Odontogenic keratocyst: A peripheral variant

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Abstract

Odontogenic keratocyst, which is developmental in nature, is an intraosseous lesion though on rare occasions it may occur in an extraosseous location. The extraosseous variant is referred to as peripheral odontogenic keratocyst. Though, clinically, peripheral odontogenic keratocyst resembles the gingival cyst of adults, it has histologic features that are pathognomonic of odontogenic keratocyst. This article presents a case of this uncommon entity.

Key words: Keratocyst, odontogenic cyst, peripheral odontogenic keratocyst

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Introduction

A variety of cysts are known to occur in the head and neck region. Amongst these the odontogenic keratocyst (OKC) is noteworthy because of its ability to attain a large size before any clinical signs and symptoms develop, its high recurrence rate, and an association with nevoid basal cell carcinoma syndrome (which has been reported by various authors).[1,2] Various dental cysts can exhibit keratinization in their epithelial lining, but OKCs predominantly show parakeratinization. The term 'odontogenic keratocyst' was coined by Philipsen.[1] The defining histologic feature – the presence of parakeratinization – is unique to OKCs among the multitude of inflammatory and developmental cysts that occur in the jaws. The presence of keratin in association with the other characteristic histologic findings is so distinctive that the diagnosis is usually obvious on microscopy.[3] These cysts are more common in males than females and tend to involve the mandible far more frequently than the maxilla. Within the mandible they are found most commonly at the angle of the mandible and often extend into the ascending ramus and the body of the mandible.[1] Their frequent presentation in the ascending ramus of the mandible has been explained by the hypothesis that offshoots of the dental lamina are probably responsible for the development of keratocysts in this region, and this has prompted Toller[4] to suggest the term 'laminal cyst.' Reports of various studies suggest that these cysts can occur anywhere in the jaws, including in the midline of the mandible and maxilla and the globulomaxillary area in the maxilla.[2]

On rare occasions the cyst can develop in the gingiva, as a soft tissue counterpart of OKC, from the remnants of dental lamina. This soft tissue OKC can clinically mimic the gingival cyst of adults.[5] According to available data only 14 cases of this lesion have been reported till date.[5–11] Dayan *et al.*[8] were the first to report this unusual entity and had suggested the term 'peripheral odontogenic keratocyst (POKC)' for the same. This peripheral counterpart of OKC appears to be less aggressive and has a lower recurrence rate than its intraosseous form, as has also been observed in peripheral variants of certain odontogenic tumors.[12] In this article we report a patient with POKC, thus adding one more case to the short list of reported cases of this rare entity.

Case Report

A 56-year-old male patient reported to the dental clinic with the complaint of a swelling in the anterior aspect of the left side of the palate. On clinical examination, the patient was completely edentulous. A swelling was observed on the anterolateral aspect of the hard palate, extending

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from the incisor to the premolar region and measuring 2.5×2.0 cm. The overlying mucosa appeared normal [Figure 1]. On palpation, the swelling was soft in consistency and had well-defined borders. The patient also gave a history of occasional discharge from the swelling on application of pressure. Radiographic examination did not show any bone involvement. Based on the clinical features it was thought to be a cystic lesion. On excisional biopsy the lesion was found to be completely extraosseous and was excised *en masse*. The tissue was submitted for histopathological examination [Figure 2].

Microscopically, hematoxylin and eosin (H and E)–stained sections revealed a cystic lumen containing keratin flakes and inflammatory cells. The lumen was lined by parakeratinized stratified squamous epithelium, which showed thickening and loss of keratinization at places. The epithelium showed surface corrugations and palisaded basal columnar cells. The epithelium–connective tissue interface was flat. The connective tissue wall of the cyst was uniform in thickness and had a chronic inflammatory cell infiltrate at places [Figures 3 and 4].

