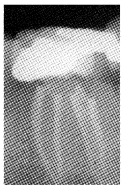
International Endodontic Journal

doi:10.1111/j.1365-2591.2008.01477.x



CASE REPORT

Persistent apical periodontitis associated with a calcifying odontogenic cyst

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Abstract

Estrela C, Decurcio DA, Silva JA, Mendonça EF, Estrela CRA. Persistent apical periodontitis associated with a calcifying odontogenic cyst. *International Endodontic Journal*, **42**, 539–545, 2009.

Aim To report a case of calcifying odontogenic cyst (COC) that was suggestive of apical periodontitis adjacent to the roots of the maxillary incisor teeth.

Summary Tooth 21 presented with clinical and radiographic signs of secondary infection, a post within the root canal and substantial internal tooth destruction; it was scheduled for endodontic surgery. Teeth 12 and 22 were root filled following the placement of a calcium hydroxide intracanal dressing for 21 days. Three attempts at root canal disinfection in tooth 11 were unsuccessful, and a persistent purulent drainage precluded completion of root canal treatment. Surgical enucleation of the periapical lesion was undertaken and the tissues submitted for histopathological examination. A diagnosis of COC was established based on the microscopic analysis. COC is an unusual benign lesion that represents 2% of all odontogenic lesions. Depending on the stage of development, it can mimic a large lesion associated with apical periodontitis and should therefore be considered in the differential diagnosis. In the case of COC, the definitive diagnosis can only be made with histopathological analysis.

Key learning points

- Persistent apical periodontitis may be of nonendodontic origin.
- Histological examination is essential to establish the cause of persistent apical periodontitis.
- Calcifying odontogenic cyst can mimic apical periodontitis.

Keywords: apical periodontitis, calcifying odontogenic cyst, differential diagnosis, Gorlin's cyst, persistent periapical lesion.

Received 14 April 2008; accepted 12 August 2008

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Introduction

Apical periodontitis is a sequel to root canal infection that can result in progressive stages of inflammation and destruction of periradicular bone. Root canal bacteria are the most common aetiologic factor responsible for apical periodontitis, which generally manifests as dental granulomas, radicular cysts or periapical abscesses (Bhaskar 1966, Nair 2004). Caution during endodontic diagnosis is mandatory to decide on the best therapeutic option, because periapical diseases of nonendodontic origin can mimic the more common infections of endodontic origin (Selden *et al.* 1998, Lee *et al.* 2007, Slutzky-Goldberg & Heling 2007, Faitaroni *et al.* 2008, Figueiredo *et al.* 2008).

The calcifying odontogenic cyst (COC) is an unusual odontogenic lesion described for the first time by Gorlin *et al.* (1962). It represents approximately 1–2% of all odontogenic tumors (Regezi *et al.* 1978) and occurs in the maxilla and mandible with similar frequency (Gorlin *et al.* 1962, Regezi *et al.* 1978, Waldron 2002). The World Health Organization (2005) classifies the COC as a benign neoplastic cyst with epithelium similar to that of an ameloblastoma and ghost cells that may produce calcifications (Waldron 2002, Carvalhosa *et al.* 2004, Praetorius & Ledesma-Montes 2005, Reyes *et al.* 2007). This cyst may be associated with other recognized odontogenic tumours, namely odontomas, adenomatoid odontogenic tumors and ameloblastomas (Waldron 2002).

The purpose of this paper is to report a case of COC that was suggestive of apical periodontitis. The treatment approach and the periapical outcome after root canal treatment and surgical enucleation of the lesion are described.

Report

A 26-year-old Caucasian male patient was referred to the Endodontic Clinic of the Federal University of Goiás, Brazil, for treatment of the maxillary right central incisor (tooth 11). A review of the patient's dental history revealed a coronal fracture in teeth #11 and 21 due to a motorcycle accident 3 years previously. At that time, the patient was referred to a general dentist for treatment and the postoperative course was uneventful. There was no history of pain in the traumatized teeth, but the patient complained of a low-intensity discomfort to pressure and palpation in the periapical region of tooth 11, with purulent drainage in the buccal region.

Clinical examination revealed the presence of a buccal fistula corresponding to the root of tooth 11 and an open cavity maintaining the root canal exposed to oral environment.

Radiographic analysis revealed a periapical radiolucent area involving teeth 12, 11, 21 and 22. The diameter of the radiolucency was approximately 15 mm in the periapical region of teeth 12 and 11, and 8 mm in the periapical region of teeth 21 and 22. The root canal of tooth 21 was poorly filled and there was a radiographic image suggestive of a fragment of an intracanal post. A pulp sensibility test with tetrafluoroethane spray (cold stimulus) was negative for teeth 12 and 22.

