

Early Detection of a Calcifying Cystic Odontogenic Tumor associated with Odontoma

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Abstract:

A 15-year-old male patient was referred for diagnosis and treatment of a radiographic lesion located in the anterior region of the maxilla. Clinically, the patient presented a prolonged retention of the element 63 and absence of the element 23. No swelling around the alveolar ridge was observed. The radiographic findings were characterized by few tooth-like radiopaque structures surrounded by a radiolucent halo around the impacted element 23. The clinical hypothesis was established as dentigerous cyst associated with compound odontoma. The excisional biopsy was performed keeping the impacted tooth in place, to which an orthodontic bracket was bond for posterior traction. The surgical piece was taken to histopathological examination and the final diagnosis was calcifying cystic odontogenic tumor (CCOT) associated with compound odontoma. The calcifying cystic odontogenic tumor (CCOT) is a rare benign neoplastic lesion that represents less than 2% of all cysts and odontogenic tumors; it is usually found at the canine region and is associated with odontoma. Knowledge on the lesion characteristics, a detailed clinical exam, and complementary exams are essential to a differential diagnosis aiming to provide appropriate treatment for the patient, which currently consists of complete enucleation, with good prognosis. The 12-month follow-up shows an injury-free patient

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INTRODUCTION

The calcifying cystic odontogenic tumor (CCOT) is a rare lesion that represents less than 2% of all cysts and odontogenic tumors^{1,2}. There is still discussion in the literature regarding its nomenclature: some authors argue that this lesion does not present itself as a neoplasia with potential growth; however, other authors consider this lesion a tumor with cystic formations, with capacity to infiltrate or even become malignant, being considered a neoplasia^{3,4}. In 2005, the World Health Organization Classification of Head and Neck Tumors described it as a benign cystic neoplasia and introduced the new nomenclature: Calcifying Cystic Odontogenic Tumor (CCOT)⁵.

The CCOT is frequently found associated with other odontogenic tumors, being odontoma the most commonly associated lesion. It is presented as a predominately intraosseous lesion, but it can occasionally be manifested peripherally. It occurs with the same frequency both in the maxilla and in the mandible bones. Most cases (approximately 65%) are found in young adults (second and third decades of life), in the canine and incisor regions, with no gender predilection⁶.

Usually, a unilocular radiolucent image is revealed by radiographic exams on CCOT. Irregular internal calcifications may be found and, occasionally, this lesion may be associated with impacted tooth⁷. The characteristic histopathological aspect of this lesion is the presence of ghost cells, which are passive of calcification, and the presence of epithelial lining similar to that of the ameloblastoma⁸.

The present report aims to describe a CCOT case associated with odontoma and review the specific literature on this subject.

CASE REPORT

A 15-year-old male sought a routine appointment and orthodontic evaluation in a Dental Clinic. The clinical exam showed the absence of the element 23 and a prolonged retention of the corresponding primary tooth. No vestibular volume alteration or any other symptom was reported.

The panoramic and intra-oral periapical radiographic images (Figure 1) showed one radiolucid, unilocular, well-delineated lesion and one unilocular radiolucency with a radiopaque halo around the crown of the developing and impacted canine. Teeth-like structures of different shapes and sizes were also

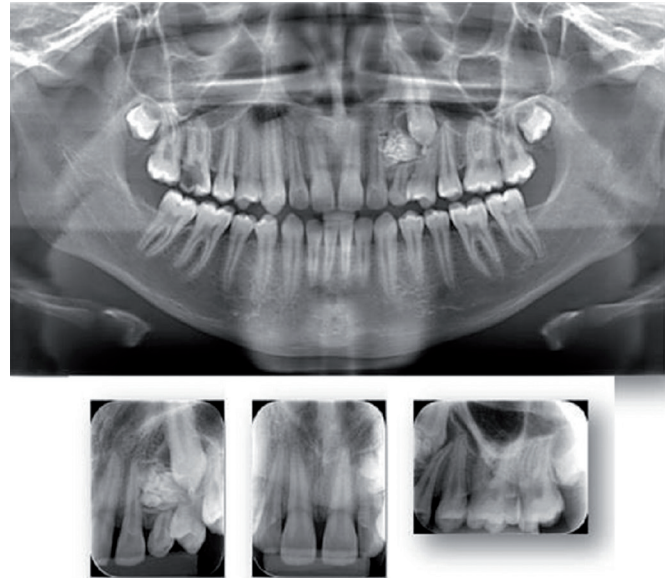


Figure 1. The panoramic and intra-oral periapical radiographic images showed a unilocular, radiolucid and well-delineated lesion close to the canine tooth.

observed. There was no resorption except for the normal on the root of element 63. No root movement/deviation/migration was noticed.

