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CASE REPORT

Management of an extensive calcifying odontogenic cyst in the maxilla of a young patient: a case report

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Abstract:

The calcifying odontogenic cyst (COC) is a rare developmental lesion with variable clinical behavior, representing less than 1% of all odontogenic cysts. Its management can be challenging but typically involves enucleation and curettage. This case report aims to describe the clinical, radiographic, histopathological findings, and therapeutic approach used in the treatment of a maxillary COC in a young patient. A 19-year-old female patient presented with a complaint of progressive, asymptomatic swelling on the right side of her face, with a 12-month history. Extraoral examination revealed obliteration of the nasolabial fold and facial asymmetry in the right malar region. Intraoral examination showed a noticeable swelling in the right maxillary vestibule. Cone-beam computed tomography revealed a unilocular, hypodense lesion surrounding the crown of the impacted maxillary canine (tooth 13) and extending to the periapical region of adjacent teeth. An incisional biopsy confirmed the diagnosis of calcifying odontogenic cyst. Initial marsupialization was performed for one year, followed by complete enucleation of the lesion with curettage and peripheral osteotomy, along with extraction of tooth 13. In conclusion, COC may clinically and radiographically mimic other more common maxillary lesions. Conservative treatment modalities should be considered to preserve anatomical structures and facilitate future rehabilitation, thereby contributing to the patient's quality of life.

Keywords: Calcifying odontogenic cyst; Odontogenic cysts; Conservative treatment.

INTRODUCTION

Although previous reports of the calcifying odontogenic cyst (COC) have been found in the United States, Russia, and Japan[1], the first detailed description was published by Gorlin et al.[2]. This lesion was identified as a pathologically distinct entity from calcifying odontogenic tumors, being characterized as an oral lesion analogous to Malherbe's calcifying epithelioma. Since then, there have been disagreements regarding its classification and terminology, reflecting the lack of precise knowledge about the pathogenesis and behavior of this cyst. In 1992, the World Health Organization (WHO) classified this lesion as a tumor[3], and later, in 2005, termed it as a calcifying cystic odontogenic tumor[4]. However, in 2017, the WHO reclassified this lesion as COC and included it in the group of developmental odontogenic cysts, dividing it into its intraosseous and peripheral variants[5]. In the latest WHO classification (2022), the COC remained listed under "cysts of the jaws". It is histologically characterized by the presence of ghost cells, which often calcify. The previously essential criterion of an ameloblastoma-like epithelial lining was removed and is now considered only a desirable feature[6].

Statement of Clinical Significance
The calcifying odontogenic cyst is a rare entity that may mimic other maxillary lesions, posing diagnostic and therapeutic challenges. This case highlights the importance of accurate diagnosis and conservative management to preserve anatomical structures and optimize future oral rehabilitation.

COC is a rare lesion with variable behavior, accounting for less than 1% of all odontogenic cysts[7]. There is no sex predilection, and it is generally diagnosed between the second and fourth decades of life. It occurs equally in the maxilla and mandible, although the anterior region is affected in approximately 65% of cases[8,9].

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The clinical and radiographic presentation of COC is diverse, especially in its early stages[1,10]. Swelling in the affected region is the most commonly observed clinical sign. Smaller lesions are usually asymptomatic and incidentally discovered during routine radiographic evaluations[10,11]. Radiographically, it appears as a well-defined unilocular radiolucency of varying sizes, with possible radiopaque foci. In some cases, root resorption, tooth displacement, and cortical perforation may be observed[12,13].

The treatment of this condition is challenging but is generally performed through enucleation and curettage. However, this cyst can reach large dimensions, requiring surgeons to choose an approach that avoids aesthetic and functional sequelae and preserves important anatomical structures[11]. In such cases, marsupialization may be employed prior to surgical removal, aiming to reduce intraluminal pressure and promote peripheral bone formation, thereby decreasing the need for extensive and radical surgery[8,9].

This study aims to report the case of a nineteen-year-old woman diagnosed with COC, who underwent a conservative treatment.

