

KERATOCYSTIC ODONTOGENIC TUMOR OF THE MAXILLARY ANTERIOR REGION MASQUERADING AS A RADICULAR CYST

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ABSTRACT

Keratocystic odontogenic tumor (KCOT) is a benign odontogenic neoplasm which is characterized by aggressive behavior and a high recurrence rate. KCOTs have a predilection for the angle and ascending ramus of the mandible. Maxillary KCOTs are usually associated with nevoid basal cell carcinoma syndrome. Very few cases of non-syndromic KCOT crossing maxillary midline have been reported. In this article, we report a rare case of KCOT which presented as a multilocular radiolucency crossing midline and involving periapical area of maxillary incisors. The lesion thus simulated a periapical cyst. However, the lesion was proved to be a KCOT on histopathology. Thus this report highlights the need for histopathological evaluation of periapical lesions for accurate diagnosis and treatment plan.

Keywords: Anterior Maxilla, Keratocystic Odontogenic tumor, Radicular cyst.

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INTRODUCTION

Keratocystic odontogenic tumor (KCOT) was traditionally termed as odontogenic keratocyst [1]. It was reclassified by WHO (2005) as a benign tumor, and the use of term KCOT was recommended as it reflects on its neoplastic nature [1]. Often due to atypical clinical and radiographic presentations, KCOTs have been misdiagnosed as odontogenic cysts (viz., dentigerous cyst, radicular cyst, and lateral periodontal cyst) and have been undertreated [2-4]. Since KCOT has an aggressive biological behavior, it is essential to distinguish it from odontogenic cysts [5]. Herein, we report a case of KCOT which presented as a multilocular radiolucency in periapical region of maxillary anterior teeth and hence was erroneously diagnosed as radicular cyst.

CASE REPORT

A 45-year-old male patient presented with a chief complaint of swelling in labial aspect of the upper front region of jaw. The patient had first noticed the swelling 6 months ago. The patient did not reveal any history of trauma to maxillary anterior teeth. Intraoral examination revealed swelling in labial aspect of 11, 21, and 22. Moreover, the swelling was approximately 2x2 cm in size (Fig. 1a). There was obliteration of labial sulcus extending from distal aspect of right maxillary central incisor to mesial aspect of the left maxillary lateral incisor. On palpation, the swelling was non-tender and was cystic in nature. Teeth 11, 21, 22 were grade I mobile and non-vital, but none of the teeth were carious. A panoramic radiograph revealed an ill-defined multilocular radiolucency involving the apices of 11, 21, 22 (Fig. 1b). Displacement of the root of left maxillary lateral incisor was evident on radiograph. Overall radiographic changes were suggestive of radicular cyst. However, the history and clinical examination were not in support of radicular cyst. An incisional biopsy was done.

On histopathological examination, hematoxylin and eosin stained sections revealed cystic epithelium which was lined by 5-8 cell layer thick para keratinized stratified squamous epithelium showing corrugated surface. The epithelium-connective tissue interface was flat. The basal cells were tall columnar with hyperchromatic palisaded nuclei placed away from the basement membrane; hence histopathological report of KCOT was given. The patient was taken up for surgery under general anesthesia. Extraction of 11, 21, and 22 was done, and cyst

enucleation was carried out. During the surgery, multilocular lesion involving the roots of maxillary anterior teeth was evident (Fig. 1c). Carnoy's solution was applied over surgical site. Moreover, platelet rich plasma was placed in the osseous defect. Closure was done with 3-0 silk suture. The patient was prescribed tranexamic acid mouthwash to prevent postsurgical bleeding. Histopathological examination of enucleated specimen confirmed the diagnosis of KCOT.

DISCUSSION

KCOT is a benign uni- or multicentric tumor of odontogenic origin. The majority of cases of KCOT occur in second to the third decade of life and have a male predilection [5]. KCOT affects mandible more commonly than maxilla, with a predilection for molar-ramus area (65-83%) [5]. Till date, only a handful of cases has been reported of KCOT crossing maxillary midline [6]. When KCOT occurs in maxilla, diagnostic difficulties arise owing to lack of characteristic clinical and radiographic features [2]. The maxillary KCOTs are usually smaller in size. This may be explained by the fact that maxillary KCOT are more frequently infected than mandibular lesions hence they are detected early before they grow to a larger size [1,5].

In the present case, radiographically an ill-defined multilocular radiolucency was noted in relation to 11, 21 and 22. The abovementioned teeth were non-vital and root resorption of 11 was evident. Hence, differential diagnosis of radicular cyst was considered. However, none of the involved teeth were carious and the patient did not have a history of trauma which ruled out the possibility of radicular cyst. Other differential diagnosis considered were nasopalatine duct cyst, central giant cell granuloma, KCOT, ameloblastoma and other odontogenic tumors. But on incisional biopsy diagnosis of KCOT was rendered.

