

Case Report

A diagnostic dilemma-endodontic lesion or keratocystic odontogenic tumor (KCOT): A case report

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We present a case of keratocystic odontogenic tumor (KCOT) in maxillary canine region which was mistaken for a periapical lesion and was treated accordingly that is, root canal treatment of the teeth involved, was attempted, without checking for their vitality status. When the symptoms didn't subside, the patient was operated under local anesthesia and the lesion was sent for histopathological examination which confirmed the diagnosis of an infected KCOT. Keratocystic Odontogenic tumor may look similar to endodontic lesions. Clinicians should carefully review the cases and consider surgical intervention with biopsy in cases that do not heal.

Key words: Keratocystic odontogenic tumor, Peri-apical lesion, odontogenic keratocyst, lateral periodontal cyst.

INTRODUCTION

There may be many aggressive, inflammatory, benign and malignant diseases that appear as periapical radiolucencies, resorbing significant amount of alveolar bone and sometimes it may become very difficult to differentially diagnose between these lesions as clinically and radiographically, they may be very similar (Figure 1), making it difficult to determine treatment strategies. Thus, a proper history and a thorough clinical and radiographic examination is recommended for evaluating pulp health and a possible etiology of bony lesion (Pitt and Patel, 2004). Generally, lack of response to tooth vitality test and the presence of periradicular radiolucency indicates the need for root canal treatment. The aim of this report is to present a clinical case of keratocystic odontogenic tumor (KCOT) which presented as a case of periapical radiolucency thus creating a diagnostic dilemma.

Case report

A 31 year old male patient (Figure 2) reported to the

department of periodontics at Rajasthan Dental College and Hospital, Jaipur, with a chief complaint of dull throbbing, continuous type of localized pain and swelling with a draining sinus in maxillary left canine region for last 4-months, which he noted to be gradually enlarging. There was no extra-oral swelling and medical history was noncontributory.

Intra-oral examination revealed:

- 1) A soft, fluctuant, non-tender swelling of approximately 1 cm diameter, involving the facial attached and marginal gingiva, with a draining sinus in maxillary left canine region, that is the 22 and 23 region,
- 2) No obliteration of the vestibule,
- 3) Palpation showed no bony expansion or egg-shell crackling and no other abnormality was detected,
- 4) No change in the teeth position and the teeth involved (22th and 23rd region) were stable with probing depth within normal limits (< 3 mm) (Figure 3).

History revealed a visit to a private clinician 4 months back due to pain in the same region where root canal treatment of 22 was initiated following a radiograph (Figure 4) which showed a periapical radiolucency involving the roots of 22 and 23. Since there was no relief from pain, the root canal of 23 was opened. After 2

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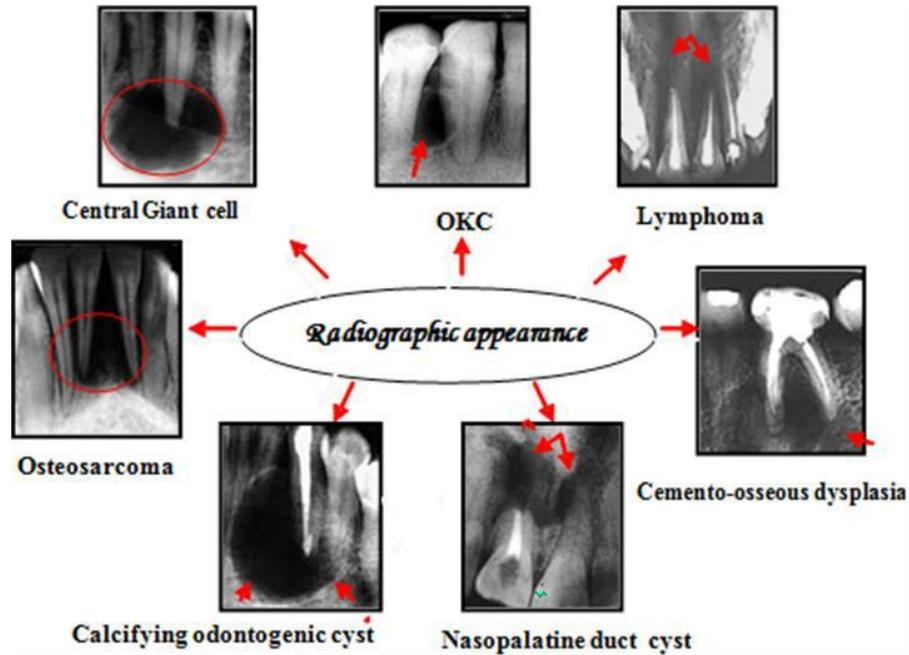


Figure 1. Radiographic appearance of various diseases that may appear as periapical radiolucencies.