CASE REPORT

A 19-year-old melanodermic female patient presented to the Oral and Maxillofacial Surgery Department of the Monsenhor Flavio D'Amato Municipal Hospital (Sete Lagoas, Minas Gerais, Brazil) with a complaint of progressive, asymptomatic swelling on the right side of the face, with a 12-month duration. During anamnesis, she denied any relevant medical or family history, habits, addictions, or medication use.

Extraoral physical examination revealed obliteration of the nasolabial fold and facial asymmetry in the right malar region (Figure 1A). Intraoral examination showed volumetric enlargement of the right maxillary vestibule, which was firm on palpation (Figure 1B).

Radiographically, a lesion was observed extending throughout the right maxilla, associated with the impacted tooth 13 and the maxillary sinus (Figure 2D). On the panoramic radiograph, the lesion exhibited ill-defined borders and was more clearly delineated on the tomographic examination. It appeared predominantly radiolucent, with areas of radiopacity near the apex of

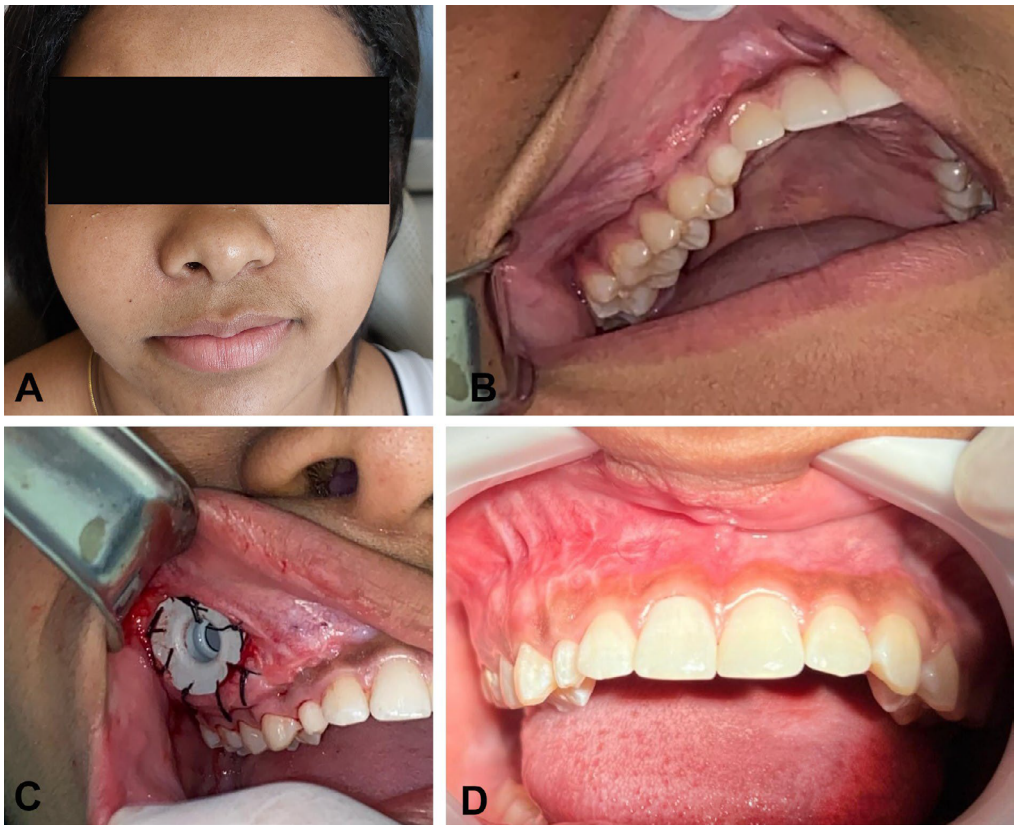

Figure 1. (A) Frontal view of the extraoral examination showing facial asymmetry. **(B)** Intraoral physical examination showing swelling in the vestibular sulcus. **(C)** Rubber device placed in the right vestibular sulcus, aiming the decompression of the lesion. **(D)** Postoperative clinical follow-up at 15 days showing favorable healing, and satisfactory clinical appearance of the surgical site.

