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Rare Association of a Calcifying Odontogenic Cyst and an Odontoma: A Case Report and Diagnostic Discussion

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Abstract: The calcifying odontogenic cyst (Gorlin cyst) is a rare benign odontogenic lesion that may be associated with other developmental anomalies, particularly an odontoma. [4]

We report the case of an 18-year-old patient presenting with a progressive, hard, non-inflammatory, and painless swelling of the right maxilla, associated with palatal bulging and cortical expansion. [1] Computed tomography revealed two well-defined radiopaque lesions closely related to the dental structures and associated with impacted permanent teeth. The main differential diagnoses included cementoblastoma, odontoma, and calcifying odontogenic cyst. Histopathological examination of the biopsy specimen established the diagnosis of a calcifying odontogenic cyst associated with an odontoma, characterized by an odontogenic epithelial lining exhibiting ameloblastic differentiation, ghost cells, dentinoid material, and rudimentary dental structures.

This case highlights the challenges of preoperative diagnosis of calcified odontogenic lesions and emphasizes the importance of clinicoradiological and histopathological correlation. [4]

Keywords: Calcifying odontogenic cyst; Odontoma; Gorlin cyst; Maxillary lesion; Differential diagnosis; Histopathology.

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1 Introduction

The calcifying odontogenic cyst (COC), also known as the Gorlin cyst, is a rare benign odontogenic lesion, accounting for less than 1% of all odontogenic cysts. [3,7]

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First described by Gorlin et al. in 1962, it is characterized histologically by an odontogenic epithelial lining exhibiting ameloblastic differentiation and, pathognomonically, by the presence of ghost cells, which may undergo secondary calcification. [1,2,6]

This lesion may occur as an isolated entity or in association with other odontogenic developmental abnormalities, particularly an odontoma, which is reported in 20–35% of cases across different series. The association of a calcifying odontogenic cyst with an odontoma occurs predominantly in young patients and most commonly presents as a progressive, asymptomatic bony swelling or as a disturbance in tooth eruption. [4,5,9]

Radiologically, the findings are often nonspecific, typically revealing a well-defined mixed radiolucent–radiopaque lesion in close proximity to adjacent or impacted teeth. [9,10]

These features may raise several differential diagnoses, including cementoblastoma, an isolated odontoma, and other calcifying odontogenic tumors. Definitive diagnosis therefore relies primarily on histopathological examination. [10,12,13]

We report a case of a calcifying odontogenic cyst associated with an odontoma, presenting as a maxillary swelling in an 18-year-old patient. This case illustrates the challenges of preoperative diagnosis and highlights the importance of clinicoradiological and histopathological correlation.

2 Case presentation

An 18-year-old patient with an unremarkable medical and surgical history presented with a hard, progressively enlarging, painless swelling of the right maxilla, without clinical signs of inflammation. Intraoral examination revealed palatal expansion associated with expansion of the buccal cortical plate. No erythema, local warmth, or purulent discharge from the adjacent teeth was noted. Two carious deciduous teeth were present in the affected region. The clinical findings were suggestive of a slowly expansile intraosseous lesion, most likely of odontogenic origin, warranting further radiological assessment.

2.1 Figure



Figure 1. Clinical examination revealed expansion of the buccal cortical plate, with no erythema, local warmth, or purulent discharge from the adjacent teeth. Two carious deciduous teeth were present in the region of the lesion.

3 Imaging and differential diagnostic

Maxillofacial computed tomography (CT) revealed two well-circumscribed, intraosseous radiopaque lesions within the maxilla, in close proximity to the adjacent dental structures. The first lesion was oval in shape, measuring approximately 14.5 × 17.5 × 17.5 mm. It involved the alveolar and palatal processes and was surrounded by a peripheral hypodense rim. The lesion caused expansion of the maxillary bone, with cortical thinning and focal areas of cortical perforation. A second lesion with similar radiological characteristics measured approximately 11 × 6 × 11.5 mm and was also in close contact with the dental roots. It was associated with cortical thinning, without any definite evidence of cortical breach. The examination also revealed multiple impacted permanent teeth within the maxilla.

