Keratinizing dentigerous cyst
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Abstract
Keratinizing dentigerous cyst is a rare entity. This article reports a case of keratinizing dentigerous cyst associated with an impacted mandibular canine. Clinical and radiological features, cone-beam computed tomography findings and histological features of the case are reported along with a discussion on keratinizing odontogenic cysts and the need for follow-up.

Keywords: Dentigerous cyst, keratinizing, odontogenic

Introduction
The dentigerous cyst is a common non-inflammatory odontogenic cyst of the oral cavity. Its pathogenesis involves the accumulation of fluid between the unerupted tooth crown and surrounding follicle, giving rise to the characteristic clinical and radiographic finding of a cystic lesion surrounding the neck of the tooth (Latin: Dens, tooth + gerere, to bear). Treatment involves surgical enucleation or marsupialization with or without preservation of the impacted tooth, followed by histopathological evaluation to rule out an odontogenic keratocyst (OKC), ameloblastoma, or rarely, malignant transformation. We report an unusual histopathological variant of the dentigerous cyst, the keratinizing dentigerous cyst, which has been reported only once previously in literature.

Case Report
The present case is about a 21-year-old male patient of Indian origin reported with a complaint of mild pain and swelling in the lower front region of the jaw for the past 1 year. He claimed that the pain was not severe and occurred intermittently. The patient’s medical history was not otherwise significant. Extra-oral examination revealed a diffuse swelling over the chin. Intra-oral examination revealed a diffuse swelling in the lower anterior vestibule extending from the region of 43 to 36. On palpation, the swelling was firm in consistency and egg shell crackling was elicited in some areas. 83 was retained and 43 was clinically absent.

The panoramic radiograph showed a well-defined unilocular radiolucency extending from the mesial aspect of 44 to the mesial root of 36, extending to the mental foramen on the left side. 43 was impacted and the cystic lesion surrounded the crown of the tooth and extended for some distance along the mesial aspect of the root (circumferential variant). The margins of the lesion were well-defined, with a sclerotic border. Cone-beam computed tomography was also performed and revealed involvement of roots of 42, 41, 31, 32, 33, 34, 35 and mesial root of 46 [Figure 1].

An excisional biopsy of the lesion was performed under general anesthesia with conventional pre-operative medication cover and endotracheal intubation. A mucoperiosteal flap was raised from 44 to 37 region and the lesion was detached from the soft-tissue using blunt dissection and curetted out from the bony walls. 32 was extracted as the bony walls were destroyed. Complete hemostasis was achieved and the wound closed primarily with Vicryl sutures. External dynaplast compression dressing was given for 24 hours. Root canal treatment was carried out for 41, 42, 31, 33, 34 and 35. A vitality test was advised for 46 and treatment planned accordingly. The specimen was sent for histopathological examination to rule out an OKC or an ameloblastoma. The patient was called for review and a post-operative Osteoprotegerin after 1 month.

Macroscopically, a cyst in-toto with a tooth, measuring 40 × 30 × 40 mm in size [Figure 2] and four bits of tissue curetted from the surgical site were sent for histopathological evaluation. The cyst was sectioned in half and processed along with the four bits of curetted tissue.

Microscopically, a fibrous connective tissue capsule in association with a non-keratinized cystic lining epithelium of varying thickness was observed. The lining epithelium
exhibited focal areas of prominent granular cell layer. The lumen exhibited keratin flakes [Figure 3]. The four bits of curetted tissue microscopically showed the presence of hematoxyphilic substance, suggestive of keratin.

Discussion

We report a case of keratinizing dentigerous cyst which, to the best of our knowledge, has been reported only once previously in the literature. Philipsen in 1956 suggested the term OKC for all Odontogenic cysts, regardless of type, showing keratinization of the epithelium. More recently, an OKC is defined by other characteristics of the epithelium such as basal palisading, hyperchromatism of nuclei and cell thickness of the epithelium and not merely the presence of keratinization. The term “keratinizing odontogenic cyst” has been suggested for any cyst, regardless of the type, that shows keratinization.

Characteristically, the epithelial lining of a dentigerous cyst is not keratinized and most of those that have been described as keratinized have been ascribed to adjacent OKCs. However, taking into consideration the established criteria, the present case did not show any of the features that currently define OKCs. The patient was followed up after 1 year [Figure 4] and showed no clinical or radiographic signs of recurrence.

The significance of keratinization in odontogenic cysts is not fully known. Keratinization is the final product of epithelial cell differentiation. However, dentigerous cysts, thought to arise from reduced enamel epithelium, are products of end cells, i.e. cells that have completed synthesis (enamel formation). Considering the age of the patient, it is possible that the dentigerous cyst is a primordial variant, arising from more primitive cells of the developing enamel organ.

Figure 1: Cone-beam computed tomography shows a unilocular lesion surrounding the crown of impacted 43 and extending along the mesial aspect of the root (circumferential type)

Figure 2: Gross finding of a tooth within a cystic bag filled with keratinaceous material

Figure 3: Focal areas of keratinizing cystic lining epithelium exhibiting a prominent granular cell layer. The cystic lumen is filled with keratinaceous material (H and E, x10)

Figure 4: (a) Intra-oral-labial aspect of the patient 1 year postoperatively (b) Intra-oral-lingual aspect of the patient 1 year postoperatively (c) Orthopantamograph (OPG) of the patient 1 year postoperatively
The keratinizing dentigerous cyst is an uncommon variant of the dentigerous cyst. Long term follow-up of patients presenting with a keratinizing dentigerous cyst is advised to observe its potential for recurrence or malignant transformation since very little is known about this unusual entity.

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