Case Report:
Keratocystic Odontogenic Tumor Involving Maxillary Antrum

Authors:
Srikanth Gadicherla, Associate Professor,
Abhay T Kamath, Professor and Head,
Vasantha Dhara B, Post Graduate,
Komal Smriti, Assistant Professor,
Department of Oral & Maxillofacial Surgery,
Deepika Pai, Department of Pedodontics and Preventive Dentistry,
Manipal College of Dental Sciences, Manipal, Manipal University, Manipal-567104, Karnataka.

Address for Correspondence
Dr Abhay T Kamath,
Professor and Head,
Department of Oral & Maxillofacial Surgery,
Manipal College of Dental Sciences,
Manipal-567104,
Karnataka, India.
E-mail: abhyataranathk@gmail.com.

Citation

Submitted: Nov 12, 2017; Accepted: Jan 1, 2018; Published: Jan 30, 2018.

Abstract: Keratocystic odontogenic tumor (KCOT) is a benign intraosseous neoplasm of the jaw which is derived from epithelial remnants of the tooth germ and basal cells of the overlying oral epithelium. The occurrence of KCOT in the maxilla is very less compared to the mandible. The incidence of KCOT in maxillary sinus with impacted third molar is very uncommon. This paper reports such a case of KCOT associated with maxillary third molar involving maxillary sinus, diagnosis and treatment of which was based on thorough radiographic evaluation. Very few cases have been reported so far in literature pertaining to the same.

Key Words: Keratocystic odontogenic tumor, maxillary third molar, maxillary sinus, computed tomography

Introduction:
The term odontogenic keratocyst (OKC) was first described by Phillipsen in 1956.[1] In 2005 World Health Organization renamed OKC as Keratocystic odontogenic tumor (KCOT) due to its aggressive and infiltrative behavior. KCOT is a benign intraosseous cyst or tumor of odontogenic in development with an distinctive parakeratinized stratified squamous epithelial lining presenting as unicystic or multicystic lesion which has potential for aggressive, infiltrative behavior. [2] The incidence of KCOT in the maxillary sinus associated with an impacted third molar is less than 1% of all cases. This case report shows one such uncommon occurrence of KCOT related with an unerupted right maxillary third molar, present in the maxillary sinus which could pose difficulty in diagnosis and treatment due to its unusual location.

Case Report
A 21 year old male patient was referred with the chief complaint of swelling in the left side cheek since 8 months. Swelling started spontaneously and enlarged slowly to the present size. He had no contributory medical history.

On extra oral examination there was gross facial asymmetry on left side with diffuse swelling extending from left preauricular region to the angle of mouth antero-posteriorly and from the left zygomatic arch to the angle of mandible supero-inferiorly. On palpation, the swelling was non tender and firm with no local rise of temperature of the overlying skin. Intra oral examination revealed palatal bulge with non-tender palatal expansion with no obliteration of buccal vestibule. The teeth in that region showed no evidence of caries. Associated teeth were vital with no mobility & 28 was missing.

The long duration of the swelling, its asymptomatic nature & absence of odontogenic abnormalities raised the suspicion of a benign bony lesion- Odontogenic cyst/tumor was considered in the clinical differential diagnosis. However due to the nondefinitive clinical history, patient was subjected to thorough radiological evaluation for affirming the diagnosis. A screening panoramic radiograph showed (Figure 1) impacted left maxillary third molar which was displaced towards the antral cavity.

Figure 1: Cropped Panoramic radiograph showing impacted left maxillary third molar within cystic cavity; CT sections showing cystic lining arising from impacted left maxillary third molar with destruction of posterior lateral wall of maxillary sinus
Figure 2: Intraoperative enucleation of cyst with extraction of tooth

To visualize the complete extent of the lesion, Computed Tomography (CT) was performed. The coronal, axial and sagittal sections (Figure 1) revealed a cystic lining arising from an impacted left maxillary third molar inside the antral cavity with destruction of posterolateral wall of antrum. There was complete increased radio opacity with respect to the left maxillary sinus suggestive of an odontogenic pathology. Based upon the clinical and radiological evaluation, a provisional diagnosis of a dentigerous cyst associated with impacted 28 was made. Differential diagnosis of keratocystic odontogenic tumor was considered.

The surgical management of the lesion in the form of enucleation was planned under general anaesthesia. The buccal mucoperiosteal flap was reflected and left maxillary third molar was elevated and removed from the buccal aspect. There was evidence of the perforation of the posterolateral wall of the maxilla. Left maxillary antrum was approached via Caldwell Luc technique and a rectangular piece of bone was removed. The cystic lining was curetted and the cyst was removed in toto and sent for histopathological evaluation (Figure 2). Prophylactically, modified Carnoy’s solution was also used for chemical cauterization to avoid risk of recurrence. The antral cavity was thoroughly irrigated and the rectangular piece of bone was plated using a 1.5mm stainless steel plate.