Based on these clinical and radiographic findings, the diagnosis was asymptomatic apical periodontitis. The treatment plan consisted of root canal treatment for teeth 12, 11 and 22, and endodontic surgery (rooted resection and root end filling with MTA; ProRoot Dentsply-Maillefer, Ballaigues, Switzerland) for tooth 21, which had a worse prognosis due to the destruction of the internal root canal walls (Fig. 1a,b).

In teeth 12, 11 and 22, root canal access was completed without anaesthesia. The canals were instrumented using a crown-down technique and the cervical third of the root canals was enlarged with numbers 3 and 4 Gates-Glidden drills (Dentsply Maillefer). Teeth 12 and 22 and tooth 11 were prepared up to a size 40 and a 50, respectively, 1 mm short of the apical foramen. In all teeth, the root canals were irrigated with 3 mL of 2.5%

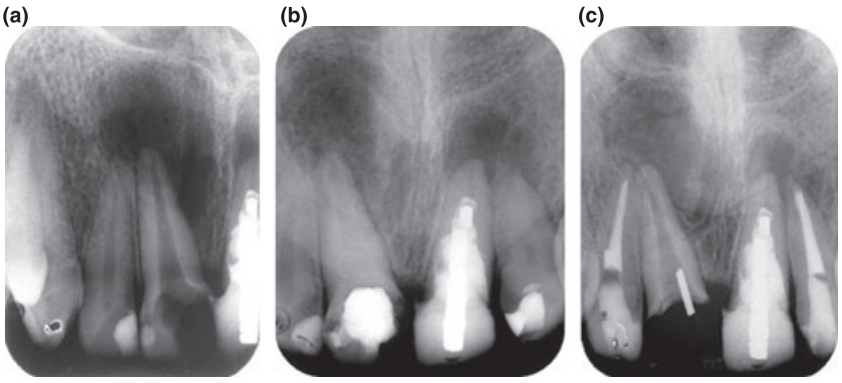

Figure 1 (a,b) Periapical radiographs of teeth 12, 11, 21 and 22 show radiolucent area characteristic of asymptomatic apical periodontitis. (c) Teeth 12 and 22 filled with gutta percha and Sealapex.

sodium hypochlorite (NaOCl) at each change of file and a calcium hydroxide paste with distilled water was used as an intracanal dressing. The quality of canal filling with the medication was confirmed radiographically. The teeth were temporarily restored with intermediate restorative material (IRM, Dentsply, Petropólis, RJ, Brazil).

After 21 days, the intracanal dressing was removed and the teeth canals of 12 and 22 were filled with gutta-percha and Sealapex (Sybron Kerr, Orange, CA, USA) using the lateral condensation technique (Fig. 1c). In tooth 11, however, a persistent purulent drainage was observed after the removal of paste, although the fistula on the buccal region had disappeared. The root canal was irrigated copiously with 2.5% NaOCl and sizes 15–20 K-Files (Dentsply Maillefer) were advanced approximately 0.5 mm beyond the apical foramen for debridement of this area. An aspiration cannula was placed for 3 min inside the root canal followed by a final flush with 2.5% NaOCl. The canal was filled again with the calcium hydroxide paste and the patient was scheduled to return 30 days later. Two weeks before the appointment, however, the fistula reappeared though with little purulent drainage. The root canal was reopened, the intracanal dressing removed and exudate observed. The calcium hydroxide paste was renewed once again and maintained for another 60 days. Within this period, the fistula disappeared and reappeared two times (Fig. 2a). On the 60-day appointment, exudate was still observed in the root canal.

At this stage the periapical lesion appeared to have increased only slightly on the basis of the preoperative radiographic analysis and compared with the radiographs taken during endodontic treatment. As the periapical infection persisted in spite of root canal treatment, the treatment approach was to fill the root canal of tooth 11, with subsequent surgery to remove the lesion adjacent to teeth 11, 21 and 22 (Fig. 2b) and submission for histopathological evaluation. As endodontic surgery had already been planned for tooth 21

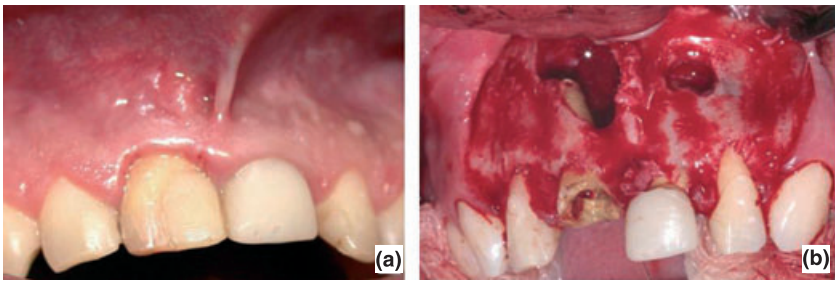

Figure 2 (a) Presence of fistula in tooth 11. (b) Clinical aspects after enucleation of periapical lesions.

