Calcifying odontogenic cyst associated with complex odontoma treated with a conservative approach: a case report

Cisto odontogênico calcificante associado a odontoma complexo tratado com abordagem conservadora: relato de caso

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ABSTRACT
Calcifying odontogenic cysts (COCs) are uncommon benign lesions arising from the odontogenic epithelium which are currently considered cysts. Histopathologically, it presents as an epithelial lining with ameloblastic characteristics, in addition to ghost cells with tendency to undergo calcifications. The objective of this study was to describe a case of a calcifying odontogenic cyst associated with odontoma, which was treated with a conservative two-step approach. A 29-year-old female presented with increased volume on the right side of the face with a lesion detected at the bottom of the buccal groove, extending from tooth 53 to 16. Radiographically, a wide, unilocular lesion was observed, involving tooth 13 that was impacted. Histopathological examination was consistent with COC associated with complex odontoma. No recurrence was seen at 6-month follow-up. The case addresses the importance of anatomopathological diagnosis of the lesion, since characterization of the cyst is fundamental to treatment.

Keywords: calcifying odontogenic cyst, decompression, odontoma.
e unilocular, envolvendo o dente 13 que foi impactado. O exame histopatológico foi consistente com o COC associado a odontoma complexo. Não se observou recidiva no seguimento de 6 meses. O caso aborda a importância do diagnóstico anatomopatológico da lesão, uma vez que a caracterização do cisto é fundamental para o tratamento.

**Palavras-chave:** calcificação do cisto odontogénico, descompressão, odontoma.

### 1 INTRODUCTION

Calcifying odontogenic cysts (COCs) were previously described by Gorlin et al. (1962) and Gold (1963). They are rare, usually unique lesions, accounting for about 1%–2% of odontogenic cysts and tumors [1], [2], [3]. Various terminologies have already been designated to refer to this lesion [1] that was reincorporated into the cyst session in the latest World Health Organization (WHO) classification for head and neck tumors in 2017 [4].

Representing a group of lesions with various clinical and pathological characteristics [5], the COC is predominantly intraosseous, expandable, asymptomatic, and in most cases, found in the anterior region of gnathic bones, with no preference for sex [2]. Radiographically, it presents itself as a radiotransparent area, more commonly unilocular, and with the visualization of irregular radiopaque foci being a typical peculiarity [6].

The presence of phantom cells in the cystic lining is a distinctive histopathological characteristic of COCs, which have a tendency to coalesce and form extensive calcified masses [5]. In addition, an epithelial lining resembling ameloblastoma is observed, which may be similar to the starry reticulum of the enamel organ [7]. Eosinophilic structures similar to dysplastic dentin can also be found adjacent to the odontogenic epithelium [6].

COCs occur alone or in association with other odontogenic tumors, such as ameloblastoma, adenomatoid odontogenic tumor (TOA), and ameloblastic fibroma [7]. In addition, it is related to dental tissue formation that resembles odontoma, an association most commonly reported [1].

The surgical approach to enucleation is most commonly used in COCs [8]. However, depending on the lesion size and the need to preserve anatomical structures, two surgical procedures can be performed: one for decompression and the other for total removal of the lesion [9]. Advantages such as new bone formation have been reported with this technique [10].

This case report presents COC associated with a complex type odontoma, treated in two surgical stages, demonstrating a diversified presentation of this lesion. We also describe the clinical and histopathological characteristics of the lesion.
2 CASE REPORT

This study followed the rules of the Research Ethics Committee of the Federal University of Ceará and was approved for execution under CAAE number: 45170721.5.0000.5054.

A 29-year-old female visited the Stomatology Clinic at the Federal University of Ceará (UFC) with increased volume on the right side of the maxilla. Intraoral examination revealed a deep groove erasure in the region of teeth 53 and 16; the mucosal lining was smooth, teleangiectatic, and softened on palpation (Fig. 1). The patient had no remarkable medical history. Panoramic radiography showed well-defined edges extending to the right maxillary sinus permeated by irregular radiopaque outbreaks.

Fig. 1. Initial clinical appearance. Pre-operative intraoral appearance, showing increased volume in the bottom of the right vestibule and absence of the right upper permanent canine.

Source: Division of Oral and Maxillofacial Surgery and Traumatology, Walter Cantídio University Hospital, Federal University of Ceará.
The tooth 13 was present inside the lesion (Fig. 2). Considering the clinical hypothesis of a dentigerous cyst, surgical decompression of the lesion was performed. After 8 months, an excisional biopsy was performed in surgery under general anesthesia in a hospital setting. An intraoral approach was performed through a modified Neumann incision, osteotomy to access the interior of the right maxillary sinus, identification and complete enucleation of the lesion and removal of the associated tooth, followed by surgical curettage (Fig 3). The specimen was sent to the UFC Oral Pathology Laboratory.
Fig. 3. Transoperative. (A) Initial appearance; (B) Surgical access via a modified Neumann approach using two vertical incisions and an intrasulcular incision. (C) Access to the right maxillary sinus and identification of the lesion. (D) Appearance after complete removal of the lesion and curettage.

Source: Division of Oral and Maxillofacial Surgery and Traumatology, Walter Cantídio University Hospital, Federal University of Ceará.

