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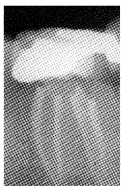

CASE REPORT

# A diagnostic dilemma: endodontic lesion or odontogenic keratocyst? A case presentation

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## Abstract

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**Aim** To present a clinical case of odontogenic keratocyst (OKC) simulating a lateral periodontal cyst.

**Summary** A 39-year-old female complaining of swelling and pain in the left mandibular premolar area was found to have a radiolucent lesion between teeth 34 and 35 (FDI). Both teeth had incomplete root fillings, and orthograde re-treatment of both premolars was performed. At 2-year follow-up, radiographic examination showed an increase in the radiolucent defect with respect to the previous examination. A surgical treatment of tooth 34 was then performed, with histological examination of the lesion. Histological features were consistent with an OKC, and the lesion was successfully treated by complete enucleation and application of Carnoy's solution. At 2-year follow-up, no clinical signs or symptoms were found and the radiolucent area had disappeared.

**Key learning points**

- Odontogenic keratocysts may mimic endodontic lesions.
- Clinicians should carefully review their cases and consider surgical intervention with biopsy in cases that do not heal.

**Keywords:** odontogenic keratocyst, periradicular lesion, radicular cyst.

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## Introduction

Differential diagnosis in endondotics may raise concerns when destructive lesions resorb significant amounts of alveolar bone. Generally, a sequence of clinical examinations is recommended for evaluating pulp health and a possible endodontic aetiology of the bony lesion (Pitt Ford & Patel 2004). The lack of a response to tooth sensibility test and the presence of a periradicular lucency generally indicates the need

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for root canal treatment. The treatment plan may be complicated when a radiolucent lesion surrounds a previously root-filled tooth. An orthograde re-treatment is usually recommended when an incomplete root filling is detected (Friedman & Stabholz 1986). When an orthograde treatment does not result in clinical and radiographic healing, a surgical approach may be indicated.

Well-defined radiolucent lesions surrounding teeth may present difficulties in differential diagnosis and in determining treatment strategies. Unilocular lesions such as periapical cysts may be located at the apical third of the root; lateral periodontal and lateral radicular cysts may present between the roots of teeth (Cairo *et al.* 2002); in the maxillary midline nasopalatine duct cysts may present (Magnusson 1978). A unilocular radiolucent lesion may also be a giant cell granuloma (Lombardi *et al.* 2006), a cystic ameloblastoma (Omura *et al.* 1997) or a schwannoma (Martins *et al.* 2007). The aim of this report is to present a clinical case of a nonhealing radiolucent lesion that created a diagnostic dilemma.

## Case report

In March 1999, a 39-year-old female was referred to the Department of Endodontics at the School of Dentistry, University of Florence, Italy. She reported an acute episode of swelling and pain a few days earlier, after which a buccal fistula with purulent discharge appeared in the mandibular left premolar region. No systemic disease was reported. On clinical examination, the patient reported pain during bi-manual palpation between teeth 34 and 35, close to the middle third of the roots. Periodontal examination revealed no loss of attachment at either site.

Radiographic examination revealed a well-circumscribed, unilocular radiolucent area between teeth 34 and 35 (Fig. 1). The radiographic defect did not involve the apex and the lesion was approximately 1 cm in diameter. Both teeth had incomplete root fillings and a coronal composite bonded restoration. The initial diagnosis was a lateral periodontal lesion resulting from endodontic infection. No alternative diagnoses were considered at this stage. The treatment plan consisted of an orthograde re-treatment of both premolars. After removing the coronal restorations, the gutta-percha was removed using a nickel titanium rotary instruments (ProFile orifice shaper; Dentsply Maillefer, Ballaigues, Switzerland) in the coronal and middle thirds of the roots. Hand instrumentation and solvent were used to remove the apical filling material and copious irrigation was performed with 5% NaOCl (Niclor 5; OGNA, Milan, Italy). A blocked canal was found in the apical third of tooth 34, making complete shaping impossible. The canal was refilled to the

**Figure 1** Intraoral radiograph revealing a well-circumscribed radiolucent region in the alveolar bone and the incomplete root canal treatment of teeth 34 and 35.

point of obstruction and the teeth were sealed coronally with enamel and dentine bonded composite resin. A radiograph was taken to check the quality of root canal treatment. Radiographic examination at the 1-year follow-up showed a reduction of more than 50% in the radiolucent area (Fig. 2), both teeth responded normally to percussion and palpation. No clinical symptoms were reported.