Figure 2. Extra-oral appearance of the patient showing complete facial symmetry and no evidence of facial swelling that is, no extraoral abnormality.

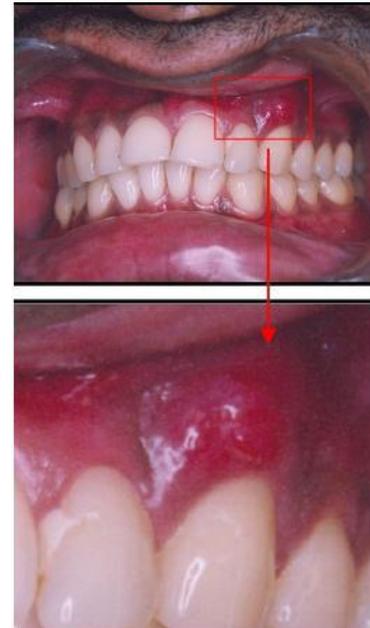


Figure 3. Soft fluctuant swelling seen intra-orally involving the gingiva in 22 and 23 region.

months, patient was referred to the Department of Periodontics, Rajasthan Dental College and Hospital with no improvement in his condition. At the hospital, an Intra-oral periapical radiograph and a true occlusal radiograph (Figure 5) were taken and revealed;

1). A clear, single, unilocular, radiolucency, measuring

approximately 1 cm mesio-distally, extending from distal surface of the root of 22 up to the mesial surface of the root of 23 and approximately 1.5 cm coronal-apically extending from alveolar crest to the apical aspect of 22 and 23.



Figure 4. Radiographic appearance of the lesion at the initiation of the root canal treatment showing a unilocular radiolucency (1 × 1.5 cms mesio-distally and corono-apically), and borders are not well circumscribed.



Figure 5. (a and b) Intraoral periapical and True occlusal radiograph taken at the hospital showing radiolucency in between the roots of 22 and 23, causing flaring of the roots.

- 2). Corticated borders were not clearly appreciated with hazy margins on the superior aspect of the lesion, 3). Roots were flared with the absence of lamina dura of both the roots,
- 4). No root resorption and no tooth displacement,
- 5). Radio-opacities seen in the coronal part of both the teeth (coronal restorations).

Differential diagnosis given initially was:

- 1) Lateral periodontal cyst
- 2) Globulomaxillary cyst
- 3) Adenomatoid odontogenic tumor
- 4) Primordial cyst
- 5) Keratocystic Odontogenic tumor (KCOT).

The treatment plan consisted of completion of root canal treatment for 22 and 23 since the root canal was already opened (Figure 6). After removing the coronal restorations, hand and rotary instrumentation were performed to shape the canals. Copious irrigation was done using 5% NaOCl (Sodium Hypochloride) and the canal was obturated to the apex and the teeth were sealed coronally with enamel and dentine bonded composite resin.

Radiographic examination at 6 months follow up showed no improvement in the radiographic defect and the patient's chief complaint persisted, that is, dull, throbbing pain, along with buccal fistula. Because of this, a surgical enucleation (Figure 7) and biopsy of the lesion was scheduled. A mucoperiosteal full-thickness flap with intrasulcular incision and vertical releasing incisions was



Figure 6. Radiograph taken after the completion of the root canal treatment of both the teeth.