Microscopic examination showed a cystic cavity partially covered by ameloblastomatous odontogenic epithelium in which the cells of the basal layer appeared similar to ameloblasts (columnar, hyperchromatic, and with inverted polarity) with loose overlying layers, reminiscent of the strange starry reticulum. Inside the epithelial component, ghost cells were also observed, some of which were calcified. The capsule was of the dense fibrovascular type with the presence of an eosinophilic matrix compatible with dentinoid material, islands of odontogenic epithelium, and an abortive enamel matrix (Fig. 4). Based on the above findings, the final diagnosis of COC associated with complex odontoma was established. We performed a panoramic radiograph 6 months after excisional biopsy, and observed new bone formation (Fig. 5). The patient was followed up, and no recurrence was observed.
Fig. 4. Histopathological characteristics of the lesion. (A) Ameloblastic odontogenic epithelial lining and phantom cell masses (Hematoxylin-eosin, original magnification 200×); (B) Large eosinophilic ghost cells (Hematoxylin-eosin, original magnification 200×); (C) Cystic capsule with eosinophilic dentinoid material (Hematoxylin-eosin, original magnification 100×); (D) Odontoma associated with a lesion with enamel deposits (Hematoxylin-eosin, original 50× magnification).

Source: Oral Pathology Laboratory of the Federal University of Ceará.

Fig. 5. Final radiography. Panoramic radiograph 6 months after surgical excision, showing no signs of recurrence.

Source: Division of Oral and Maxillofacial Surgery and Traumatology, Walter Cantídio University Hospital, Federal University of Ceará.
3 DISCUSSION

The classification on the neoplastic or cystic nature of COC has been modified over time. This modification directly impacts the treatment plan that changes depending on the nature of the injury [10]. However, the latest WHO classification considers COC as a pathology of a cystic nature, while phantom cell dentinogenic tumor being its solid variant [4].

Clinically, COCs usually affect patients in the 2nd and 4th decades of life, with the anterior region of the mandible being the most affected [11]. Most patients with COCs (89.2%) show only slow and asymptomatic growth in the region of the gnathic bones [8]. In the present case, despite the involvement of the maxillary sinus, the patient did not report any symptoms, which is common in other odontogenic cysts and tumors, such as dentigerous cysts, central giant cell granulomas, and odontogenic keratocysts [2], demonstrating that the clinical characteristics are not sufficient for the definitive diagnosis of the lesion.

Radiographically, COCs commonly present a mixture of radiolucent and radiopaque areas with well-defined edges and unilocular appearance, as in the case reported. Due to the radiographic detection of calcified material, COCs can occur in other odontogenic tumors such as the TOA, fibroma central ossifier, and odontoma. Thus, as the lesion included tooth 13, extended into the amelo-cementary junction, and was located in the anterior region of the maxilla, we made a diagnosis of TOA [8]. Even though radiography is a tool for the diagnosis of COCs, it has insufficient pathognomonic characteristics to guide the final resolution of the case, which is dependent on histopathological examination [12].

In addition to dentinoid tissue in the fibrous capsule, microscopic findings characteristic of COCs include cystic lining containing anucleated and slightly eosinophilic cells, called ghost cells, which may be calcified [1].

Phantom cells have an epithelial origin and are controversial in nature. These cells can be found in odontogenics such as odontomas, in addition to craniopharyngioma and pilomatrixoma, which are non-odontogenic in nature. It is believed that its formation occurs as a result of an abnormal keratinization and as it presents a positive marker for a capillary protein called α-keratin, it is necessary that it can represent a differentiation of the hair in several varied local lesions [13].

Some authors have proposed the division of COCs into three variants: simple unicystic, unicystic odontoma producer, which characterizes the present case, and proliferative ameloblastomatous [3]. Arruda [8] reviewed 367 cases of COCs and found that odontogenics were present in 28.6% of home cases, with odontoma being the most common (86.8% of clinical reports). We detected a complex type of odontoma associated with COC; however, Han [14]
reported a COC associated with a compound type odontoma, with well-developed denticles in the COC structure.

Theories have been proposed to explain the association between the COC and odontomas. It is suggested that the lesions appear juxtaposed and coincidentally, since COCs can occur in association with other odontogenic tumors such as TOA, ameloblastic fibroma, and ameloblastoma. In other hypotheses, COCs were secondary to the odontogenic epithelium in the formation of the odontoma; however, the formation of the odontoma from the epithelial lining of the COC was also considered. In addition, they may be different clinical entities [1].

Surgical treatment of COCs is enucleation followed by curettage, and post-surgery recurrence is rare [10], [5]. We chose to perform decompression first and then proceed with enucleation and curettage due to the extensive size of the lesion and the patient's age. In this way, we can guarantee the preservation of noble structures, as well as those responsible for the masticatory function, which is affected by the injury [15], [10]. In the present case, decompression resulted in a reduction in the intra-and extra-oral size of the lesion after 8 months.

4 CONCLUSION

COCs show clinical and radiographic similarities with other odontogenic lesions, which indicates the need for histopathological examination for conclusive diagnosis. Microscopic characterization of COCs is important for establishing a treatment plan, since it is a rare tumor variant. In addition, a conservative two-step approach (decompression followed by enucleation and curettage) is a safe option that does not require surgical reconstruction.

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Conflict of interest

None

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