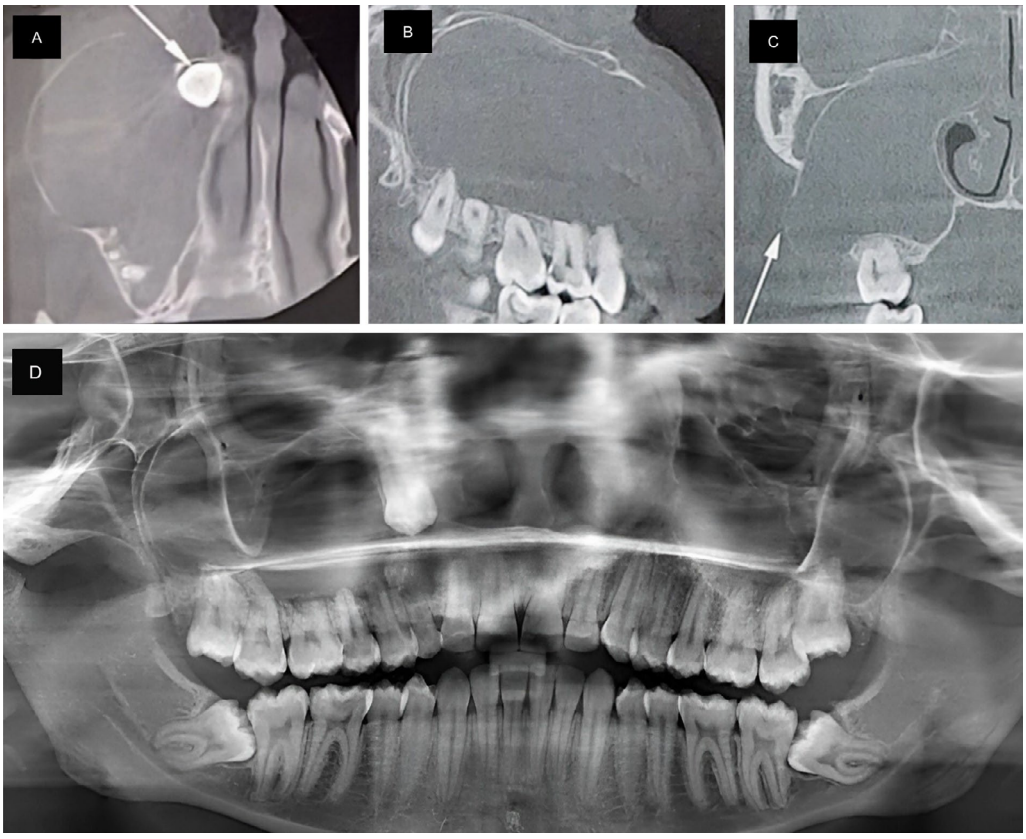

Figure 2. (A) Axial view showing an expansive lesion involving the right maxilla and associated with the impacted tooth 13. **(B)** Sagittal view revealing the lesion extending toward the maxillary sinus with cortical thinning. **(C)** Coronal view, with arrow indicating cortical bone thinning. **(D)** Panoramic radiograph showing an ill-defined radiolucent lesion extending throughout the right maxilla, with a radiopaque area near the apex of tooth 53, displacement of adjacent teeth, and root resorption of teeth 16, 15, 14, and 12.

tooth 53. Additionally, root resorption was noted in teeth 16, 15, 14, and 12.

On the tomographic examination, a unilocular, hypodense, well-circumscribed area was noted, partially surrounded by a hyperdense halo in the right maxillary region, extending from the periapical area of tooth 53 to tooth 18, and associated with the impacted tooth 13. The lesion measured 51.5 mm in the anteroposterior dimension, 34.2 mm mesiodistally, and 33.3 mm superoinferiorly. Moreover, expansion, thinning, and areas of broken vestibular cortical bone were observed. In addition, displacement of tooth 13 toward the laterobasal wall of the nasal fossa was observed, along with external apical third root resorption of the involved teeth (Figures 2 A, B, C).

After correlating the clinical, radiographic, and tomographic findings, the diagnostic hypotheses were unicystic ameloblastoma and dentigerous cyst. An aspiration puncture under local anesthesia was performed to support the diagnosis, yielding a citrine-colored fluid.

At the same time as the incisional biopsy, a rubber device (Figure 1C) was placed and secured in position between teeth 14 and 16 using 4-0 silk sutures, aiming at local decompression prior to surgical enucleation. The patient was instructed to irrigate and clean the drain site with 0.12% chlorhexidine.

