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Peripheral odontogenic keratocyst causing bone resorption: Report of two cases

Lado Lako Loro [a, b, *], Anne Christine Johannessen [b, c, d], Kathrine Skarstein [b, c, d], Sølve Hellem [b]

[a] Section for Oral and Maxillofacial Surgery, Aalesund Hospital, Norway
[b] Institute of Clinical Dentistry, Faculty of Medicine, University of Bergen, Norway
[c] Department of Pathology, Institute of Clinical Medicine, University of Bergen, Norway
[d] The Gade Laboratory for Pathology, Haukeland University Hospital, Norway

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ABSTRACT

Two cases of peripheral odontogenic keratocyst (POKC) of the gingiva presenting as asymptomatic nodules are described. The exceptional finding of cystic radiolucency in POKCs was observed on dental radiography in one case, which had a diameter of 10 mm. Conservative surgical excision was performed on both cases, and histopathological examination showed features consistent with keratocysts. POKC may recur and should thus be followed up like intraosseous keratocysts. No recurrences were observed after 10-years in our two cases. POKC is managed by conservative treatment. Recurrences should be excised more radically. Adjunct treatment with modified Carnoy's solution or topical 5Fluorouracil should be considered. POKC should be included in the differential diagnosis of asymptomatic gingival nodules.

1. Introduction

Odontogenic keratocyst (OKC), also known as keratinizing cystic odontogenic tumour, is a jaw cyst with a characteristic lining of parakeratinized-stratified squamous epithelium. OKC arise from remnants of the dental lamina or hamartia in the submucosa [1]. OKCs typically occur as intraosseous cysts in the jaws. They have a predilection for the posterior part of the body of the mandible and the ascending ramus and occur twice as often in the mandible as in the maxilla. Their size varies from a few millimetres to several centimetres [2]. Larger cysts may cause bone expansion and resorption. OKCs are generally asymptomatic but larger cysts may rupture and leak keratin to surrounding structures causing an inflammatory response, pain and swelling [2]. There is a preponderance of literature showing the aggressive, infiltrative and recurrent behaviour of OKCs [3]. Factors significantly associated with recurrence of OKC include variable age groups, size larger than 4 cm, multilocular cysts with cortical perforation, association with dentition, presence of daughter cysts, and epithelial budding [3].

Very rarely, cysts with the histopathological features of OKCs may present as peripheral cysts in the maxillofacial region. There are only 19 reported cases of peripheral OKCs (POKCs) in the gingiva and 20 in the soft tissues of the mouth [4–12]. Summeries of the POKC cases reported in English literature from 1975 to 2020 have previously been published [11,12]. Two additional cases of POKC in the maxilla canine-premolar region causing resorption of the alveolar bone are presented in this report.

Abbreviations: OKC, odontogenic keratocyst; POKC, peripheral odontogenic keratocyst.

* Corresponding author. Section for Oral and Maxillofacial Surgery, Aalesund Hospital, Moere & Romsdal Health Trust, Norway.
E-mail addresses: lado.lako.loro@helse-mr.no (L.L. Loro), Anne.Johannessen@uib.no (A.C. Johannessen), Kathrine.Skarstein@uib.no (K. Skarstein), solve.hellem@outlook.com (S. Hellem).

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2. Case 1

A 71-year-old man, in good general health, was referred by his dentist with a yellowish buccal symptomless nodule about one cm in diameter between the right upper canine and first premolar (Fig. 1). The patient had been unaware of the nodule until discovery by his dentist. On initial inspection, a dentoalveolar abscess was suspected, but was ruled out since the patient had no symptoms and teeth adjacent to the nodule were vital. Radiographic examination revealed a diffuse oval cystic appearance between the maxillary canine and first premolar (Fig. 2). A mucogingival flap was elevated to remove the nodule. A greyish, yellowish material was released upon rupture during surgery. The nodule was carefully dissected from normal tissue and sent for histopathological examination. A large crater-like defect in the buccal alveolar bone between the roots of the maxillary right canine and first premolar was seen after removal of the nodule (Fig. 3). The area was carefully curetted before the mucoperiosteal flap was repositioned and sutured with resorbable Vicryl 4.0 sutures.

3. Case 2

A 33-year-old woman, in good general health, was referred for treatment of a buccal nodule in the right canine-premolar region. The patient had been aware of the nodule for some time but had no symptoms.

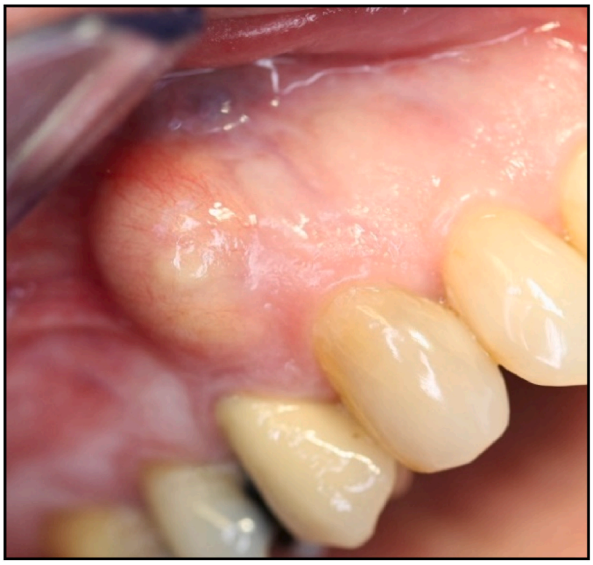

Fig. 1. Case 1. Preoperative clinical appearance of the peripheral odontogenic keratocyst showing a yellowish nodule about 10 mm in diameter between the maxillary right canine and first premolar.

