



A CASE REPORT OF KERATOCYST ODONTOGENIC TUMOR MIMICKING A LATERAL PERIODONTAL CYST

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Abstract

Keratocyst odontogenic tumor is a benign cyst usually affecting the posterior segments of the mandible in the second and third decades of life. This lesion is detected by a multi locular radiolucency in radiographs. This cyst is rare in anterior segments of the mandible and older ages. The aim of this case report is to report a 63-year-old male affected by keratocyst odontogenic in the anterior segment of the mandible.

Key words: Biopsy, Diagnosis, Keratocyst odontogenic tumor, Lateral periodontal cyst, Mandible.

Introduction

Odontogenic keratocyst is a benign however a locally aggressive cyst, which was introduced for the first time by Phillippsen in 1956. The most important consideration about this cyst is great tendency to recurrence and extension prior to exposure in oral cavity (1).

It has been proposed that this cyst arises from rests of dental lamina in mandible or maxilla. However, some other studies have suggested that the proliferation of the basal layer of oral cavity lining cells may result in formation of this cyst (2).

Odontogenic keratocyst is characterized by its unique histopathologic features and multiple recurrences. The most commonly affected sites are the posterior body of mandible. However, the lesion can occur in any region in mandible and maxilla. According to radiographic features, the lesion alters from an asymptomatic unilocular radiolucency to a multilocular radiolucency (3). keratocyst Odontogenic is more prevalent in males and the most commonly affected groups are in the 2nd and 3rd decades while it is less prevalent in older ages (4).

The aim of this case report is to report a 63-year-old male affected by keratocyst odontogenic in the anterior segment of the mandible.

Case Description:

A 63-year-old man with swelling in the anterior part of the mandible was referred to the Oral Medicine Department of Qazvin University of Medical Sciences, Iran. According to the patient's history, the swelling was detected from 5 months earlier while it was gradually enlarged in the past month. There were no significant medical problems and he was not taking any medication.

Extra oral examination revealed no lesions, masses, or swelling. In clinical intraoral examination, a firm and well-defined 1.5 × 2 cm swelling was observed in the left lateral and canine site of the mandible. The surface of the lesion was completely intact, and the color was similar to the normal mucosa with telangiectasia in some sites (figure 1).

In radiologic examination, a well-defined radiolucency was detected in the lateral and canine region, which was not corticated (figure 2). An aspiration biopsy was performed in order to differentiate cyst and tumor. The result of aspiration was indicative of pus, which is a sign of infection superimposed on the site.

In the microscopic sections, a cyst lined with a wavy parakeratinized stratified squamous cell epithelium was observed. The epithelium was composed of 4-8 layers of uniformly thin cells, which possesses a hyperchromatic palisading pattern. Detachment of the epithelium from the inferior connective tissue was evident in some sections. Interface of epithelium-connective tissue was flat and devoid of rete ridges. The connective tissue located beneath the epithelium was consisted of fibrocollagenous tissue among few congested blood vessels and some sites of hemorrhage (figure 3).

According to histopathologic examinations, the diagnosis was made as keratocyst odontogenic.

Discussions

Keratocyst odontogenic is an inflammatory lesion comprising 4-12% of the jaw cysts. According to World Health Organization (WHO) category, this cyst is classified as developmental and non-inflammatory odontogenic cyst which arises from dental lamina rests (7). Recently, WHO regarded this lesion as keratocyst odontogenic tumor (KCOT) and defined it as "a benign uni- or multi-cystic, intraosseous tumor of odontogenic origin (8). Generally, this lesion is a solitary lesion unless associated with a hereditary condition (9, 10).

The previous studies suggest that the cyst is prevalent in the 5th and the 6th decades, while most of the studies reveal the predilection of this cyst to the 2nd and 3rd decades and its association with an impacted tooth (5, 6, 11). Therefore, this cyst is not a prevalent finding in our patient's age range. Men are more commonly affected by this lesion, (6, 11) and majority of keratocyst odontogenic are detected in mandible, especially ramus, posterior part of the mandible, and third molar region (6, 7, 11, 12). In this case, in accordance to the literature, mandible was the affected jaw, but on the contrary, the epicenter was in the anterior part of the mandible. The symptoms include pain, swelling, drainage and bone perforation; however, in half of the cases, the cyst is asymptomatic and is discovered in routine radiographs. In the present case, swelling was the chief complaint of the patient though this swelling was due to the infection superimposed on the lesion that was confirmed by the aspiration.