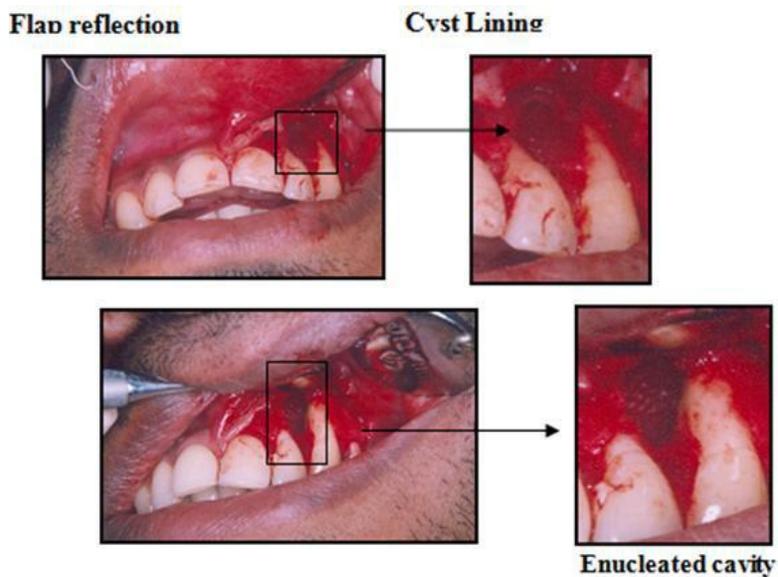


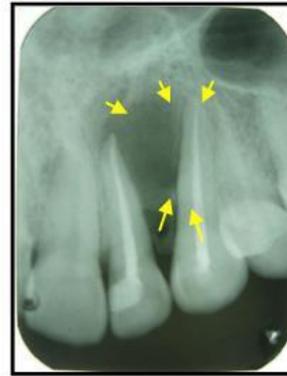
Figure 7. Surgical enucleation of the lesion.

although the lining was removed in pieces due to its fragile nature. After enucleation the bony cavity was seen extending from distal surface of 22 till the mesial surface of root of 23 (both the root surfaces were visible in the bony cavity). While enucleating, a thick white cheesy material was aspirated. Thus, the provisional diagnosis of KCOT was thought to be most appropriate. The enucleated tissue was sent for histologic examination, following which, the flap was sutured back. A follow-up after 1-week was performed which showed no sign of any swelling or suppuration and the patient was completely relieved of his chief complaint (Figure 8). Histopathologic picture showed:

- 1) Stratified squamous parakeratinized epithelium 6 to 7 cells thick with a corrugated layer of parakeratin and palisaded, polarised basal cells,
 - 2) Connective tissue consisted of Engorged blood vessels, moderate chronic inflammatory cell infiltrate and RBC extravasation. Thus, a diagnosis of INFECTED KCOT was confirmed (Figure 9).
- After this, a second surgical procedure to treat the lesion was recommended and was scheduled after 6 months. For the 2nd procedure, a mucoperiosteal trapezoidal flap was raised with an intra-sulcular incision in the incisor – premolar region with 2 vertical releasing incisions.



After 1 week...



After 3 months

Figure 8. Post-operative clinical picture after 1 week.

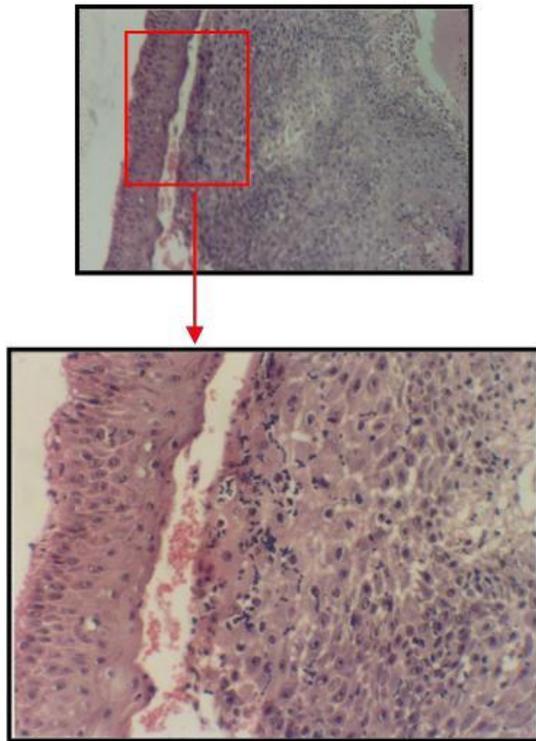


Figure 9. Histopathologic picture confirming the diagnosis of infected KCOT.

Curettage of the adjacent healthy bone was performed with a round bur. Carnoy's solution (glacial acetic acid, ferric chloride, chloroform and absolute alcohol) was then applied in the cystic lumen for 3 min (Blanas et al., 2000). The flap was then repositioned at the presurgical level by means of 5 to 0 silk interrupted sutures. The patient was given post-operative instructions; to avoid tooth-brushing or any trauma in that area, and to rinse

with chlorhexidine 0.2% twice in a day. Patient was called 7 days later for suture removal and healing was seen to be normal. At 1-year follow-up, no clinical signs or symptoms were reported; the teeth were functionally stable and the radiolucent area had reduced significantly (Figure 10).

DISCUSSION

As said earlier, many diseases may mimic periapical radiolucencies and may need to be differentially diagnosed. The diseases are listed as follows:

Inflammatory diseases

- a) Periapical Granuloma / Scar (Cairo et al., 2002),
- b) Periapical cyst,
- c) Periapical abscess.