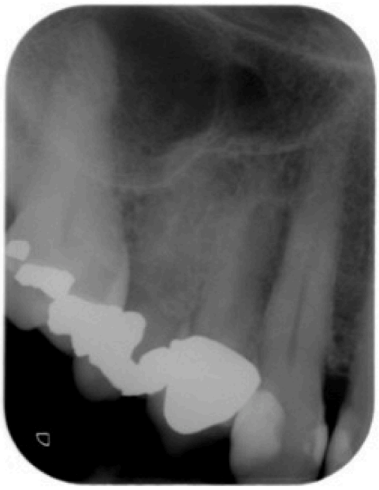

Fig. 2. Case 1. Dental radiograph showing a diffuse oval cystic appearance between the roots of the right maxillary canine and first premolar.

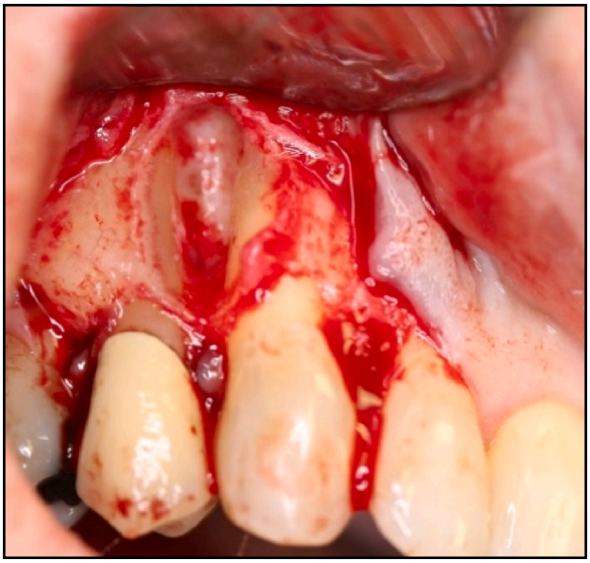

Fig. 3. Case 1. Intraoperative photograph showing a large area of alveolar bone resorption after removal of the odontogenic keratocyst.

On clinical examination, a firm well-defined bluish-grey nodule about 3 mm in diameter was observed on the buccal gingiva between the right upper canine and second premolar where the first premolar was missing. Both the right maxillary canine and second premolar were vital, and radiographs indicated no pathological changes in the region. An attempt was made to aspirate the contents of the nodule without success. However, from the needle puncture a greyish material could be pressed out. A decision was made to excise the nodule.

A mucogingival flap was elevated, exposing a soft tissue nodule associated with saucerization/cupping resorption of the alveolar bone between the right maxillary canine and second premolar (Fig. 4). The mass was enucleated and sent for histopathological diagnosis. The mucogingival flap was repositioned and sutured with resorbable Vicryl 4.0 sutures.

Histopathological evaluation of hematoxylin and eosin-stained sections from the two cases showed similar features. The soft tissue was lined with uniformly stratified squamous epithelium comprising 4–6 layers with a parakeratinized surface and a prominent palisaded hyperchromatic basal cell layer. The cyst wall from both biopsies consisted of fibrous connective tissue with sparse inflammatory cells (Fig. 5).

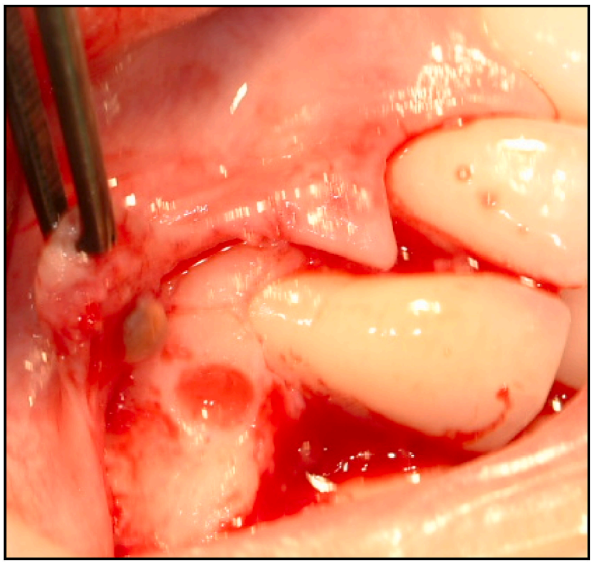

Fig. 4. Case 2. Intraoperative photograph showing part of the peripheral odontogenic keratocyst (brownish) at the base of the flap and saucerisation/cupping resorption of the alveolar bone.