The radiological findings primarily suggested a benign calcifying odontogenic lesion. The main differential diagnoses included cementoblastoma, odontoma, and calcifying odontogenic cyst with intralesional calcifications.

Cementoblastoma was considered because of the well-defined radiopaque appearance and close relationship with the dental roots. However, the absence of clear evidence of root fusion made this diagnosis less likely.

Odontoma was also an important diagnostic consideration, particularly in view of the associated tooth impaction and the presence of well-defined calcified structures.

Given the well-circumscribed nature of the lesion, its presentation in a young patient, and the presence of intralesional calcified components, ...

3.1 Figure

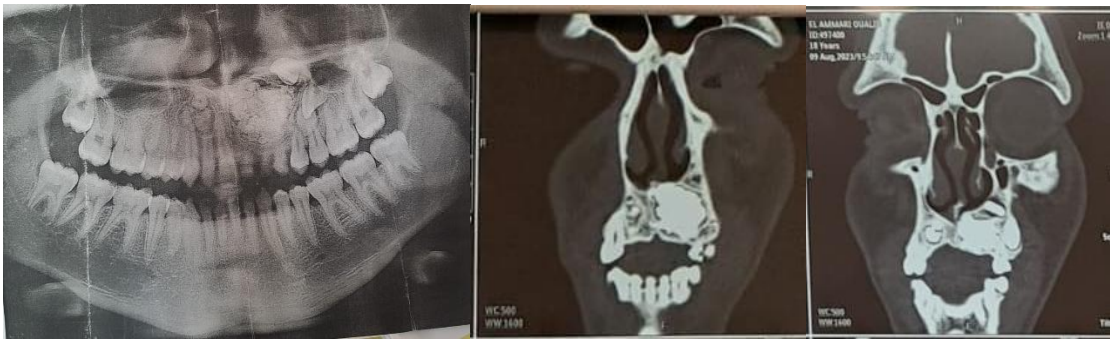


Figure 2. Imaging of the lesion on panoramic radiography and computed tomography (CT)Two well-circumscribed, intraosseous radiopaque lesions were identified within the maxilla.

4 Management

In view of the clinical and radiological findings, a biopsy of the right maxillary lesion was performed. The specimen was submitted for histopathological examination to establish the definitive diagnosis. This step was essential, as imaging alone could not reliably distinguish between the various calcifying odontogenic lesions that may present with similar clinical and radiological features. The diagnostic approach therefore relied on close correlation of the clinical, radiological, and histopathological findings.

Surgical management

Treatment consisted of curettage, fenestration, and decortication of the lesion. The procedure was performed under general anesthesia with endotracheal intubation. Extraction of teeth 61, 62, and 63 was followed by an extended sulcular incision from tooth 15 to tooth 25, after infiltration with epinephrine-containing lidocaine.

Following mucoperiosteal flap elevation, two maxillary lesions were exposed: a right-sided lesion with an intact cortical plate and a left-sided lesion associated with buccal and palatal cortical expansion. Curettage, fenestration, and decortication were performed on the radiopaque lesion, which was suggestive of an odontoma. The immediate postoperative course was uneventful, and the patient recovered from general anesthesia without complications.

