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CASE REPORT

Keratocystic odontogenic tumour arising as a periapical lesion

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Abstract

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Aim To document a case of a keratocystic odontogenic tumour (KOT) involving the apical region in the maxilla mimicking a periapical lesion of endodontic origin.

Summary Benign and malignant tumours, including odontogenic lesions, can be erroneously diagnosed as periapical radiolucencies. KOTs mimicking periapical lesions of endodontic origin are uncommon, especially when the lesions involve the maxilla. This article describes a 55-year-old man with a well-delimited, oval-shaped, radiolucent lesion, occupying the middle and apical third of teeth 22 and 23. After 30 days, the clinical and radiographic findings remained unchanged and the patient was referred for surgical removal of the lesion. Clinical, radiographic and histopathological features are also discussed and compared with current literature.

Key learning points

- Keratocystic odontogenic tumours are benign, but locally aggressive, cystic neoplasms that may be misinterpreted as periapical lesions of endodontic origin and result in unnecessary and ineffective root canal treatment.
- Lesions of nonendodontic origin such as odontogenic tumours should be included in the differential diagnosis of periapical inflammatory lesions.
- All tissue removed from the apical region must be sent for histopathological examination to establish the final diagnosis and to prevent misdiagnosis.

Keywords: keratocystic odontogenic tumour, odontogenic keratocyst, periapical lesion.

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Introduction

Keratocystic odontogenic tumours (KOTs) are benign, but locally aggressive, cystic neoplasms that arise from the dental lamina or its remnants or from basal cell extensions of the lining oral epithelium (Stoelinga 2001, Shear 2003, Agaram *et al.* 2004, Saraçoğlu *et al.* 2005, Mello *et al.* 2011). These tumours have a high rate of recurrence and are often associated with nevoid basal cell carcinoma syndrome (Shear 2003, Agaram *et al.* 2004, Saraçoğlu *et al.* 2005). Radiographically, KOTs appear as uni- or multilocular images with sclerotic borders that may or may not be associated with impacted teeth (Shear 2003, Chirapathomsakul *et al.* 2006, Mello *et al.* 2011). Microscopically, the World Health Organization defines KOT as a neoplasm with a cystic wall lined by parakeratinized and corrugated stratified squamous epithelium that exhibits a palisaded basal layer with nuclei intensely basophilic (Philipsen 2005).

Keratocystic odontogenic tumours are more frequent in the mandible (Philipsen 2005, Tejasvi *et al.* 2010, Khanna *et al.* 2011, Mello *et al.* 2011), but the maxilla can also be affected, although at a lower proportion (Ali & Baughman 2003, Habibi *et al.* 2007, Melo *et al.* 2012). Nevertheless, KOTs that mimic periapical lesions of endodontic origin are uncommon, especially when the lesions involve the maxilla (Wright *et al.* 1983, Stajcić & Paljm 1987, Nohl & Gulabivala 1996, Garlock *et al.* 1998, Tan 2001, Pace *et al.* 2008, Tejasvi *et al.* 2010, Khanna *et al.* 2011).

This article reports a case of KOT that was clinically and radiographically diagnosed as a radicular cyst and draws the attention of dental practitioners and pathologists to the occurrence of this tumour in the periapical region, a fact that can lead to misinterpretation and misdiagnosis.

Case report

A 55-year-old man was seen by an endodontist for a painless maxillary swelling in the region of the left lateral incisor that extended to the first premolar on the same side. Intra-oral examination revealed a discrete swelling in the corresponding region and a single crown restoration on tooth 24. Panoramic radiography (Fig. 1a) showed bilateral thickening of the sinus mucosa, the absence of some teeth and the presence of endodontic material that partially filled the root canal of teeth 23 and 24. In addition, a well-delimited, radiolucent, oval-shape image that measured approximately 3 cm in its major diameter was observed, which occupied the middle and apical third of teeth 22 and 23. Taking into account the coronal restoration of tooth 23 whose root canal was only partially filled, the absence of pulp vitality of tooth 22 and the clinical suspicion of a radicular cyst, the endodontist suggested root canal retreatment of tooth 23 and endodontic treatment of tooth 22. After 30 days, the clinical and periapical radiographic findings (Fig. 1b) remained unchanged and the patient was referred for surgical enucleation followed by bone curettage of the lesion. The specimen was sent for histopathological analysis, and the diagnosis was keratocystic odontogenic tumour (KOT). The patient continues with clinical and radiographic follow-up and bone repair and no signs of recurrence were observed after eleven months (Fig. 1c,d).