Benign diseases

- 1) Traumatic bone cyst,
- 2) Nasopalatine duct cyst (Magnusson, 1978),
- 3) Adenomatoid odontogenic tumor,
- 4) Periapical cemento-osseous dysplasia,
- 5) Ossifying /cementifying fibroma.

Benign aggressive diseases

- a) Keratocystic odontogenic tumor,
- b) Central Giant Cell Granuloma (Lombardi et al., 2006),
- c) Calcifying epithelial odontogenic tumor (CEOT).

Malignant diseases

- 1) Lymphoma/Leukemia,



Figure 10. Radiographic appearance at 1 year follow up showing considerable healing of the radiouency.

2) Multiple myeloma.

KCOT was formerly called OKC (Odontogenic Keratocyst) and was first described by Philipsen (1956). It was classified by the World Health Organization as a developmental, noninflammatory Odontogenic cyst (Kramer et al., 1992) which arises from rests of dental amina cells (Tsukamoto et al., 2001). This lesion was recently renamed by him as keratocystic odontogenic tumour (KCOT) and reclassified as an odontogenic neoplasm in the world health organization's edition of its histo-logical classification of odontogenic tumours (Philipsen 2005). According to this edition the KCOT has been defined as "A benign uni or multicystic intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behavior and having a very high recurrence rate. KCOT are less common in

maxilla than mandible with only 31.3% in maxilla (Mervyn, 2007). But when they do occur, they are more common in the canine region and more commonly affect males as compared to females (Ratio 1.3:1), which was the case in our patient also. KCOT may occur as solitary cysts, multiple cysts or they may occur as components of a syndrome. These lesions may be unilocular or multilocular radiolucent areas with clear contours (Brannon, 1976). KCOT has been shown to have a bimodal age distribution with first peak in 2nd and 3rd decade and the second peak in the 5th decade or older. It is said that the lesions in the second peak are more common in maxilla which corresponded with our case (Mervyn, 2007).

An important feature that helps to distinguish the KCOT is stated by White and Pharoah (2009) stating that KCOTs exhibits "propensity to grow along the internal aspect of the jaws, causing minimal expansion". The extension is more in antero-posterior direction and the

pressure of the fluid is quite low and grows by extension of the cystic lining that is enlarges slowly but relentlessly along the path of least resistance (R A Cawson). So, not much of cortical expansion is seen in the initial stages, as

in this case. The ultimate diagnosis is histological, revealing the presence of para-keratinised epithelial tissue surrounding the cystic cavity (Brannon and Colonel, 1976; Ali and Baughman, 2003); as proved by the histological analysis performed after the first surgical procedure in this case. In a systemic review, Blanas et al. (2000) analysed treatment methods and the associated prognosis of KCOT. Treatment may be of 2 types:

- 1). Conservative,
 - a. Marsupialisation,
- 2). Aggressive,
 - a. Enucleation,
 - b. Use of Carnoy's solution,
 - c. Enucleation along with peripheral ostectomy.

Surgical approaches based on complete resection showed the lowest recurrence rate (0%) but the highest morbidity rate (including discomfort, pain and paresthesia). Simple enucleation was reported to have a recurrence rate of 17 to 56%. Simple enucleation combined with adjunctive therapy, such as the application of Carnoy's solution or marsupialisation before enucleation, was reported to have a recurrence rate of 1 to 8.7%. In this report, the episode of draining sinus reported by the patient led the practitioner to diagnose a lesion with an endodontic origin. Since the lesion failed to resolve even after completion of the endodontic treatment, a surgical procedure was indicated. A surgical re-treatment was advised because of the nature of the bony lesion, as revealed by the histologic examination.

The treatment of KCOT was a cystic enucleation and application of Carnoy's solution, based on its efficacy (application of this solution improves result of complete enucleation) and minimal post-operative discomfort, when compared with marsupialisation or extensive bony resection (Blanas et al., 2000). At 1 year follow-up, healing was uneventful and no signs of recurrence.

Conclusion

A simple case of purulent gingival swelling, along with the presence of a radiolucent lesion which was thought to be of endodontic origin and was treated likewise, was subsequently diagnosed to be a case of keratocystic odontogenic tumor.

Clinical significance

- 1) KCOTs can be mistaken for inflammatory lesions of perio-endo origin,

- 2) Vitality status of the associated teeth should be known,
- 3) Histopathological examination of the tissue is mandatory for a definitive diagnosis,

- 4) Due to high recurrence rate and aggressive behavior of this lesion, periodic follow up is very important (Ali and Baughman, 2003).

A good doctor is a person who observes things with a third eye and listens with a third ear."

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