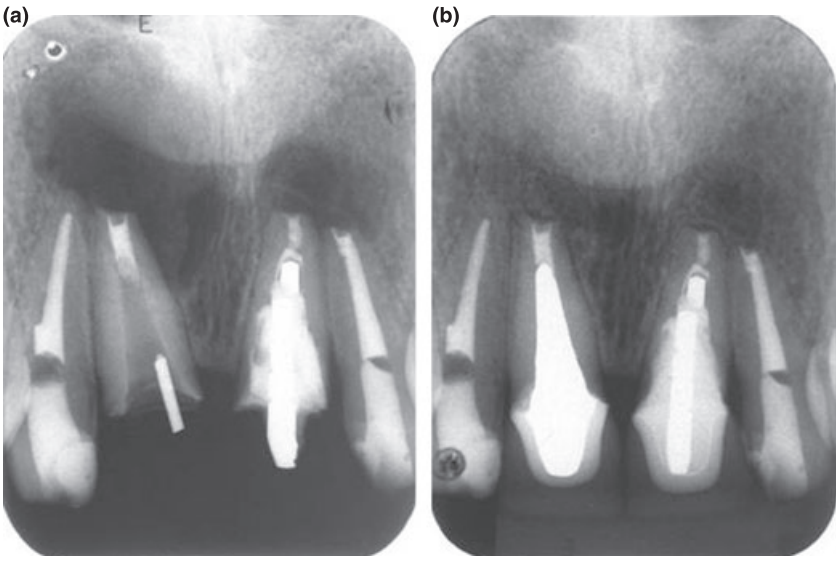

Figure 3 (a) Apicoectomy with MTA retrofilling on teeth 11, 21 and 22. (b) Follow-up of 1 year with final restoration and periapical repair.

(rooted resection and root-end filling), the surgical procedures were performed in a single intervention (Fig. 3a).

The histological examination confirmed the diagnosis of COC (Fig. 4a–d). Two weeks postoperatively, there was complete healing of the mucosa and absence of fistula. Teeth 12, 11 and 21 were restored. At the 1-year visit, periapical radiographic examination

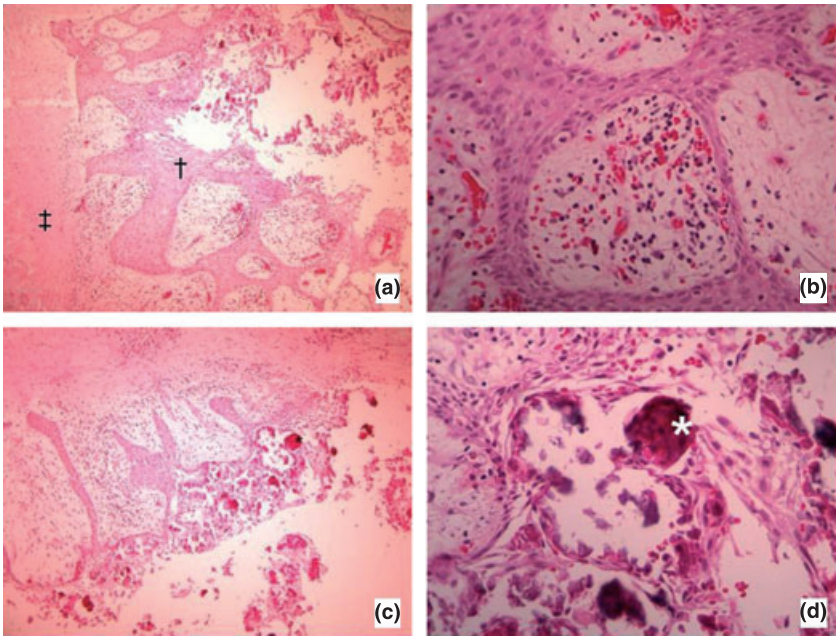

Figure 4 (a,d) Histology of odontogenic calcificant cystic tumor. Fibrous connective tissue with epithelial coating and hyperplasic characteristics. Presence of keratinization of coating epithelial cells characterizing ghost cells and presence of peripheral calcifications. Figures (b) and (d) are magnified views from Figures (a) and (c), respectively. (*calcifications, †epithelial coating, ‡connective membrane).

revealed bone tissue formation without clinical symptoms of pain or signs of lesion recurrence (Fig. 3b). The patient was informed of the need of continuing recall appointments during subsequent years.