Discussion

This study reports the case of an odontogenic lesion that involved the periapex of teeth 22 and 23 in a 55-year-old man. Although uncommon, the finding of tumours that arise as periapical radiolucencies is not surprising because benign (Morais *et al.* 2011, Rodrigues *et al.* 2011), malignant (Favia *et al.* 2000, Kuc *et al.* 2000, Chen *et al.* 2004, Choi

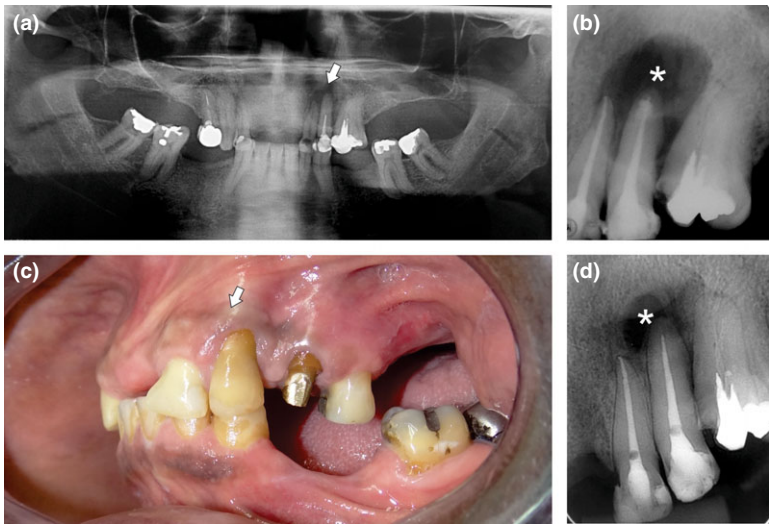

Figure 1 (a) Well-delimited, radiolucent, oval-shape image located on the middle and apical third of teeth 22 and 23; (b) unchanged lesion following endodontic treatment of tooth 22 and retreatment of tooth 23; (c) note absence of swelling; (d) and bone repair in the affected region after 30 days.

et al. 2012) and even metastatic tumours can be found in this region (Milobsky *et al.* 1975, Block *et al.* 1977, Nevins *et al.* 1988). However, although cases of odontogenic cysts and tumours that mimic periapical lesions have been previously reported (Kuc *et al.* 2000, Cunha *et al.* 2005, Nikitakis *et al.* 2010, Tejasvi *et al.* 2010, Gondak *et al.* 2013), few cases of keratocysts in the periapical region exist (Wright *et al.* 1983, Stajcić & Paljm 1987, Nohl & Gulabivala 1996, Garlock *et al.* 1998, Tan 2001, Pace *et al.* 2008, Tejasvi *et al.* 2010, Khanna *et al.* 2011).

A search of the English-language literature for cases of KOT involving the periapical region identified 27 case reports, including this case report (Table 1). The mean age of the patients was 50 years (range: 27–78 years). Men were more frequently affected than women were. There was a slight predominance of cases in the periapical region that affected the maxilla compared with the mandible and most of them involved the anterior region.

According to Brannon (1976), Chuong *et al.* (1982) and Stajcić & Paljm (1987), the frequency of keratinizing cysts in the periapical region ranges from 0.5 to 7%. In addition, other tumours that arise in the tooth periapex less frequently have been also described as lymphomas (Mendonça *et al.* 2013), and sarcomas (Davido *et al.* 2011), salivary gland carcinomas (Favia *et al.* 2000, Chen *et al.* 2004), haemangiomas (Orsini *et al.* 2000), schwannomas (Martins *et al.* 2007) and lipomas (Morais *et al.* 2011). With respect to odontogenic tumours, Wright *et al.* (1983) reported four cases of periapical keratocysts, with three of them involving the anterior maxilla. Stajcić & Paljm (1987) described four cases of odontogenic keratocysts that were diagnosed clinically as radicular cysts, with two of them involving the maxilla. Pace *et al.* (2008) reported a case of KOT affecting the mandible, which mimicked a lateral periodontal cyst. Garlock *et al.* (1998) described a series of 239 cases of KOT, in which 12 cases were associated with root filled teeth, thus mimicking lesions of endodontic origin. Half the cases involved the mandible and half the maxilla. Cunha *et al.* (2005) reported a case of unicystic ameloblastoma that involved the distal root of tooth 36, which had been previously diagnosed as a cystic lesion. Recently, Gondak *et al.* (2013) described five cases of unicystic ameloblastoma that affected the posterior mandible in young adults, which manifested as apical periodontitis.