At the 2-year follow-up, the teeth were functionally stable, with no loss of periodontal attachment but the patient complained of occasional pain on chewing. The radiographic examination showed an increase in the radiolucent defect with respect to the previous examination; the size was similar to the findings at the first visit (Fig. 3). Because of the blocked canals encountered during an orthograde re-treatment and an uncertain origin of the radiolucent defect, a surgical treatment of tooth 34 and a biopsy of the radiolucent area was scheduled. A mucoperiosteal full-thickness flap with intrasulcular incision and vertical releasing incisions was raised with papillary preservation. A microsurgical blade (Stainless Fine surgical blades; Swann Morton, Sheffield, UK) was used. The lesion was enucleated and the periradicular tissue was removed for biopsy. Following flap reflection, resection of the apical 3 mm of first premolar root was performed approximately perpendicularly to the long axis of the root. The retro-cavity was prepared using ultrasonic retro-tips (KIS 3D; Spartan Obtura, Fenton, MI, USA). Root-end cavities were dried and MTA cement (ProRoot™ MTA; Dentsply Endodontics, Tulsa, OK, USA) was used to seal the canal. The radiographic examination immediately following root-end filling is shown in Fig. 4.

**Figure 2** One-year follow-up: intraoral radiograph after orthograde endodontic re-treatment of teeth 34 and 35. The radiolucent lesion appeared substantially decreased.

**Figure 3** Recurrence of periradicular radiolucent lesion between teeth 34 and 35 2 years after root canal re-treatment.

 

**Figure 4** Periapical radiograph after endodontic surgery.

Histological examination revealed the presence of epithelial lining with 5–6 layers of columnar basal cells. Epithelial islands were present in the lumen of the cystic cavity, demonstrating a corrugated surface and the presence of residual para-keratinized epithelium. The histological diagnosis was odontogenic keratocyst (OKC) (Fig. 5). These results were presented to the patient and a second surgical procedure to treat the OKC was recommended. As the symptoms had lessened following the first surgical procedure, the patient preferred to wait before the next surgery, which was scheduled 6 months later.

Second surgical procedure: a muco-periosteal trapezoidal flap was raised with an intrasulcular incision in the premolar area and two vertical incisions. The interproximal papilla between the premolars was completely preserved. The vestibular bone plate was found to be completely resorbed. Curettage of the adjacent healthy bone was performed with a round bur (Fig. 6) Carnoy's solution (glacial acetic acid, ferric chloride, chloroform and absolute alcohol) was then applied into the cyst lumen for 3 min (Blanas *et al.* 2000).

The flap was then repositioned at the pre-surgical level by means of 5-0 silk interrupted sutures (Fig. 7). All the procedures were performed under microscopic vision (10×). The patient was instructed to avoid tooth-brushing or any trauma in the area, and to rinse with

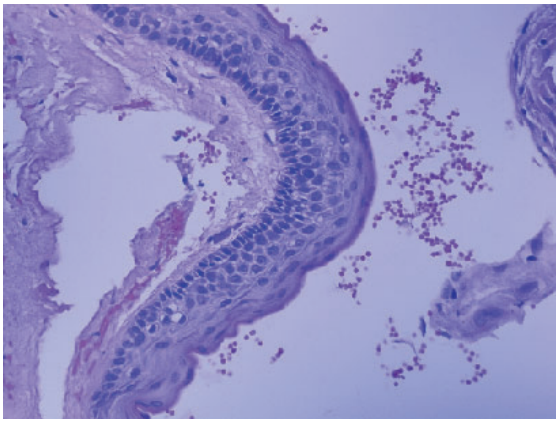

**Figure 5** Histologic appearance of odontogenic keratocyst. The figure shows a characteristic stratified squamous epithelium 5 or 6 cells thick with a corrugated para-keratinized surface. Epithelial islands were present in the lumen of the cystic cavity (Haematoxylin eosin stain; original magnification at 40×).

