

Keratocystic odontogenic tumor invading the right maxillary sinus: a case report

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Abstract: Keratocystic odontogenic tumor (KCOT) is a benign intraosseous neoplasm of the jaw. Involvement of the maxillary sinus is an unusual presentation. We present the case of a 23-year-old man with extensive KCOT and impacted third molar in the right maxillary sinus. The clinical, radiological, and histological features of this tumor and its surgical management are discussed. (*J. Oral Sci.* 50, 345-349, 2008)

Keywords: third molar tooth; maxillary sinus; odontogenic keratocyst; keratocystic odontogenic tumor.

Introduction

The odontogenic keratocyst (OKC), first described by Phillipson in 1956, differs from other cysts; it shows more aggressive clinical behavior including a high recurrence rate and demonstrates a high mitotic count and high epithelial turnover rate. Because of these neoplastic features, the term "odontogenic keratocyst" was changed to "keratocystic odontogenic tumor" in the WHO classification of head and neck tumors in 2005 (1).

The keratocystic odontogenic tumor (KCOT) is a benign uni- or multicystic, intraosseous tumor of odontogenic origin (2) that has a slight male predilection and commonly occurs in the second and third decades of life (3).

Jaw cysts and tumors commonly occur due to the presence of odontogenic epithelial remnants (4). Another source of these lesions is basal cells of the overlying oral epithelium in the jaw (5). Remnants of odontogenic epithelium persist in oral tissues after odontogenesis is complete, and a variety of tumors and cysts might possibly arise from these remnants (6). Generally, KCOTs are solitary lesions unless they are associated with nevoid basal cell carcinoma syndrome (7). The most common site for KCOT is the mandibular molar region (3,8-10). While the most common location is the posterior portion of the mandible or the mandibular ramus, unusual locations have also been reported, such as the anterior portion of the maxilla, the maxillary sinus, and the maxillary third molar area (9,11). Radiographically, KCOTs present as well-defined radiolucent lesions with smooth and usually corticated margins. They may be either multilocular or unilocular on radiography. An unerupted tooth is involved in the lesion in 25% to 40% of cases (12).

Histological features are characterized by the presence of a thin bandlike parakeratinized or orthokeratinized stratified squamous epithelium, with a prominent basal layer composed of either columnar or cuboidal cells, and a connective tissue wall that is usually free of inflammation (13). KCOT has a slight male predilection and commonly occurs in the second and third decades of life (7). Therapeutic approaches vary in different studies from marsupialization and enucleation, which may be combined with adjuvant therapy such as cryotherapy or Carnoy's solution, to marginal or radical resection (14). The recurrence rate varies from approximately 20% to 62% (8,11,14,15). Successful treatment depends on precise diagnosis, adequate surgical procedure, and thorough follow-up (11,16).

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Case Report

A 23-year-old man was referred to the Department of Oral Diagnosis and Radiology of the School of Dentistry of Ataturk University after developing pain and swelling of the right maxillary region, accompanied by a discharge into the mouth. The medical history was unremarkable. Plain radiographs of the face and skull including the paranasal sinuses were performed. Water's view showed a dense area indicating the presence of a tooth in the right maxillary sinus (Fig. 1). Subsequently, panoramic radiography revealed a lytic lesion in the right maxillary sinus associated with an impacted third molar (Fig. 2). In order to visualize the lytic lesion in more detail, computed

tomography (CT) was performed. CT revealed that the lesion involved the upper third molar, and that it had destroyed and displaced the roof and the lateral wall of the right maxillary sinus (Fig. 3). Imaging studies demonstrated an oval, well-defined mass including the third molar in the right maxillary sinus region (Figs. 1-3). The lesion was examined at biopsy. Histopathologically, the lesion had a uniform squamous epithelial lining, five to eight cells thick, with fairly flat base. The epithelium demonstrated a well-developed basal layer of palisaded cuboidal and columnar cells with polarized, hyperchromatic nuclei. The squamous cells progressively flattened toward the