Table 1 KOT cases in the periapical region published in the English-language literature.

Author (year)	Region	Age	Gender
Wright *et al.* (1983)	1.1, 1.2 and 1.3	69	M
	2.1	37	M
	2.1	50	M
	3.3 and 3.4	64	M
Nohl & Gulabivala (1996)	3.2 and 3.3/4.2 and 4.3	38	F
	3.1	33	M
Garlock *et al.* (1998)	2.5	76	M
	2.5	36	M
	1.4	70	F
	1.4	48	M
	1.2	68	M
	2.3	37	M
	3.3 and 3.4	78	M
	3.5	52	M
	3.2	52	M
	3.2	61	M
	4.1 and 4.2	34	M
	4.1 and 4.2	54	F
Stajcić & Paljm (1987)	Two cases in maxilla[a]	NI	NI
	Two cases in mandible[a]		
Tan (2001)	1.3	27	M
Pace *et al.* (2008)	3.4	39	F
Tejasvi *et al.* (2010)	4.6	41	M
Khanna *et al.* (2011)	2.3	31	M
Santos *et al.* (current case)	2.2 and 2.3	55	M

[a]Unknown tooth.
NI, Not informed; F, female; M, male; KOT, keratocystic odontogenic tumour.

Other lesions such as an orthokeratinized odontogenic cyst (Hancock *et al.* 1986), a lateral periodontal cyst (Nikitakis *et al.* 2010), a traumatic bone cyst and cysts of the nasopalatine duct (Suter *et al.* 2011) have also been reported as mimicking inflammatory periapical lesions. Nevertheless, inflammatory periapical lesions with extensive accumulation of Russel bodies may also mimic a malignant neoplasm (Dos Santos *et al.* 2008).

In the present case, the lesion's clinical appearance was a discrete swelling of the affected region. On radiographical examination, the lesion seemed to be a commonly occurring lesion such as a radicular cyst because the affected region contained teeth with crown restorations or fillings. However, marked radiolucency and an oval shape are uncommon findings for radicular cysts (Ørtavik *et al.* 1986, White *et al.* 1994).

On the basis of the patient's history and radiographic images, the initial diagnosis was a lesion of endodontic origin. Periapical lesions can be differentiated radiographically from KOTs by their destructive nature, with a more marked radiolucency observed in the cases of KOTs. Shared findings include well-demarcated margins and the location of the lesion in the periapical region. Regarding the latter feature, according to the systematic reviews of MacDonald-Jankowski (2011) and Sansare *et al.* (2013), KOTs in the maxilla are rare.

The present case fulfils the histopathological criteria for KOT (Philipsen 2005). The lesion was a cystic tumour consisting of a thin fibrous wall lined by corrugated and atrophic parakeratinized stratified pavement epithelium that exhibited a palisaded basal layer of cells (Fig. 2a). An interesting microscopic observation was the marked presence of haemorrhagic content and keratin laminae between cystic fibrous walls without lining epithelium (Fig. 2b,c). These keratin laminae were the first clue to the lesion's noninflammatory nature of the lesion. At a lower magnification, small fragments of keratinized lining epithelium that were either supported or not supported by a fibrous wall