The histological sections showed a cystic capsule lined by epithelium containing ameloblast-like cells in the basal layer. Just above, ghost cells were loosely arranged, interspersed with others showing eosinophilic cytoplasm and loss of nuclei, but with preservation of their outline. In some areas, deposition of mineralized material was observed within the ghost cells (Figures 3 A, B, C, D). Adjacent to the epithelial lining, dentinoid material formation was also noted. The diagnosis was calcifying odontogenic cyst.

On clinical and radiographic follow-up after one year of decompression, slight reduction of the lesion with adjacent bone neoformation was observed, and total removal of the lesion was indicated.

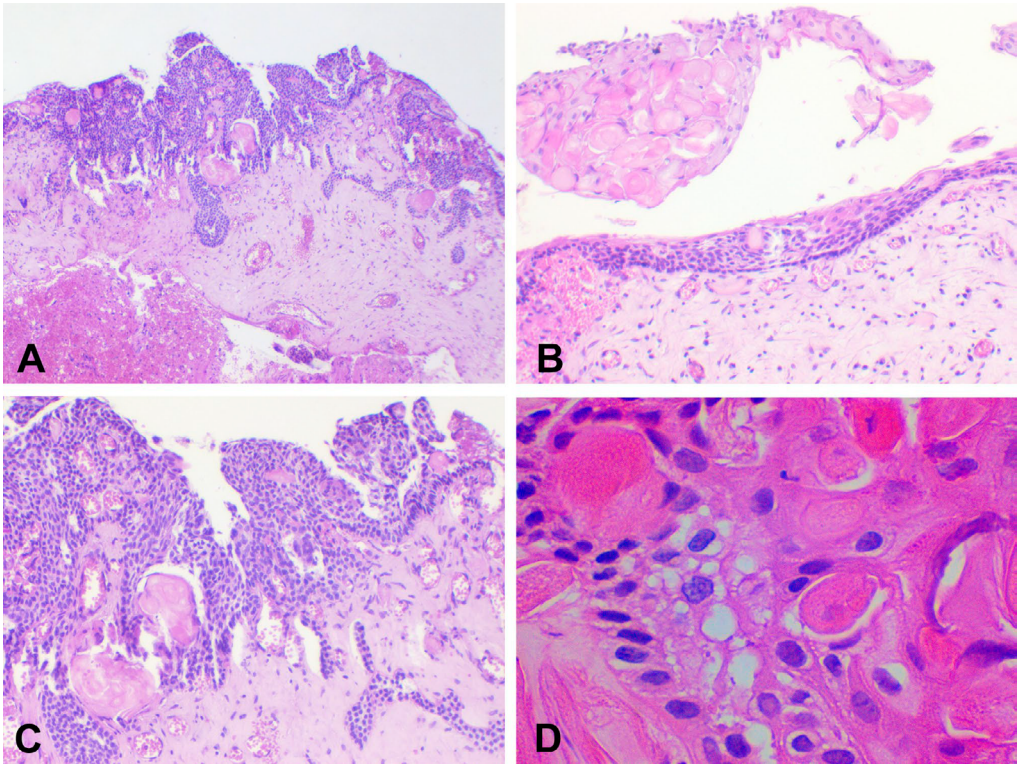

Figure 3. Histopathological sections. **(A)** Fibrous cystic capsule lined by ameloblastic odontogenic epithelium (H&E, 4x). **(B)** Odontogenic epithelium with deposition of amorphous mineralized material (H&E, 10x). **(C)** Proliferation of odontogenic epithelium, loosely arranged cells interspersed with others exhibiting eosinophilic cytoplasm and nuclear degeneration, but with preservation of their outline, referred to as ghost cells (H&E, 10x). **(D)** Palisaded and hyperchromatic columnar ameloblast-like basal cells, with ghost cells (H&E, 40x).

A second surgical procedure was planned and performed under general anesthesia with orotracheal intubation. A mucoperiosteal incision and flap elevation were carried out using an electric scalpel, exposing the entire anterior wall of the maxillary sinus. Then, osteotomy was performed, followed by exposure of the cystic membrane, enucleation by dissection and curettage, and extraction of tooth 13, concluding with continuous suturing. The excised material was again submitted for histopathological analysis, confirming the diagnosis of calcifying odontogenic cyst.