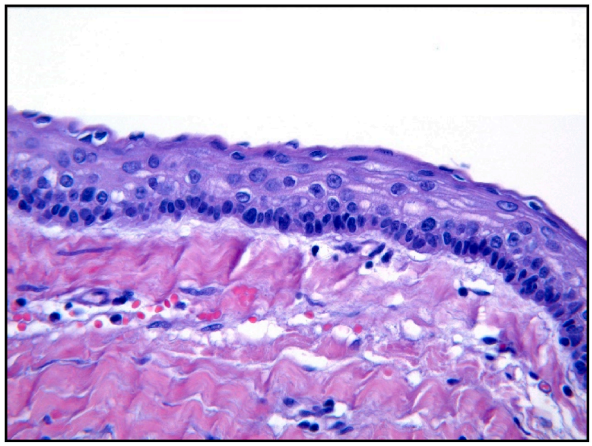

Fig. 5. Case 2. Photomicrographs of odontogenic keratocyst lining comprising a corrugated parakeratinized surface and palisaded basal cells with dark staining nuclei. A flat epithelial-connective interface and fibrous connective tissue with scanty inflammatory cells can be seen. Hematoxylin and eosin stain, 40-x magnification.

4. Discussion

A variety of odontogenic cysts and tumours can present in the soft tissues of the mouth. There is controversy in the literature as to the true nature of POKCs. Some authors maintain that they are soft tissue counterparts of intraosseous OKCs while others classify them as keratinizing variants of gingival cysts [5–10]. The location, clinical presentation, and presence of the "cupping resorption" during surgery in our two cases may suggest a lateral periodontal cyst. However, the histopathological features were entirely consistent with OKC. Gingival cysts of the adult are reported to have a range of histological features, including an "epidermoid type", with a keratinized stratified squamous epithelium and a lumen filled with ortho- or parakeratin [13]. A variant of gingival cysts of the adult termed the "keratocyst type" has a stratified squamous epithelium with palisading basal cells and a corrugated parakeratin layer [13]. Other reports in the literature maintain that the above features are more consistent with OKCs and thus, the "keratocyst type" of lateral periodontal cysts of the adult should be reclassified as OKCs [5,7]. Moreover, POKCs have been shown to contain daughter cysts and may recur after excision [5,6,10]. The two cases presented here exhibited the typical histological features of OKCs and have thus been diagnosed as POKCs. No recurrences were observed more than a decade after surgical removal in our two cases. POKCs should be followed up for a least 10 years.

A review of radiographic imaging of POKCs showed that most reported cases had no apparent pathologic changes [14]. Although the two cases presented here showed obvious clinical resorption of the alveolar bone, only the larger POKC could be imaged on radiographs as a cystic radiolucency in the alveolar bone. Clearly, the two cases show that POKCs can resorb bone like their intraosseous counterpart. The potential of dental lamina to resorb bone including grafted bone has been established [1]. However, cystic radiolucency on radiographs appears to be dependent on cyst size. Most reported cases in the literature are in the size range of two-five mm [11]. Larger cysts may be visible on radiographs, as seen in this report.

Presently, there are too few reports to ascertain the relative frequency, clinical behaviour and molecular basis of POKCs. These cysts have generally been managed by conservative surgical excision. This should be sufficient given the low recurrence rate. However, a more aggressive approach should be considered for recurrent cases. Excision including removal of the surrounding mucosa, electrocautery and peripheral osteotomy to remove dental lamina remnants, harmatia and daughter cysts should be considered. Recent reviews have shown that the use of modified Carnoy's solution or topical 5-Fluorouracil is effective in reducing the recurrence of keratocysts [1,15]. These adjunct measures may also be considered in the management of recurrent POKCs.

Immunohistochemical investigations have shown that POKCs express cytokeratin 7, 14 and 19 like intraosseous OKCs [16]. However, nothing is known about their genetic signature. Sporadic OKCs frequently exhibit mutations in the *PTCH 1* gene and other rare genetic aberrations of the SHH signalling pathway [17]. It would be of interest to investigate genetic changes in POKCs to ascertain their similarity to intraosseous keratocysts.

5. Conclusion

Peripheral odontogenic keratocyst is a very rare entity. Two cases exhibiting bone resorption are presented. The larger of the two cysts could be imaged on radiographs. Surgical excision is the main treatment. POKC may recur and should be followed up like intraosseous keratocyst for at least 10 years. Recurrences should be managed more aggressively with excision of surrounding soft tissues and peripheral osteotomy. Adjunct treatment with modified Carnoy's solution or topical 5-Fluorouracil may be considered. POKCs should be included in the differential diagnosis of asymptomatic gingival nodules.

Proof of consent

At the time of writing case 1 was deseased. No consent is necessary by Norwegian law. Consent to publish images was obtained from case 2.

Credit author statement

LLL: Conceptualisation, Writing – original draft and editing, ACJ: Histopathological examination, writing-reviewing and editing, KS: Histopathological examination, writing-reviewing and editing, SH: Writing-reviewing and editing.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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