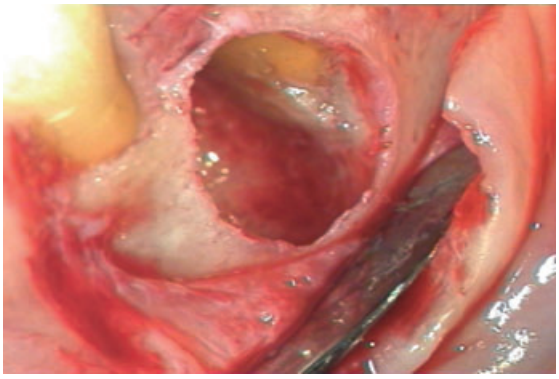

**Figure 6** Clinical view of the vestibular bone plate after total enucleation of the cyst.

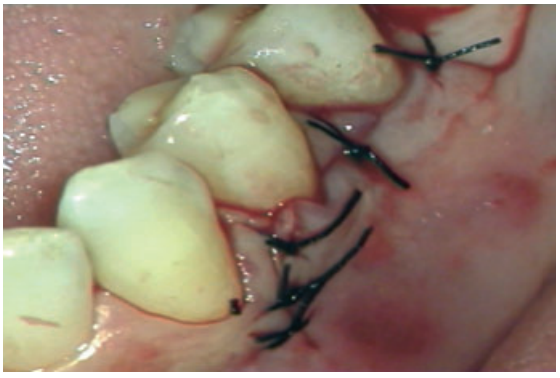

**Figure 7** Clinical situation at the conclusion of the surgery.

chlorhexidine 0.12% (Curaprox; Curadent International, Kriens, Switzerland) three times a day. Analgesics were prescribed for use as needed. The sutures were removed 7 days after surgery. Healing was uneventful. At the 2-year follow-up, no clinical signs or symptoms were reported; the teeth were functionally stable and the radiolucent area had disappeared (Fig. 8).

**Figure 8** Intraoral radiograph 2 years after the removal of OKS. Although the bony healing of the defect is not complete, reduction of the radiolucent region, with no periapical lesion on the first and second mandibular premolar being evident.

 

## Discussion

The OKC was first described by Phillipsen (1956). This lesion is classified by the World Health Organization as a developmental, noninflammatory odontogenic cyst (Kramer *et al.* 1992) arising from rests of dental lamina cells (Tsukamoto *et al.* 2001). Histologically, it is characterized by a uniform epithelial layer with a corrugated para-keratinized luminal layer and a prominent basal-cell layer (Ali & Baughman 2003).

The OKC is associated with a high recurrence rate (Payne 1972, Shear 1994) and the most common location is the third molar and ramus area of the mandible. OKC are characterized by an aggressive behaviour and can demonstrate a greater expansion or recurrence when compared with other jaw cysts (Nakamura *et al.* 2002). In this clinical report, a second surgical procedure was indicated because of the nature of the bony lesion, identified following histological examination of periradicular tissue. Therefore, a surgical procedure for complete cystic enucleation was then performed, and Carnoy's solution was applied in order to reduce the risk of recurrence (Blanas *et al.* 2000).

Upon radiographic examination, these lesions appear as unilocular or multilocular radiolucent areas with clear contours, although clinical and radiographic findings are not considered unequivocal proof for a definitive diagnosis (Brannon 1976). The ultimate diagnosis is histological, revealing the presence of para-keratinized epithelial tissue surrounding the cystic cavity (Brannon & Colonel 1976, Ali & Baughman 2003); as proved by the histological analysis performed after the first surgical procedure.

In a systematic review, Blanas *et al.* (2000) analysed treatment methods and the associated prognoses of OKC. The authors reported that the literature concerning OKC was limited to retrospective consecutive case series. Surgical approaches based on complete resection showed the lowest recurrence rate (0%) but the highest morbidity rate (including discomfort, pain and paraesthesia). Simple enucleation was reported to have a recurrence rate of 17–56%. Simple enucleation combined with adjunctive therapy, such as the application of Carnoy's solution or decompression before enucleation, was reported to have recurrence rates of 1–8.7%. In this report, the presence of incomplete root filling and the episode of swelling reported by the patient, led the practitioner to diagnose a lesion with an endodontic origin, even though its location was unusual (lateral and not periapical).

An orthograde root canal re-treatment was scheduled because of the inadequate primary treatment. It seemed reasonable to assume that the bacteria present in the root canal system caused the subsequent infection in the lesion, and the swelling reported by the patient. The partial resolution of the radiolucent lesion at 1-year follow-up, after orthograde re-treatment, may be attributed both to the elimination of the endodontic supra-infection, and to drainage via the vestibular fistula. After 2 years, the recurrence of the periradicular radiolucent lesion supported a surgical re-treatment of the first premolar and a biopsy of the lesion. The histological evaluation revealed the bony lesion to be OKC. Therefore, a second surgical procedure was indicated. The treatment of the OKC was a cystic enuclation and application of Carnoy's solution, based on its efficacy (application of Carnoy's solution improves results of complete enucleation only) and minimal postoperative discomfort if compared with marsupialization or extensive bony resection (Blanas *et al.* 2000). At the 3-year follow-up, healing was uneventful. Given the known high-risk of recurrence of OKS, the patient continues to be recalled for check-ups every 6 months (Maurette *et al.* 2006).

## Conclusion

The radiographic and clinical characteristics of OKCs are not pathognomonic signs and may lead to a difficult diagnosis especially when this lesion is adjacent to teeth with nonvital pulps or inadequate root fillings. When an orthograde re-treatment is not feasible

or is ineffective, a retrograde re-treatment with a biopsy is recommended because an OKC in an unusual location may simulate a periapical lesion.

## Disclaimer

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