Fifteen days after the surgical procedure, at the postoperative clinical follow-up, the patient showed favorable healing, with stable occlusion, good clinical appearance of the surgical site, and no painful symptoms (Figure 1D). Over time, the postoperative evolution remained satisfactory, and she is currently in a six-month postoperative follow-up, showing good local healing and signs of bone formation in the affected region (Figure 4).

Pulp sensitivity was assessed using thermal tests with tetrafluoroethane spray (Endo-Ice, Maquira – *Indústria de Produtos Odontológicos* LTDA., Paraná, Brazil). Teeth 12 and 53 tested positive, while teeth 14, 15, 16, and 17 tested negative. The Pulp-Tester® Digital VCR-200 device (ODOUS de Deus, Belo Horizonte, Minas Gerais, Brazil) was used for the electric pulp test. Values of 32, 52, 80, 46, and 61 were recorded for teeth 53, 14, 15, 16, and 17, respectively. Based on these results, only tooth 15 was diagnosed with pulp necrosis, and the

Figure 4. Panoramic radiograph six months after the last surgical intervention, showing bone formation in the previously affected region.

patient was subsequently referred to the endodontics team for appropriate management.

Literature research

A comprehensive literature search was conducted using PubMed, Embase, Web of Science, and LILACS, to identify articles related to the conservative treatments in COCs, with no restrictions regarding language or year of publication. Full-article review was performed for eligible articles based on title and abstracts. The review included case reports and case series publications, with exclusion criteria for articles not describing any conservative approach for treating COC.

The following search strategy was used for the four electronic databases: ('calcifying odontogenic cyst' OR 'calcifying cystic odontogenic tumor' OR 'calcifying odontogenic tumor' OR 'Gorlin cyst' OR 'calcifying ghost cell odontogenic cyst' OR 'odontogenic ghost cell cyst' OR 'ghost cell odontogenic cyst' OR 'calcifying ghost cell cyst') AND ('conservative treatment' OR 'marsupialization' OR 'decompression').

All the references obtained in the search were imported to the web software Rayyan (Qatar Computing Research Institute, Doha, Qatar) for duplicate removal, and then continued the selection phases in the same software.

The first search strategy resulted in 175 studies (Figure 5). Following the removal of duplicate records, 100 articles underwent screening based on titles and abstracts (phase-1). Following the initial selection process, 18 articles progressed to the phase-2 of selection. After the selection process, 12 articles were included.

Furthermore, the authors extracted the useful information and data from the full-text articles onto a customized data extraction sheet. There was extracted the following data from each included study: study identification, age, gender, site, side, lesion size, treatment, interval between interventions, outcomes, and follow-up. Table 1 summarizes the analyzed articles[8,10,14-23].

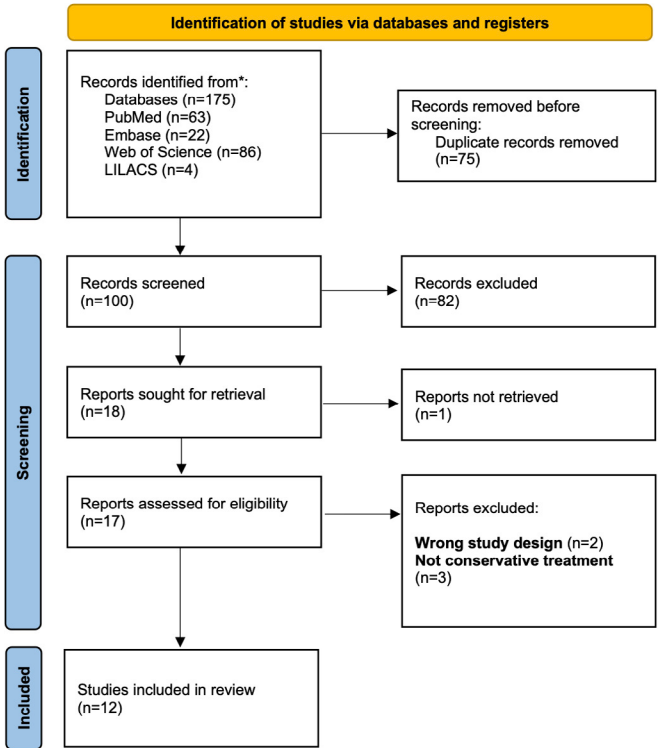

Figure 5. Flowchart depicting the process of search and selection of included studies.