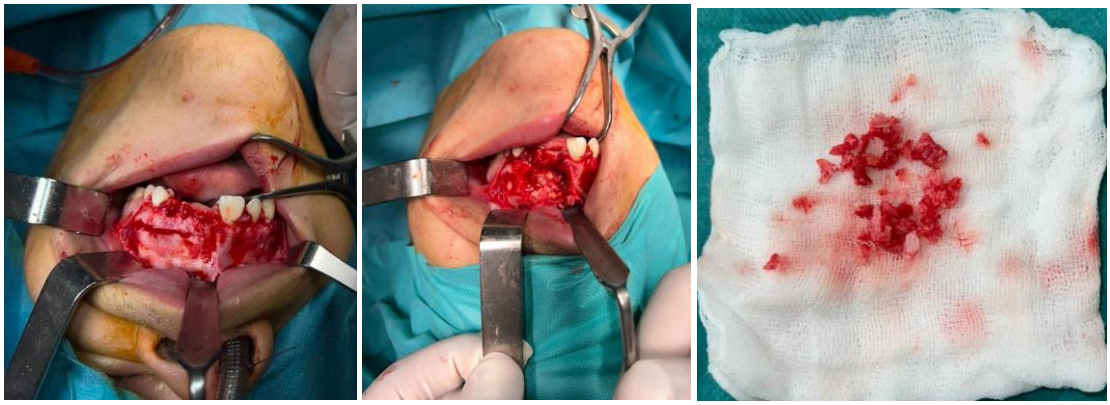


Figure 3. Aspect per-operatoire

5 Anatomo-pathologie

Histopathological examination revealed fragmented tissue specimens comprising an atrophic stratified squamous epithelial lining, with focal epithelial proliferations exhibiting marked ameloblastic differentiation. The basal cell layer consisted of columnar cells overlying a stellate reticulum-like layer, consistent with odontogenic differentiation. Prominent keratinization with numerous large, anucleate ghost cells was also observed, together with an amorphous eosinophilic calcified material corresponding to dentinoid. In addition, multiple rudimentary tooth-like structures were identified, composed of enamel, dentin, and cementum, with dental pulp present in some areas. Scattered trabeculae of mature, regular cancellous bone were also noted.

Taken together, these histopathological findings established the diagnosis of a **calcifying odontogenic cyst associated with an odontoma**.

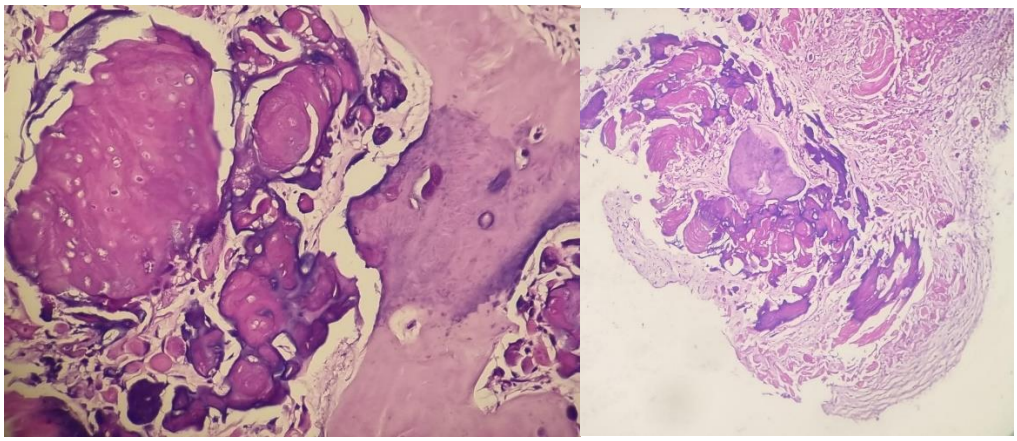


Figure 4. Histopathological features of a calcifying odontogenic cyst associated with an odontoma.

6 Disscussion

Calcifying odontogenic lesions comprise a heterogeneous group of benign entities characterized by considerable clinical, radiological, and histological variability [2]. Their diagnosis can be particularly challenging, as they often share common features, including slow growth, progressive bone expansion, internal calcifications, and a close relationship with impacted or adjacent teeth. In this context, the present case illustrates the difficulty of preoperative diagnosis and underscores the critical importance of histopathological examination.

The patient's clinical presentation was suggestive of a benign, slowly progressive lesion. The swelling was hard, painless, and non-inflammatory, with localized maxillary deformity characterized by palatal bulging and cortical expansion. Such a presentation is typical of several odontogenic lesions, which may remain asymptomatic for an extended period before being detected because of facial asymmetry, disturbances in tooth eruption, or incidental radiological findings. The absence of signs of infection, suppuration, or acute pain initially argued against a primary odontogenic inflammatory process, despite the presence of carious deciduous teeth in the affected region.