Discussion

Calcifying odontogenic cyst (Gorlin's cyst) was recognized by the WHO as a distinct entity, and classified as a non-neoplastic cystic lesion by the WHO's international reference centre for the histological definition and classification of odontogenic tumors, jaw cysts and similar lesions (Carvalhosa *et al.* 2004, Praetorius & Ledesma-Montes 2005). Other definitions of COC are keratinizing and/or calcifying epithelial odontogenic cyst, Gorlin's cyst, cystic keratinizing tumor, calcifying ghost cell odontogenic tumor and cystic calcifying odontogenic tumor (Langlais *et al.* 1995).

Calcifying odontogenic cyst is predominantly an intraosseous lesion, but 13–21% of cases have an extraosseous location. It is asymptomatic and has similar appearance in the maxilla and mandible. Sixty-five percent of the lesions are found in the incisor-to-canine region (Waldron 2002). There is no preference for gender or race, emerging in most cases in the 2nd to 4th decade of life (Waldron 2002, Carvalhosa *et al.* 2004). Radiographically, COC usually has an unilocular aspect (Sapp *et al.* 2004, Praetorius & Ledesma-Montes 2005), but it can also appear as a multilocular lesion (Gorlin *et al.* 1962, Rushton & Horner 1997, Erasmus *et al.* 1998, Waldron 2002) with well circumscribed margins and containing diffuse radiopaque areas (Erasmus *et al.* 1998, Waldron 2002, Sapp *et al.* 2004, Praetorius & Ledesma-Montes 2005).

In the present case, the first diagnosis was an asymptomatic apical periodontitis. The routine protocol for primary endodontic infection with an open access cavity was initially undertaken, i.e. copious 2.5% NaOCl irrigation at each session and calcium hydroxide inter-appointment dressing for a prolonged period. These are the antimicrobial agents most frequently used in infected root canals (Estrela *et al.* 1999, 2007). However, persistence of the endodontic infection with exudative drainage in spite of root canal treatment required further consideration of the differential diagnosis. The histopathologic examination indicated a true diagnosis of COC. The characteristics of the case were consistent with those found in the literature (Gorlin *et al.* 1962, Regezi *et al.* 1978, Langlais *et al.* 1995, Rushton & Horner 1997, Erasmus *et al.* 1998, Regezi & Sciubba 1999, Waldron 2002, Carvalhosa *et al.* 2004, Sapp *et al.* 2004, Praetorius & Ledesma-Montes 2005, Reyes *et al.* 2007, Shear & Speight 2007), that is an asymptomatic maxillary intra-osseous lesion with unilocular radiographic appearance and discreet radiopaque areas occurring in a young adult. According to Waldron (2002), this non-neoplastic cyst presents a well-defined cystic cavity with a fibrous capsule and an odontogenic epithelium lining. The basal cells of the epithelial lining may be cuboidal or columnar and are similar to ameloblasts. The most typical histopathological feature of COC is the presence of variable numbers of ghost cells within the epithelium. Calcification within the ghost cells is common.

Radiolucent areas observed in the mandible or maxilla surrounding the apices of a tooth or various teeth may lead to an erroneous diagnosis of apical periodontitis because of pulpal involvement. Differential diagnosis should include other possibilities (Selden *et al.* 1998, Lee *et al.* 2007, Slutzky-Goldberg & Heling 2007, Faitaroni *et al.* 2008, Figueiredo *et al.* 2008). The definitive diagnosis of COC is only established based on histopathology findings (Regezi *et al.* 1978, Waldron 2002, Carvalhosa *et al.* 2004). A conservative treatment approach is indicated by lesion enucleation (Langlais *et al.* 1995, Waldron 2002, Carvalhosa *et al.* 2004, Sapp *et al.* 2004, Praetorius & Ledesma-Montes 2005, Reyes *et al.* 2007) with a reportedly low recurrence rate (Gorlin *et al.* 1962, Regezi *et al.* 1978,

Waldron 2002, Carvalhosa *et al.* 2004, Praetorius & Ledesma-Montes 2005, Reyes *et al.* 2007).

Conclusions

Apical periodontitis is generally a sequel to endodontic infection caused by micro-organisms from pulp necrosis. However, after several unsuccessful attempts to treat conventionally a root canal with an associated periapical lesion, other conditions must be suspected. A conservative treatment option can be surgical enucleation of the lesion and histopathological analysis. Nonendodontic diseases that mimic large apical periodontitis must be considered in the differential diagnosis. In the case of COC, the definitive diagnosis can only be made with histopathological analysis.

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