DISCUSSION

COCs may present in a central/intraosseous form, as reported in our study, or in a peripheral/extraosseous form[11]. Arruda et al.[24] conducted a systematic review and found that, clinically, the central variant is usually painless (89.2%), presenting as swelling (76.7%). Although it can be found in any maxillomandibular region, the anterior region is more common, accounting for 65% of cases[25]. Most patients are adults, with involvement in children and adolescents aged 0 to 19 years representing 35.1%[10], and a similar distribution between males and females[24]. It is generally an isolated lesion, but rare bilateral cases have been reported in the literature[26,27]. The present case involves a young female patient, in whom the clinical features included obliteration of the nasolabial fold and facial asymmetry in the right malar region.

However, in 8% of cases, the disease may be completely silent, with no signs or symptoms. Most often, this cyst is discovered incidentally through imaging exams[11]. Radiographically, it usually presents well-defined borders (87%), a mixed appearance (61.1%), associated with bone expansion (81.9%), cortical perforation (61.6%), and association with impacted teeth (57%), generally canines[11]. The present case presented as a unilocular lesion involving the crown of the impacted tooth 13, as well as expansion and thinning of the cortical bone.

The average lesion size at diagnosis was 29.5 mm, ranging from 2 to 90 mm[24]. The reported lesion measured 51.5 mm at its greatest extent, being considered an extensive lesion. Arruda et al.[12] observed that radiolucent, mixed, and radiopaque lesions showed increasing sizes, with averages of 32, 36, and 50 mm, respectively.

Table 1. Clinical features of 12 reported cases treated with conservative approaches.

Study	Age (y)/ gender	Site	Side	Lesion size (cm)	Treatment	Interval between interventions (months)	Outcomes	Follow-up
Moreno-Rodríguez et al.[14]	14/F	Mandible	Anterior	10x4.5	D+E	7	Bone formation; no recurrence	10 years
Sakai et al.[15]	12/F	Maxilla	R	NA	M+E	24	Bone formation; no recurrence	NA
Silva et al.[16]	34/M	Maxilla	R	4.5x5.7	D+E	5	Bone formation; no recurrence	13 months
Alarcón et al.[17]	12/M	Mandible	Anterior	NA	D+E	12	Bone formation; no recurrence	9 years
Emam et al.[18]	36/M	Maxilla	R	5.9x3.6x4.6	D+E	9	Bone formation; no recurrence	9 months
Kim et al.[19]	24/M	Maxilla	L	NA	D+E	5	Bone formation; no recurrence	36 months
Samir et al.[10]	17/ NA	Maxilla	R	2.7x2.1	D+E	9	Beginning of bone formation; no recurrence	9 months
Moraes et al.[8]	11/M	Mandible	R	NA	M+E	4	Bone formation; no recurrence	>10 years
Moraes et al.[20]	15/F	Maxilla	R	NA	D+E	8	Bone formation; no recurrence	4 years
Masuda et al.[21]	8/F	Mandible	R	NA	M+E	4	Bone formation; no recurrence	7 years
	20/M	Mandible	Anterior	NA	M+E	21	Bone formation; no recurrence	24 months
Souza et al.[22]	14/M	Mandible	R	1.5	M+E	NA	Bone formation; no recurrence	10 years
	8/F	Maxilla	R	NA	M+E	7	Bone formation; no recurrence	5 years
Santana et al.[23]	13/F	Maxilla	L	7.5	D+E	6	Bone formation; no recurrence	1 year

L: Left; R: Right; M: Marsupialization; D: Decompression; E: Enucleation; NA: Not available.

Thus, the deposition and number of calcifications may be related to the lesion size, but this hypothesis is still not well elucidated in the literature[12].