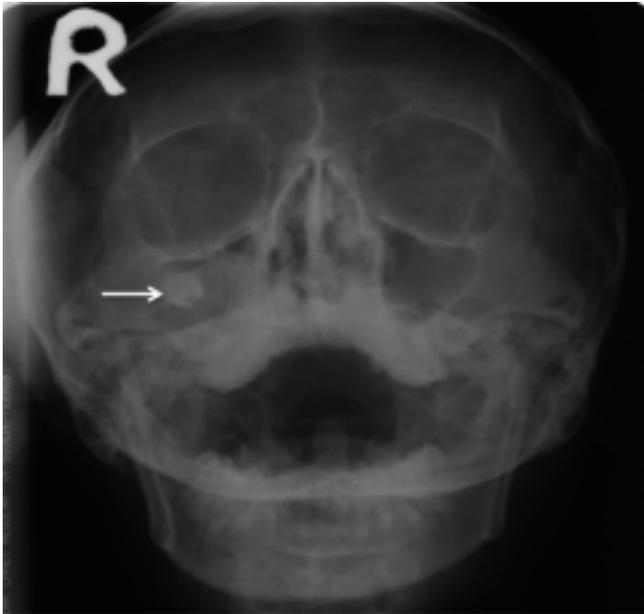


Fig. 1 Water's view revealed an impacted third molar crown in the right maxillary sinus.

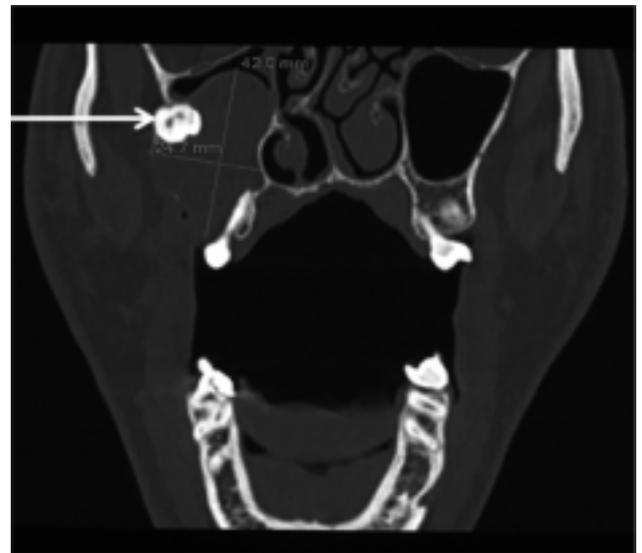


Fig. 3 CT scan of the paranasal sinuses revealed that the lesion involved the upper third molar crown in the right maxillary sinus.



Fig. 2 Panoramic radiography demonstrated an impacted third molar in the right maxillary sinus.

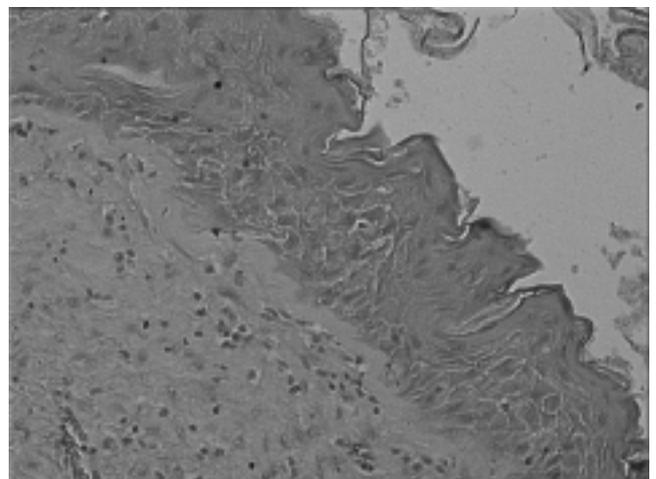


Fig. 4 Biopsy specimen shows a parakeratotic KCOT (H-E staining, ×115).

lumen. The luminal surface was corrugated and lined at least in part by parakeratotic cells. The lumen had an irregular, folded contour and contained keratin (Fig. 4). Under general anesthesia, the lesion was completely excised with a Caldwell-Luc approach (Fig. 5), and the walls of the bone cavity were curetted. Inspection of the cavity showed that the distal maxillary wall and roof had gradually been destroyed and displaced. No recurrence has been determined after six months' post-operative follow up.

Discussion

KCOT has a slight predilection for men and commonly occurs in the second and third decades of life (7,10). However, other studies have reported that peak frequency occurs in the fifth and sixth decades (17,18). In our case, the patient was in his second decade of life. Approximately three quarters of all KCOTs occur in the body of the mandible, most commonly in the molar region and vertical ramus (19). In the present case, we report maxillary sinus involvement as a rare site of occurrence. The literature suggests that less than 1% of all cases of KCOT occur in the maxilla and exhibit sinus involvement (11,20).

