



OLGU SUNUMU / CASE REPORT

Keratocystic odontogenic tumour occurring in an unusual location

Sıradışı bir yerde meydana gelen keratokistik odontojenik tümör

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Abstract

Keratocystic odontogenic tumor, formerly known as odontogenic keratocyst is a benign intraosseous neoplasm of the jaw. Maxillary involvement is very unusual with few cases being documented in literature. Hence we present the case of a 35-year-old male patient with keratocystic odontogenic tumor in the anterior maxilla.

Key words: Keratocystic, odontogenic, tumor, maxilla.

Öz

Keratokistik odontenik tümör, daha önce Odontojenik Keratosis olarak bilinen, çenenin iyi huylu bir intraosseöz tümörüdür. Maksiller tutulum literatürde az sayıda olgu ile belgelenmiş olup, çok sık karşılaşılmayan bir durumdur. Bu yazıda anterior maksillada keratokistik odontojenik tümörlü 35 yaşında bir erkek hasta sunulmuştur.

Anahtar kelimeler: Keratokistik, odontojenik, tümör, maksilla.

INTRODUCTION

Philipsen described “Odontogenic Keratocyst” in 1956¹ World Health Organization (WHO) coined the term “Keratocystic Odontogenic Tumor” (KCOT). WHO “recommends the term keratocystic odontogenic tumour as it better reflects its neoplastic nature. It is defined as “a benign uni or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior”².

The overall ratio of occurrence of the tumour in the mandible to maxilla of was 5:1. 81% of the lesions were located in the mandible, most often in the body [20%], angle [18%] and vertical ramus [10%]. In 30% of the mandibular cases the tumour involved more than one anatomical site, most frequently affecting the body, angle and vertical ramus [10%]. Only 16% of the lesions occurred in the maxilla, mainly in the posterior region [13%]³. It has also occurred in unusual locations such as the anterior portion of the maxilla, the maxillary sinus,

and the maxillary third molar⁴. It has a slight male predilection. It occurs in the second and third decades of life⁵.

Owing to its aggressive nature, pathognomonic microscopic features and high recurrence rate, the KCOT is unique among odontogenic cysts^{6,7}. KCOT may manifest as solitary lesions, multiple cysts or as a component of the basal cell nevus syndrome⁸. The most frequently confronted complication with KCOTs is the high frequency of recurrence related to residues of cyst epithelium which follow excision and an intrinsic growth potential which cause cortical expansion and erosion^{6,7}. KCOTs have a high recurrence rate ranging between 25% and 60%⁹.

The radiographic features can be unilocular or multilocular, with a well-defined margin rarely causing root resorption¹⁰ The treatment options include a simple enucleation, which can be held in a single surgical procedure. Marsupialization can also be done which consists of a store to open surgical suturing the wall injury to the epithelium of the oral mucosa. The decompression procedure differs from

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marsupialization by installing a device for progressive reduction in size keratocyst¹¹.

CASE

A 35-year-old male patient had visited the Oral Medicine and Radiology department with a chief complaint of swelling in the palate since 6 months. There was no history of pain or any other associated symptoms. Medical history and history of drug allergy were non contributory. The patient was moderately built and nourished. On extraoral examination, no abnormalities were detected.

On intraoral examination, a well defined swelling 2x2 cm was seen in the left anterior region of the hard palate. (Figure 1) Anteriorly, it was 1cm behind the lingual surface of left maxillary central incisor, distal extent was corresponding to the lingual surface of the left maxillary premolars. It medially involved the median palatine raphe. The posterior extent was 2cm behind the lingual surface of the maxillary left central incisor. On palpation, inspeactory findings were confirmed. The swelling was non tender and cystic in consistency. It was not associated with any discharge. The left maxillary canine was missing. Aspiration of the swelling yielded a yellowish white exudate followed by a blood tinged one. (Figure 2) Based on the clinical findings, a provisional diagnosis of Periapical cyst wrt respect to 21, 22 was made. Keratocystic odontogenic tumor and Adenomatoid odontogenic tumor were also considered as the differential diagnosis.

Panoramic radiograph showed a well defined radiolucent lesion extending from the roots of 21 till the periapical areas of 24 (Figure 3). Crown of 23 was seen projecting into the lesion. Occlusal radiograph showed a radiolucent area which was scalloped and had sclerotic borders. Crown of 23 was seen projecting into the lesion. (Figure 4). Water s view was made, which showed an irregular radiopaque elevation in the floor of the left maxillary sinus suggestive of the root tip of the impacted maxillary canine. (Figure 5). A radiographic differential diagnosis of Keratocystic odontogenic tumor with respect to 23 was made.