Histopathologically KCOT is characterized by the presence of cystic epithelium lined by 5-8 cell layer thick stratified squamous epithelium [1]. Basal layer shows palisading and the nuclei are hyperchromatic. The superficial epithelium is para keratinized and often shows corrugation. The epithelium-connective tissue interface is flat. These histopathological features were evident in the present case (Fig. 2).

In the literature, a few cases of KCOT involving the periapical areas which were clinically misdiagnosed as a radicular cyst have been

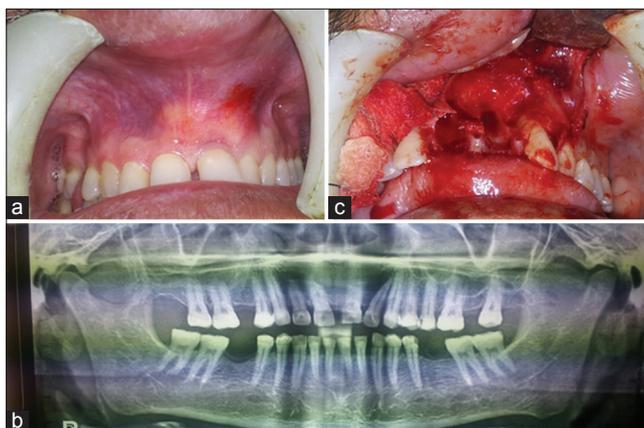


Fig. 1: (a) Swelling on the maxillary anterior region in relation to 11 and 21, (b) orthopantomograph showing multilocular radiolucency in maxillary anterior area in relation to 12,11,21 and 22, (c) intraoperative photograph showing a multilocular lesion involving the roots of maxillary anterior teeth

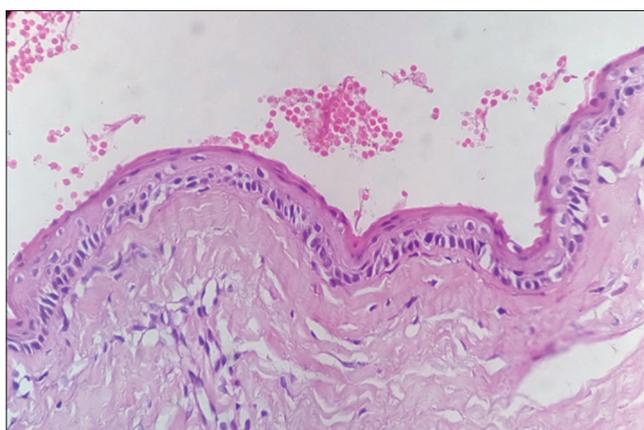


Fig. 2: Photomicrograph showing H and E stained section showing 5-8 cell layer thick stratified squamous epithelium. The basal layer of the cystic epithelium is made of tall columnar cells with palisaded hyperchromatic nuclei. The superficial epithelium is parakeratinized with corrugated surface

reported [4,6,7]. However, the diagnosis was established only after histopathological evaluation. This highlights the pivotal role of histopathology in diagnosis of periapical lesions and it is essential to submit all the excised tissue from the periapical region for histopathological examination.

Based on the histopathological diagnosis, enucleation of the lesion was done along with application of Carnoy's solution. The involved teeth,

i.e., 11, 21 and 22 were extracted. The use of platelet-rich plasma in healing of osseous defect is established [8]. Hence at the time of surgery, platelet rich plasma was placed in the osseous defect to augment healing of the bone. Maxillary anterior teeth are important from the point of view of esthetics and function. Hence, rehabilitation of missing teeth was planned for the patient.

The rationale behind extraction of the teeth in relation to the tumor is that there might be residual epithelium in relation to the roots of the teeth which may lead to recurrence of the tumor [9]. Thus, use of Carnoy's solution and extraction of involved teeth significantly reduces the risk of recurrence of KCOT [9-11]. Achieving hemostasis after surgery is of utmost importance and tranexamic acid is often used as hemostatic agent [12,13]. In this case, the patient was prescribed tranexamic acid mouthwash to prevent post-operative bleeding.

Thus, we present an unusual case of KCOT mimicking radicular cyst. If the diagnosis is based only on clinical and radiographic features, it can be erroneous. Hence, histopathological examination is necessary to reach to an appropriate diagnosis and to initiate accurate treatment.

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