Clinically KCOT generally presents with swelling, pain, discharge, aggressive growth, invasion of adjacent structures, and recurrence (7-10,14,21,22). A localized asymptomatic swelling is the most common symptom; spontaneous drainage of the tumor into the oral cavity and mobility of the teeth are also common. Nasal obstruction, paresthesia, and root erosion are more rare symptoms. Some reports emphasize that KCOTs can undergo malignant transformation at a frequency of 5% to 62.5%

(8,11,23-25). It is important to note that KCOTs can be confused with inflammatory lesions since patients with this type of cystic neoplasm usually have inflammatory symptoms such as pain, swelling, and discharge (26). In the presented case, the patient also had intermittent pain and swelling of the right cheek, which can also be seen in inflammatory lesions such as sinusitis.

Radiographically KCOT usually presents as a unilocular or a multilocular radiolucency with scalloped and well-defined margins (7,21). As KCOT is rare in the maxillary sinus, its radiographic appearance in this situation may be misinterpreted. Computed tomography can provide information on the extent of these lesions, contributing to diagnosis and preoperative preparation. KCOT is difficult to diagnose clinically due to a relative lack of specific clinical and radiographic characteristics (27). Since KCOT can be associated with the crown of an involved tooth (7), the lesion must be distinguished from a dentigerous cyst (on radiography). Other cystic and neoplastic diseases, such as traumatic bone cyst, lateral periodontal cyst, central giant cell granuloma, fissural cyst, minimally calcifying odontogenic cyst, radicular cyst, arteriovenous malformation, benign bone tumor, ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma, and plasmacytoma can present with the same radiologic features (28,29,30). Therefore, even if both radiology and clinical features indicate KCOT, a definitive diagnosis cannot be made without histology, and ideally a biopsy specimen examination and accurate clinical, radiographic, and intraoperative observation are essential to determine the most effective treatment in order to avoid recurrence (14,31).

Histologically KCOTs have been classified by some authors into parakeratotic and orthokeratotic subtypes (19,32). These types refer to the histologic characteristics of the lining and the type of keratin produced. Compared with the parakeratotic subtype, the orthokeratotic subtype produces keratin more closely resembling the normal keratin produced by the skin, with a keratohyaline granular layer immediately adjacent to the layers of keratin, which do not contain nuclei. The parakeratotic subtype has more disordered production of keratin; no keratohyaline granules are present, and cells slough into the keratin layer. The keratin contains nuclei and is referred to as parakeratin. The parakeratotic type is the most frequent (80%) and has a more aggressive clinical presentation than the orthokeratotic variant (33-35). Some pathologists think that the orthokeratotic subtype should be classified as a separate entity and termed orthokeratotic odontogenic cyst, because of its distinct histologic features and substantially less aggressive behavior. The lesion reported in this case is a

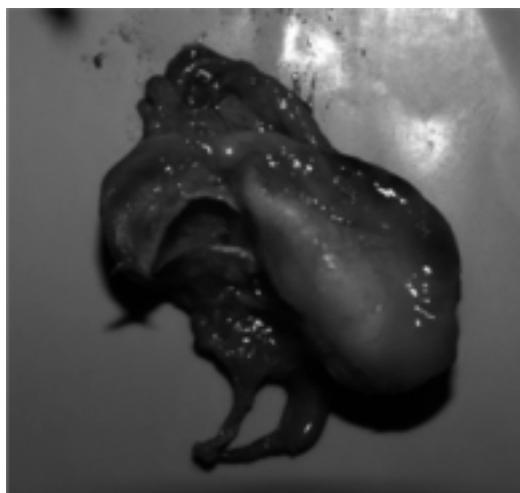


Fig. 5 Excised specimen of the odontogenic keratocyst including upper third molar.

parakeratotic KCOT.

Treatment of KCOT is controversial (10). If these lesions are left untreated, they can become quite large and locally destructive. According to Blanas et al., treatment options for KCOT are simple curettage, enucleation (intact shelling with or without the use of Carnoy's solution or cryotherapy to kill the epithelial remnants or satellite cysts), radical enucleation, marsupialization, and resection (marginal or segmental) (14). Recurrence can occur with all treatment methods except marginal resection (7). In the present study, we used Carnoy's solution in addition to enucleation and aggressive curettage because of the high recurrence rate. The main difference between KCOTs and other jaw cysts is their potentially aggressive behavior. Recurrence is documented even after 10 years of follow up and treatment. However, it is difficult to diagnose recurrence of maxillary sinus cysts after surgical removal of the initial lesion.

In conclusion, KCOTs are relatively rare in the maxillary sinus. It is suggested that if clinical signs and symptoms are absent, the radiological appearance on both conventional and panoramic radiography may be misinterpreted. Therefore CT is important in assessing the full extent of the recurrent lesion preoperatively, and a definitive diagnosis must be made histologically. Post-operative follow up is essential for at least five years following surgery.

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