The lesion was surgically enucleated along with removal of impacted 23 under local anaesthesia. Histopathological examination of stained H & E sections, 10X showed epithelium and connective tissue. Epithelium was parakeratinized stratified

squamous type which was corrugated with a palisaded polarized basal layer of cells with flat reteridges .The connective tissue showed odontogenic islands, few satellite cysts were also present. Numerous rushton bodies were seen in the areas of inflammation. There were areas showing separation of epithelium from the connective tissue interface, connective tissue stroma is dense and collagenous with fibroblasts, inflammatory cells and blood vessels and was suggestive of Keratocystic odontogenic tumor. (Figure 6).

The patient was recalled for periodic review. A post operative panoramic radiograph was made a year post surgery, which did not show any signs of recurrence.(Figure 7)



Figure 1. Palatal swelling on the left



Figure 2. Yellowish white exudate



Figure 3. Unilocular lesion in the anterior left maxillary region



Figure 4. Occlusal radiograph showed a radiolucent area which was scalloped and had sclerotic borders



Figure 5. Irregular radiopaque elevation in the floor of the left maxillary sinus suggestive of the root tip of the impacted maxillary canine.

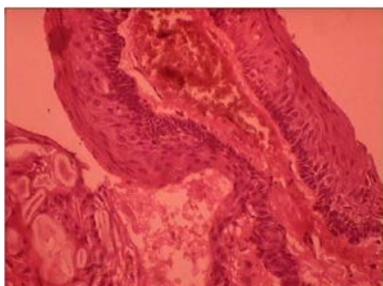


Figure 6. Histopathology

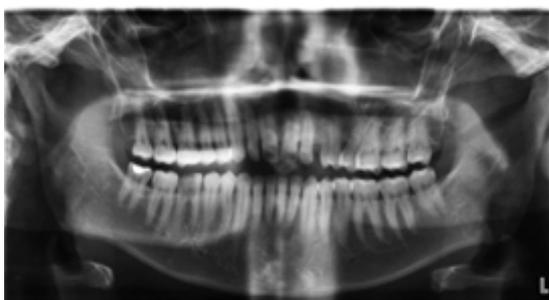


Figure 7. Post operative radiograph showing no signs of recurrence.

DISCUSSION

According to the literature, only 16% of cases KCOT have occurred in the maxilla, of which 13% involve posterior maxilla³. In our case, the lesion was present in the anterior maxilla. A localized swelling, which is asymptomatic is the most common symptom. Spontaneous drainage of the tumor into the oral cavity and teeth mobility are also seen¹².

Radiographic picture of KCOT usually presents a unilocular or a multilocular radiolucency with scalloped and well defined margins¹³. Keratocystic odontogenic tumors, which are large frequently appear similar to other pathologies, such as the dentigerous cyst, lateral periodontal cyst and the ameloblastoma. KCOTs which are smaller, usually present as asymptomatic unilocular radiolucencies with corticated peripheries¹⁴. An unerupted tooth is usually associated with the lesion in 25% to 40% of cases¹⁵. Since KCOT is usually associated with the crown of an involved tooth, it is necessary to radiographically distinguish the lesion from dentigerous cysts¹⁶.

Keratocystic odontogenic tumor is believed to be a developmental cyst which has its origin from the dental lamina or its remnants (glands of Serres)¹⁷. In 2001 Mervyn Shear had observed that epithelium of KCOT, dentigerous and radicular cyst positively reacted for epithelial growth factor receptor (EGFr). The trend indicating that the most intense staining was in the KCOTs, followed by the dentigerous and then the radicular cyst linings. These findings led to the conclusion that the KCOTs have an intrinsic growth potential which is not present in other odontogenic cysts¹⁸.

The surgical modalities which have been suggested for the treatment of KCOTs include enucleation with primary closure, enucleation with open packing and resection with or without loss of jaw continuity. The treatment is dependent upon several factors, such as age, location and size of lesion, and if the lesion is primary or recurrent. Total enucleation with or without “peripheral ostectomy” is treatment of choice for most KCOTs unless lesion is recurrent or has significant soft tissue invasion⁸.

Periodic recall and follow up of the patient is mandatory. This is owing to that fact that the KCOT is an aggressive lesion and that the lesion has a high recurrence rate, despite the unusual location.

The potential regressive behavior is of chief concern and differentiates this tumor from other odontogenic cysts.

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