Discussion

The term 'cyst' comes from the Greek word *kysits* which means 'sac' or 'bladder.' Cysts are relatively common and may be encountered virtually in any organ or tissue in the body. The head and neck region and the jaws, in particular, collectively comprise one of the more common sites for the occurrence of cysts. The frequency of the occurrence of cysts within the jaws is usually ascribed to the peculiar embryology of the facial skeleton and the presence of teeth which, both pre- and post-eruptively, may be associated with epithelium and epithelial residues, which are potentially capable of being involved in the genesis of cysts.[1]

Among all the jaw cysts, OKCs have received special attention in the literature because of their relatively high recurrence rate and also due to their tendency to grow within the medullary spaces of the bone in the anteroposterior direction.[13] This growth pattern rules out the possibility of the detection of these lesions at an early stage. On rare occasions, this cyst can occur in the soft tissues, when it

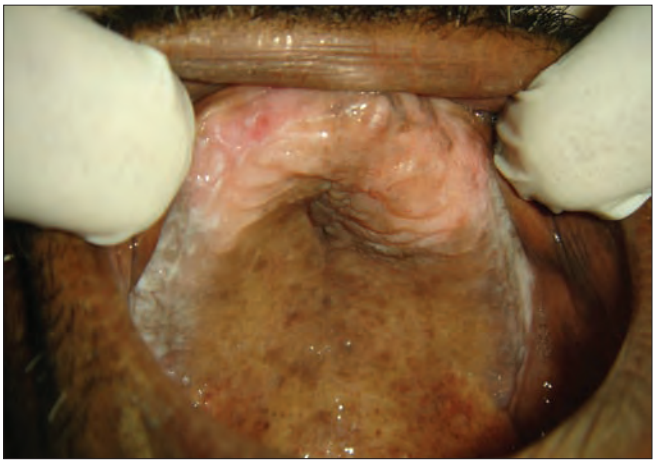

Figure 1: Palatal swelling in the incisor–canine region

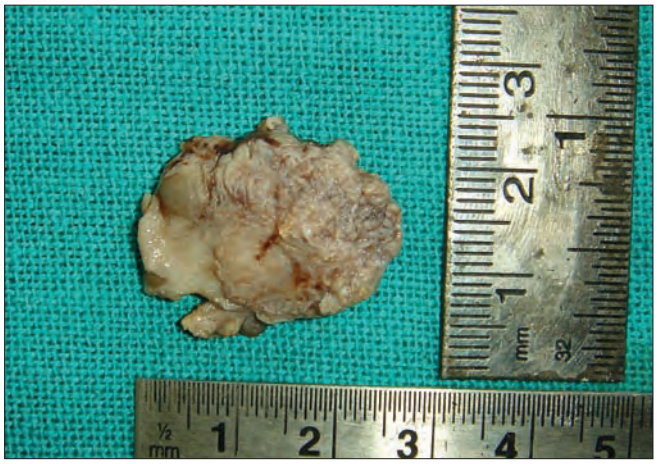

Figure 2: Gross specimen

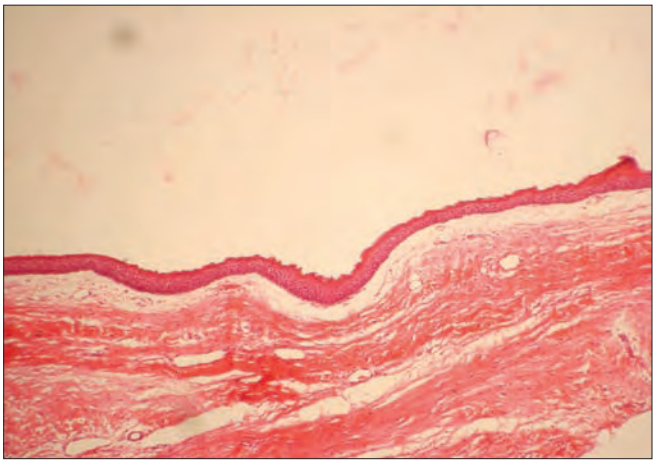

Figure 3: Cyst lining showing corrugated surface and lack of rete ridges (H and E, ×40)