When presenting as mixed, it may exhibit three radiopacity patterns:

1. Resembling salt-and-pepper spots;
2. A diffuse and uniform amorphous pattern; and
3. A crescent moon-like appearance[11].

COCs have clinical and radiographic presentations similar to various other odontogenic lesions, such as dentigerous cyst, ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibro-odontoma, and calcifying epithelial odontogenic tumor[1,10]. Therefore, an accurate histopathological examination is necessary for confirmation[13]. Histologically, this cyst is generally composed of cuboidal or columnar cells resembling ameloblasts. The suprabasal layer is composed of loosely arranged epithelial cells resembling the stellate reticulum of an ameloblastoma[25]. The most significant feature is the presence of ghost cells, in varying amounts and distributions[10]. These are altered eosinophilic epithelial cells that retain basic nuclear and cellular outlines but lack cellular vitality. Occasionally, ghost cells may become calcified, thus losing their cellular configuration and resulting in foci of calcified keratin[11]. Immunohistochemical analysis showed moderate to high expression of amelogenin (92.3%) and cytokeratin 6 (77%)[28]. Other odontogenic lesions containing ghost cells that should be considered as possible differential diagnoses are the dentinogenic ghost cell tumors and the ghost cell odontogenic carcinoma[7,12].

Reports of COC associated with other lesions are also found in the literature, such as orthokeratinized odontogenic cysts, ameloblastomas, myxofibromas, ameloblastic fibromas, ameloblastic fibro-odontomas, and adenomatoid odontogenic tumors[29]. However, odontoma is the lesion most commonly associated with COC, found in 24% of cases[29]. Three theories have been proposed for the pathogenesis of the association of these lesions. The first suggests that they are found together by coincidence, since other lesions have also been associated. The second indicates that COC is formed secondarily from the odontogenic epithelium that initially formed the odontoma. The third suggests that some authors consider the association of these lesions to be a distinct and unique clinical entity[30]. Our case presented as an isolated lesion, without other associated cysts or tumors.

Treatment strategies vary and depend on the lesion's aggressiveness[11]. Conservative surgery, using enucleation, is the most common treatment for COCs, employed in 70% of cases[24]. Furthermore, Yeh et al.[9] suggested marsupialization or decompression for lesions larger than 4 cm, aiming to reduce the size and facilitate treatment, decreasing the need for more extensive and radical surgeries[8], and it has been used in 0.7% of cases[24]. In our patient, decompression was chosen due to the large extent of the lesion. Surgical resection is a more aggressive treatment and was performed in less than 5% of cases. However, the recurrence rate is low, at about only 4%[24]. Therefore, a conservative approach is favored, and the prognosis is good. Despite this, Samir et al.[10] suggest that COCs should be monitored for 10 years.

The reviewed cases demonstrate that conservative approaches followed by enucleation are effective treatment strategies for large COCs. The patients' ages ranged from 8 to 36 years, which supports the preference for less invasive procedures aimed at preserving bone and vital structure, and teeth. Moreover, the clinical outcomes were consistently favorable, with bone regeneration observed in nearly all cases. The interval between treatments ranged from 4 to 24 months, reflecting the individualized nature of treatment planning bases on lesion behavior and radiographic evolution.

CONCLUSION

COC is a rare developmental odontogenic cyst that can clinically and radiographically mimic other more common maxillary entities. Conservative treatment should be considered to preserve anatomical structures and thus enable subsequent rehabilitation, maintaining quality of life. Our case demonstrated the clinical, radiographic, and histopathological features of a COC in a young patient, showing the success of the treatment employed.

AUTHORS'CONTRIBUTIONS

RMOM: conceptualization, data curation, formal analysis, writing – original draft, writing – review& editing. VFSF: data curation, formal analysis, writing – review & editing. LRDM: data curation, formal analysis, writing – review & editing. CHA: data curation, formal analysis, writing – review & editing. LOF: formal analysis, investigation, writing – review & editing. MSN: formal analysis, investigation, writing – review & editing. PHAT: conceptualization, data curation, formal analysis, project administration, 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: The study was conducted in accordance with the ethical principles established by the Institutional Research Ethics Committee, ensuring compliance with the current ethical and scientific guidelines. Approval protocol: 79800624.0.0000.8164.

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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