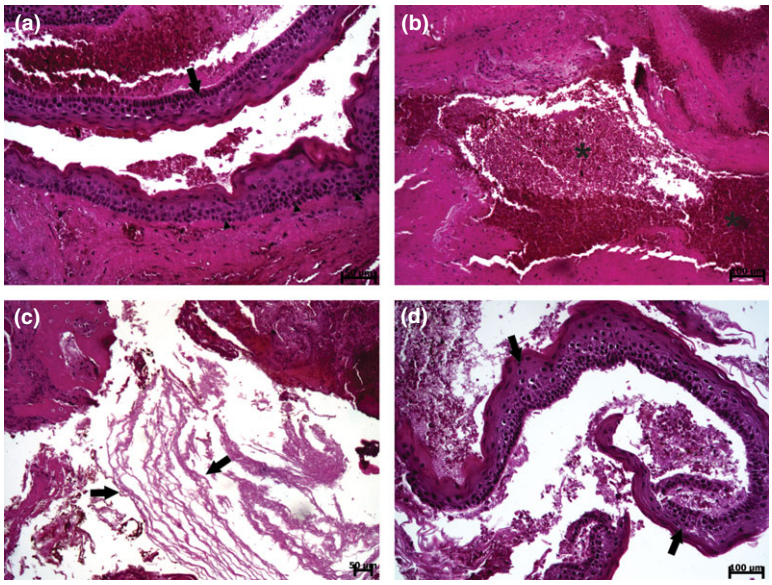

Figure 2 (a) Fibrous wall lined by corrugated parakeratinized squamous epithelium exhibiting a palisaded basal layer of cells (arrow); note basal layer with cuboidal cells as well (arrowhead); (b) cystic fibrous wall surrounding haemorrhagic content (asterisk); (c) remnants of fibrous wall surrounding numerous laminae of keratin (arrow); note absence of epithelial lining; (d) cystic epithelial lining with a parakeratinized and irregular surface (arrow).

were observed, confirming the histopathological diagnosis of KOT (Fig. 2d). The friable aspect of the lesion, significant haemorrhagic content and treatment by curettage may explain the histological profile described. Taken together, these findings indicate the need for rigorous microscopic analysis combined with clinical and surgical data to prevent an incorrect diagnosis. Although radicular cysts and periapical KOTs are histologically distinct lesions, their radiographic appearance may be similar (Wright *et al.* 1983, Pace *et al.* 2008, Tejasvi *et al.* 2010). In addition, these lesions are similar in terms of cytokine production in tissue cultures, suggesting that their mechanisms of expansion may involve similar biological mechanisms, except for infection (Hayashi *et al.* 2008). These features are important for the understanding of the biological profile of these lesions. However, in a clinical–pathological study of 100 cases of periapical lesions arising from endodontically treated teeth, Love & Firth (2009) attributed the absence of lesions such as KOTs and other developmental cysts to the diagnostic accuracy of the method used by the endodontist and general dentists, which should be able to differentiate lesions of nonendodontic origin and refer them for adequate treatment. Kuc *et al.* (2000), analysing biopsies from more than 800 periapical specimens, observed that 1% of these cases that were histologically classified as periapical lesions are unrelated to pulp necrosis. In fact, endodontists and general dentists are able to clinically differentiate periapical lesions of endodontic and nonendodontic origin, although their experience combined with new techniques effectively contribute to the diagnosis of periapical lesions that are unrelated to pulp necrosis. At the present institution, there are cases histopathologically diagnosed as periapical cemental dysplasia that were treated endodontically, probably due to the fact that the early stages of development of these lesions mimic inflammatory periapical lesions. In this occasion, the diagnosis is difficult to make based solely on radiographical findings.

In the present case, conventional root canal treatment and retreatment were ineffective. Thus, the periapical lesion was treated surgically as reported in previous studies

(Gagliani *et al.* 2005), followed by curettage (Natkin *et al.* 1984). The treatment of choice for keratocyst odontogenic tumours is variable, but it includes surgical enucleation followed by bone curettage (Bell & Dierks 2003, Ghali & Connor 2003, Mello *et al.* 2011). In this case report, the patient showed significant resolution of the maxillary radiolucency and no signs of recurrence eleven months after this therapy. Therefore, benign odontogenic tumours should be included in the differential diagnosis of periapical lesions.

Conclusion

Because different neoplastic lesions can mimic a large number of periapical lesions other than radicular cysts, it is important that the endodontist and general dentist are aware of their existence, especially when these lesions are located in the maxilla.

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