The patient was referred to the Department of Stomatology, College of Dentistry, University of São Paulo for evaluation, where a cone-beam computed tomography of the maxilla (Figure 2 e Figure 3) was requested in order to obtain improved details of the lesion characteristics and canine location.

Based on the clinical, radiographic and tomographic examinations, a differential diagnosis of compound odontoma associated with dentigerous cyst was considered. Thereby the treatment plan consisted of an excisional biopsy (Figure 4) with complete curettage of the lesion under general anesthesia due to the proximity of the lesion to the nasal cavity, extraction of the element 63, and an orthodontic button attachment for further traction of the permanent canine. After the curettage, a healthy adjacent bone was observed.

The surgical specimen consisted of eight yellow hard tissue fragments and irregular surfaces measuring 15 x 14 x 05 mm altogether, and one hard tissue fragment associated with a soft fragment grayish in color, of rubbery consistency and irregular shape, with surface measuring 12 x 08 x 05 mm (Figure 4).

Histopathological examination revealed deposit of dental tissue, majority represented by a tubular dentin. Tissue similar to cementum, dental pulp and, in great proportions, enamel matrix were also present (Figure

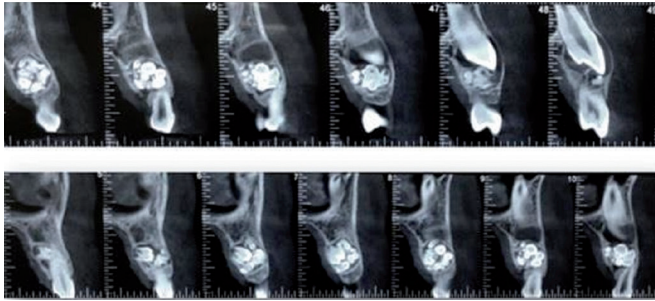


Figure 2. Sagittal cross-sectional tomography in anterior maxilla showed a radiopaque image with a tooth aspect in different sizes and shapes and a radiolucent lesion with cystic aspect.



Figure 3. Cone beam computed tomography reconstruction demonstrating the localization of the lesion.

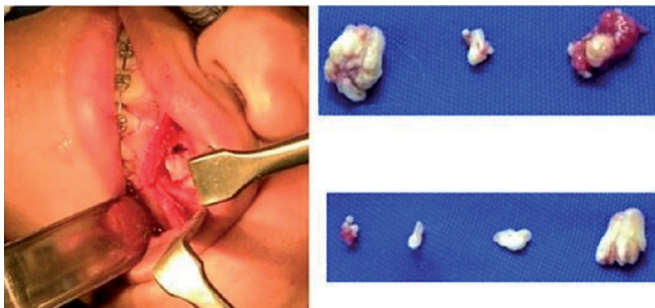


Figure 4. Trans operative and macroscopic aspect of the lesion.

5). In association with these tissues an odontogenic epithelium was nothed, with ameloblastous aspect and presence of ghosts cell masses. Foci of calcification along these cells were also present (Figure 6). The diagnosis of calcifying cystic odontogenic tumor associated with odontoma was made.

Over the 12-month follow-up, the patient did not present any complaints or signs of recurrence. Currently, the patient is undergoing orthodontic treatment for dental correction and traction of the permanent tooth.

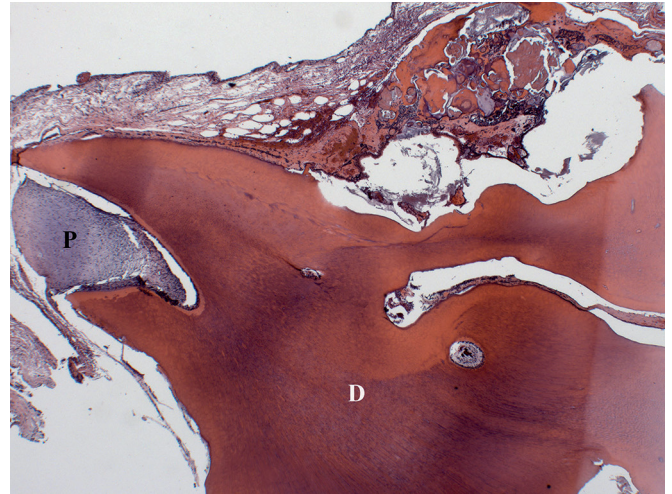


Figure 5. Microscopic examination presented a deposit of dental tissues (D) and dental pulp (P) (Hematoxylin-eosin, 40x).

