Peculiar case of orthokeratinised odontogenic cyst: a peripheral counterpart of the intraosseous entity?

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ABSTRACT

We describe a case of an orthokeratinised odontogenic cyst (OOC) in a 49-year-old man, which presented as a submucosal fluctuant, non-tender, soft nodule distal to the inferior right first molar that could be considered a peripheral counterpart of the intraosseous OOC.

INTRODUCTION

Orthokeratinised odontogenic cyst (OOC) is a relatively uncommon developmental cyst that is separate from keratocystic odontogenic tumours (KCOT),[1, 2, 3, 4 and 5]in agreement with the 2005 WHO histological classification of odontogenic tumours.[6]
Unlike KCOT, OOC is lined predominantly by orthokeratinised epithelium, presents less-developed basal cells and a well-developed granular layer, is not associated with nevoid basal cell carcinoma syndrome, and behaves less aggressively, with a lower recurrence rate.[5]The term “peripheral odontogenic keratocyst” has been used to describe a gingival cyst in an adult with an epithelial lining characteristic of an odontogenic keratocyst. It has been proposed for lesions that occur as asymptomatic nodules, mimic the gingival cyst in adults, and usually have the histopathological features of a parakeratinised odontogenic keratocyst. It has therefore been described as the extraosseous counterpart of the KCOT.[5, 7, 8, 9 and 10]
We know of only one report of a peripheral odontogenic keratocyst that had orthokeratinised epithelium.[8]
We present an OOC in a 49-year-old patient that involved the mandibular retromolar region and may have been a peripheral counterpart of the intraosseous OOC.

CASE REPORT

A 49-year-old man was referred for the assessment of an asymptomatic gingival nodule in the retromolar region of the right mandible. The patient gave no history of pain or discomfort, and his history was unremarkable. On clinical examination there was a soft submucosal nodule distal to the inferior right first molar in the retromolar region, which was fluctuant and not tender on palpation.
A previous panoramic radiograph (taken 7 years previously) showed no pathological lesion (Fig. 1).
A current panoramic radiograph and cone beam computed tomographic scans showed a unilocular, well-circumscribed, radiolucent lesion associated with a depression in the alveolar bony crest (Fig. 2 and Fig. 3).
The lesion was enucleated under local anaesthesia. After the incisions had been made and a mucoperiosteal flap reflected, the lesion was removed to show a well-defined defect in the surface of the adjacent alveolar bone. That was curetted to ensure removal of any residual epithelial remnants and the flap closed primarily with a 3/0 silk suture.

Histopathological examination showed a cyst, the wall of which was lined with orthokeratinised squamous epithelium. The flattened basal layer of cells lacked palisading, and a prominent granular layer of cells was apparent. These findings identified the lesion as an OOC.
Healing was uneventful with no sign of infection. At follow-up 24 months later there were no signs of recurrence.

DISCUSSION

Peripheral KCOT have been considered to be rare odontogenic lesions that present the same microscopic features as their central intraosseous counterpart.[7] According to some authors, they reflect the biological behaviour of intraosseous KCOT because of bony resorption and recurrence,[5 and 8]whereas others think that although peripheral and central KCOT share the same histological features, peripheral lesions are not aggressive and differ from ordinary KCOT.[10]
We know of only one reported case of a peripheral OOC, by Chehade et al.[8]
Clinically, peripheral KCOT often present as asymptomatic and fluctuant nodules, with pressure ("cupping") resorption or fenestration of the adjacent alveolar bone.[5]Because of bony resorption and recurrence, peripheral odontogenic keratocysts seem to reflect the biological behaviour of their central counterparts.[8]
Our patient had the clinical aspect of an asymptomatic and fluctuant nodule in the mandibular retromolar region, with pressure resorption of the adjacent alveolar bone. Histologically it was diagnosed as an OOC, with a thin, uniform, orthokeratinised lining epithelium.
The differential diagnoses of peripheral KCOT and OOC include other gingival cysts or tumours, such as the peripheral counterparts of several central odontogenic tumours (ameloblastoma, adenomatoid odontogenic tumour, calcifying epithelial odontogenic tumour, odontogenic fibroma, squamous odontogenic tumour, and granular cell ameloblastic fibroma).[5 and 7]Peripheral odontogenictumours have a benign, non-aggressive clinical course, are located within the gingival soft tissues, do not involve bone (except for occasional instances of underlying cupping resorption), and have histomorphological features comparable to those of their central counterparts.[5] Therefore, by analogy we think that our case is a peripheral counterpart of the OOC.

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