The radiographic characteristics of a keratocyst odontogenic are as follow: (1) a multilocular radiolucency with a well defined and usually scalloped corticated border; (2) expansion in the medial side and extension towards length of the mandibular bone; (3) displacement of a developing

tooth, separation or resorption of the erupted tooth's roots (5). Therefore, according to the radiologic features and the site of the lesion, the cyst mimicked a lateral periodontal cyst. Lateral periodontal cyst is a rare developmental cyst located interdental. Lateral periodontal cyst is usually symptomless and possesses a predilection to men and fifth decade. It is commonly located in the mandibular canine- premolar area (13).

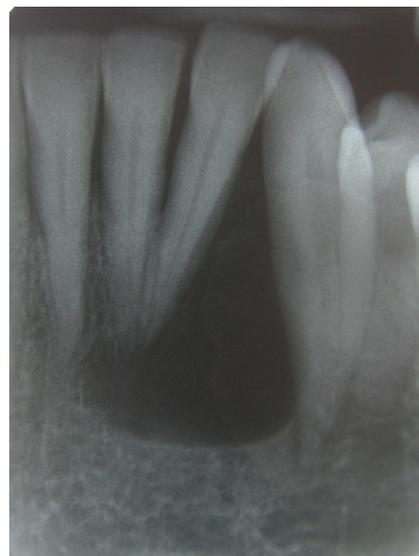
To date, a great number of studies have aimed to reveal the pathogenesis of keratocyst odontogenic. Results of several researches have proposed a role for point mutation in PTCH gene in pathogenesis of nevoid basal cell carcinoma syndrome, a hereditary condition characterized by several recurrent keratocysts odontogenic, (14) and sporadic keratocysts odontogenic as well (15-18). On the other hand, other studies revealed over expression of some genes such as EGFR and ki-67, cytokeratin 6B, epidermal growth factor receptor ERBB3, and glioma-associated oncogen homologue 1 (GLI1) (19, 20). Protein expression analysis of keratocyst odontogenic, have revealed that this specimen are positive for PCNA, Ki67 and p53 protein, markers routinely detected in neoplasms (21). However, specific etiology of this condition still remains unknown (22).

Treatment of keratocyst odontogenic tumors is can be affected by several factors such as age, location, size, extension of the lesion, and soft tissue involvement (23). Based on these factors, the treatment falls into two categories: aggressive or conservative. Peripheral ostectomy, chemical curettage with Carnoy's solution, cryotherapy, electrocautery and resection are considered as aggressive treatment approaches. Conservative treatments include enucleation, with or without curettage, or marsupialization (24,25). Consecutively, the ideal treatment must induce the least morbidity and decrease the risk of recurrence (23).

Figure 1- Swelling in the left lateral and canine segment of mandible



Figure 2- Radiographic examination revealed a corticated well defined radiolucency between





Conclusion:

According to the age, gender, site of the lesion, and other clinical manifestations in the present patient, keratocyst odontogenic wouldn't be categorized in the first choices of differential diagnosis. This case report emphasizes that radiographic and clinical features do not provide sufficient information on cysts. Therefore, definite diagnosis of whole lesions must be based on histological examinations.

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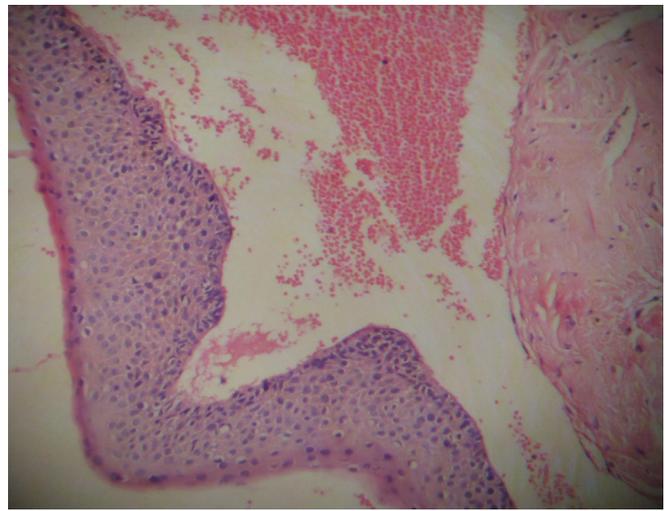


Figure 3- Microscopic appearance of the lesion indicative of keratocyst odontogenic tumor