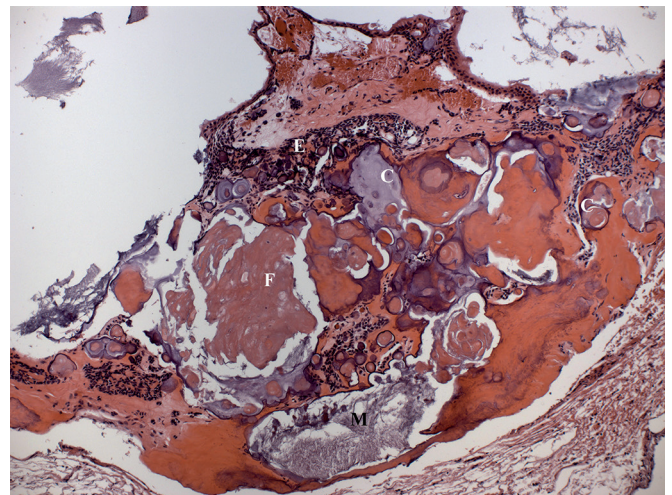


Figure 6. Odontogenic epithelium with ameloblastous aspect and presence of ghost cell masses. Foci of calcification along these cells were observed.

DISCUSSION

In the oral cavity, the odontogenic cysts present higher prevalence compared with odontogenic tumors⁹. The most common cysts are the radicular cyst, the dentigerous cyst, and the odontogenic keratocyst^{10,11}. A study with 351 cases of odontoma revealed that 27.6% of the sample was associated with dentigerous cysts, whereas only 0.9% was associated with CCOTs (10). In this context, the initial hypothesis should include this possibility of diagnosis despite its lower frequency.

Buchner¹² evaluated 215 intraosseous CCOT cases, and observed that 65% of the sample was located in the anterior region of incisors and canines, and that half of these cases were associated with odontoma, as

in the present case. In theory, this association commonly occurs due to the rich presence of odontogenic epithelium in CCOT cases, exhibiting the potential to develop other odontogenic lesions¹². The average age for CCOT occurrence, when related to odontoma, is 17 years old^{6,12}, which shows the accordance of this report with the literature.

In intraosseous cases, this cyst originates an expansion of the affected region, along with possible perforation of the vestibular bone, besides the possibility of root movement. In the present case, due to its yet small size, no expansion and/or bone perforation were observed.

Radiographically, the dentigerous cyst appears as an unilocular, radiolucid, well-delineated defect related to a tooth unerupted crown in its enamel cement junction, whereas the CCOT presents different radiographic aspects, from small unilocular, radiolucid areas to radiopaque structures on the lesion¹². In this case report, the radiographic characteristics were consistent with the possibilities presented by the studies on the matter.

Calcifying epithelial odontogenic tumor (CEOT), adenomatoid odontogenic tumor (AOC), and ameloblastic fibro-odontoma (AFO) should be considered for differential diagnoses, especially in more advanced lesions. These tumors are revealed as radiolucid lesions with some radiopaque areas on the interior. The CCOT occurs more frequently in the region of the incisors and canines, the CEOT occurs in third molar regions, and the AFO in premolar regions². The final diagnosis is always reached after histopathological examination.

The treatment of these types of lesions depends on their location and histopathological characteristics. They are normally conservative and consist of enucleation and curettage, as conducted in the reported case.

The prognosis is always favorable with no recurrence. However, if the CCOT is associated with another pathology, the treatment must consider the related lesion⁷, as proceeded in the present case, in which the enucleation of the odontoma and extraction of the deciduous tooth were executed. Currently, the patient is on his 12th postoperative month without clinical or radiographic signs of recurrence.

CONCLUSION

This case of CCOT presented imaging findings compatible with Dentigerous Cyst (light expansion

of the dental follicle). Considering this, professionals must always be aware of the tooth eruption chronology, investigating whether there is any delay in the permanent tooth eruption and/or any lesions associated with this delay. In this case report, the histopathological diagnosis was necessary to accomplish the correct follow-up of the patient, leading to the best prognosis. The accurate and early diagnosis of CCOT contributes to less invasive treatment and lowers the chance, although rare, of evolution to ameloblastoma.

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