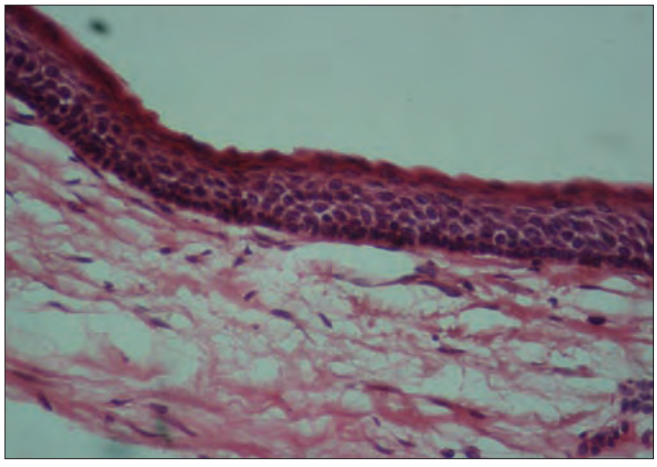

Figure 4: Cyst lining showing corrugated parakeratin layer and prominent palisaded basal cell layer (H and E, ×100)

Table 1: Case reports of peripheral odontogenic keratocyst

Author	No. of cases reported	Age (in years)	Gender	Site	Radiographic findings
Stoelinga *et al.*[6]	1	-	-	Maxillary gingiva and premolar	-
Buchner and Hansen[7]	2	-	-	Buccal gingival	-
		-	-	Buccal gingival	-
Dayan *et al.*[8]	1	42	M	Left maxillary gingiva, canine, and 1st premolar	None
Chehade *et al.*[9]	5	37	M	Right mandibular, lateral incisor, and canine	-
		66	F	Left maxillary gingiva, and 1st and 2nd premolar	-
		70	M	Left mandibular gingiva, canine, and 1st premolar	-
		57	F	Right maxillary gingiva and canine	-
		42	M	Right posterior mandibular alveolar mucosa	-
Ide *et al.*[10]	2	38	F	Left mandibular gingiva and lateral incisor	None
		46	F	Right mandibular gingiva and 1st premolar	None
Chi *et al.*[5]	2	81	F	Left mandibular gingiva, canine, and 1st premolar	None
		64	F	Left maxillary gingiva and premolar area	None
Faustino *et al.*[11]	1	57	F	Left mandibular gingiva and premolar	Superficial erosion or depression of the alveolar bone
Present case	1	56	M	Left maxillary gingiva, lateral incisor, and canine	None

clinically resembles the gingival cyst in adults (GCA). Many investigators have noted that both these cysts (OKC and GCA) originate from remnants of the dental lamina.[1,8] GCA may arise from traumatic implantation of epithelium[14] but, according to Dayan *et al.*[8] and Chehade *et al.*,[9] no history of traumatic insult or surgical intervention has been recorded in a patient with POKC.

Due to the dearth of reported cases, the exact epidemiology of this lesion cannot be established. As per the review by Chi *et al.*,[5] the average age of occurrence is 54 years (range: 37–81 years). Our patient, at 56 years of age, was also within this age range. According to these authors the most frequent anatomic location of the lesion is the canine–premolar region; however, in the present case, the lesion involved the incisor and canine region. This entity shows no significant changes on radiographic examination, though pressure/cupping resorption may be observed. Our case also did not exhibit any radiographic changes, though slight gnawing away of bone was noted at the site after surgical excision. The treatment commonly used for intraosseous OKC is total enucleation with peripheral ostectomy;[13] however, since POKC is considered to be less aggressive, we employed a more conservative line of treatment in the present case.

The histopathological features of the present case of POKC were similar to those of a traditional OKC, with parakeratinized stratified squamous epithelium that was 6-8 cell layers thick, a thin spinous layer, intercellular edema at places in the spinous layer, a flat and weak epithelium–connective tissue interface, and a thin connective tissue wall made up of immature collagen fibers; similar findings have been reported in other reported cases.[5,10,13] Certain areas in the present case exhibited epithelial thickening and a mature fibrous connective tissue wall; these features may be attributable to the long-standing nature of the lesion. This altered nature of epithelium and connective tissue can also be credited to the inflammatory component of the lesion in the present case because inflammation is known to affect the overlying epithelium (atypical changes/hyperplasia) and the surrounding connective tissue [Table 1].[15]

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