In summary, studies suggest a decompression period ranging from 3 to 14 months and a reduction of approximately 50–60% in lesion size as optimal before pro-

ceeding to enucleation[32-34]. Over the 21-month follow-up period following the initial surgery for drain insertion, all cystic lesions demonstrated considerable reduction compared to their initial volumes, as assessed by imaging studies.

In cases where complete regression does not occur, most authors advocate marsupialization or decompression as the initial treatment modality, followed by enucleation combined with curettage, peripheral ostectomy, application of Carnoy's solution, or liquid nitrogen cryotherapy[27]. When lesion size is reduced after decompression and adjacent critical anatomical structures—such as the inferior alveolar nerve and neighboring teeth—are no longer compromised, the timing of cyst enucleation should be determined based on the surgeon's clinical judgment[32].

Decompression proved to be an effective procedure for the management of multiple and extensive odontogenic keratocysts, particularly in young patients. Overall, the outcomes were positive and met the authors' expectations regarding lesion volume reduction and bone neoformation, as determined by volumetric analysis using computed tomography, as well as preservation of the patient's anatomical structures, maintaining function, form, and facial aesthetics.

The conservative approach adopted in this case requires a high level of patient commitment to postoperative care, extending beyond the responsibilities of the oral and maxillofacial surgeon alone. Therefore, it is essential that the patient and caregivers are fully informed of this requirement prior to the placement of decompression drains, in order to ensure lesion regression without complications such as infection resulting from inadequate or infrequent irrigation.

As with all cases of odontogenic keratocysts, the patient will remain under long-term clinical and radiographic follow-up by the oral and maxillofacial surgery team at the Hospital de Clínicas de Porto Alegre due to the high recurrence potential of this lesion. In addition, continued follow-up with the genetics and dermatology teams is required for evaluation of basal cell carcinoma lesions and other manifestations associated with Gorlin–Goltz syndrome.

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Conflict of Interest

The authors deny any conflicts of interest related to this study.

Data Availability

Datasets related to this article will be available to the corresponding author upon request.

Author Contribution

Angelo Luiz Freddo: Investigation, Supervision, Writing – Review & Editing. **Henrique Tedesco de Oliveira:** Investigation, Visualization, Writing – Review & Editing. **Amália**

Pletsch Furlanetto: Investigation, Visualization, Writing – Review & Editing. **Júlia Heidrich:** Investigation, Visualization, Writing – Original Draft. **Adriana Corsetti:** Investigation, Supervision, Writing – Original Draft. All authors actively revised and approved the final version of the manuscript.

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