Imaging played a key role in guiding the diagnostic process. CT demonstrated well-circumscribed, radiopaque lesions that were relatively small but associated with significant osseous remodeling. The peripheral hypodense rim, expansion of the maxillary bone, and cortical thinning were suggestive of a benign expansile process. However, the radiological appearance remained nonspecific. Indeed, several odontogenic lesions may present as mixed radiolucent–radiopaque lesions, with variable internal architecture depending on their degree of maturation and mineralization. The close relationship between the lesions and impacted teeth or dental roots further supported an odontogenic origin but was insufficient to distinguish between the possible entities.

Cementoblastoma was one of the main differential diagnoses. This rare benign odontogenic neoplasm predominantly affects adolescents and young adults and develops in association with a tooth root, to which it is often intimately fused. Radiographically, it typically appears as a well-defined radiopaque mass surrounded by a radiolucent rim, with possible root resorption or root fusion. In the present case, although the lesion was radiopaque and located near dental structures, histopathological examination did not reveal the characteristic features of cementoblastoma, allowing this diagnosis to be excluded.

Odontoma was another important diagnostic consideration. It is the most common odontogenic tumor and is widely regarded by many authors as a hamartoma rather than a true neoplasm. Odontomas are classified as compound or complex. The compound type consists of multiple tooth-like structures, whereas the complex type presents as a disorganized calcified mass. The presence of multiple rudimentary tooth-like structures composed of enamel, dentin, and cementum strongly supported the diagnosis of a compound odontoma. However, an isolated odontoma could not account for all the histopathological findings, particularly the presence of a cystic epithelial component exhibiting ameloblastic differentiation and prominent keratinization. This distinctive morphological association ultimately established the diagnosis of a calcifying odontogenic cyst associated with an odontoma. [10]

The calcifying odontogenic cyst (COC) is a rare odontogenic lesion that may exhibit considerable clinical and radiological variability [1]. It predominantly affects young patients and may present as bony swelling, be associated with an impacted tooth, or be detected incidentally on imaging. Histologically, it is classically characterized by an odontogenic epithelial lining, foci of ameloblastic differentiation, and, most notably, ghost cells that may undergo secondary calcification. [1,6]

In some cases, the lesion may be associated with other odontogenic developmental abnormalities, particularly odontomas. Although uncommon, this association is consistent with the developmental nature of the lesion and accounts for the complexity of its histopathological presentation. [4,5]

The significance of this case lies in the coexistence of distinct pathological components within the same lesion. [10] The cystic component accounted for the observed epithelial differentiation, whereas the odontomatous component explained the presence of well-defined rudimentary dental structures. This association complicates the initial radiological diagnosis and highlights the importance of looking beyond the radiopaque appearance of a maxillary lesion. [12]

In clinical practice, this case emphasizes that histopathological confirmation is essential before establishing a definitive diagnosis, particularly when imaging suggests several competing diagnostic possibilities. [13]

Regarding treatment, biopsy followed by complete surgical excision is a standard approach for benign odontogenic lesions of this type. The prognosis is generally favorable following appropriate surgical management, provided that complete removal is achieved and clinical and radiological follow-up is maintained. [9,11] An accurate diagnosis is also essential for follow-up planning, as it guides the surgical approach, determines the extent of resection, and informs surveillance for potential recurrence. In the present case, the diagnostic workup established concordance between the clinical, radiological, and histopathological findings, leading to a reliable final diagnosis.

In conclusion, this case describes a benign odontogenic lesion of the maxilla whose clinical and radiological presentation initially raised several differential diagnoses, particularly cementoblastoma and odontoma. Histopathological examination established the diagnosis of a calcifying odontogenic cyst associated with an odontoma. This case highlights the importance of a multidisciplinary approach and clinicoradiological–histopathological correlation in the diagnosis of calcifying odontogenic lesions. [3]

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