The enucleated specimen was sent for histopathological examination. The microscopic section revealed a cystic lumen associated with left maxillary third molar lined by a para keratinized stratified epithelium of 4-6 cell layer thickness with a corrugated surface. The basal cells were tall, columnar with a hyperchromatic palisading nucleus and the capular tissue comprised of dense bundles of collagen, fibroblasts, fibrocytes and blood vessels with dense infiltration of chronic inflammatory cells. The above-mentioned findings confirmed the lesion as odontogenic keratocyst. Post-surgical healing was satisfactory with no evidence of recurrence for a period of 6 years.

Discussion

The odontogenic keratocyst is an epithelial developmental cyst of the jaws, frequently derived from the remnants of dental lamina. Due to its aggressive and infiltrative behavior, the WHO has redefined it as Keratocystic Odontogenic tumor. KCOT has been known for high recurrence due to the presence of daughter cysts occurring beyond the margin of the lesion and thin friable cystic lining. [3] The presence of tumor markers which include Ki67, proliferating cell nuclear antigen (PCNA), p53, BCK 2 sequence of the enzyme dithiolopoyl acetyltransferase and matrix metalloproteinase (MMP) 2 within the cyst have also attributed to the same.[4,5] KCOT has a predilection for occurrence in mandible especially in the mandibular posterior region particularly ramus and molar region. In the maxilla, the third molar area followed by canine region are common site of occurrence.[6] The incidence of KCOT in maxillary sinus is rare with frequency of less than 1% of all reported cases in maxilla[7] (Table 1).

Table 1: Clinical data on reported cases on odontogenic keratocyst associated with maxillary sinus

<table>
<thead>
<tr>
<th>Author</th>
<th>Cases</th>
<th>Treatment</th>
<th>Follow up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Houpis et al, 1971</td>
<td>1</td>
<td>Enucleation and Curettage</td>
<td>19 months</td>
</tr>
<tr>
<td>Abhishek Gupta et al, 2011</td>
<td>1</td>
<td>Enucleation &amp; Curettage</td>
<td>12 months</td>
</tr>
<tr>
<td>Marcelo et al, 2014</td>
<td>1</td>
<td>Decompression, followed by enucleation and curettage</td>
<td>3 years</td>
</tr>
<tr>
<td>Absi et al, 1994</td>
<td>2</td>
<td>Enucleation, curettage and cryotherapy</td>
<td>8 months</td>
</tr>
<tr>
<td>Carvalho Silva et al, 2005</td>
<td>2</td>
<td>Enucleation &amp; Curettage</td>
<td>18 - 5 years</td>
</tr>
<tr>
<td>Cioffi et al, 1987</td>
<td>1</td>
<td>Enucleation &amp; Curettage</td>
<td>2 years</td>
</tr>
<tr>
<td>Yadav et al, 2013</td>
<td>1</td>
<td>Enucleation, Curettage, Carnoy’s solution</td>
<td>5 years</td>
</tr>
<tr>
<td>Barry et al, 2016</td>
<td>4</td>
<td>Endoscopic medial maxillectomy, enucleation and curettage</td>
<td>2 years</td>
</tr>
<tr>
<td>Nahvi et al, 2016</td>
<td>1</td>
<td>Enucleation, Curettage</td>
<td>1 year</td>
</tr>
<tr>
<td>Kim et al, 2011</td>
<td>2</td>
<td>Enucleation, Curettage</td>
<td>4 months, 3 months</td>
</tr>
</tbody>
</table>

The causes of the cystic lesions of the maxillary antrum can be odontogenic which include developmental and inflammatory and nonodontogenic like chronic maxillary sinusitis, post-surgery, foreign bodies and trauma. It has been suggested that sinus pathologies can be differentiated from odontogenic cysts through radiological evaluation like computed tomography. Odontogenic cysts are identified by their demarcated expansile cystic mass and are related to the root or crown of a tooth. Moreover, by identification of the bony plate between the cystic mass and the maxillary antrum, an extra-antral cystic lesion can be distinguished from an antral lesion such as a mucocele.[8] In our case the patient did not report any symptoms suggesting a sinus pathology. Thus, the diagnosis mostly relied on radiographic evaluation. A routine panoramic radiograph can show a maxillary third molar in maxillary antrum identifying the odontogenic cause. CT serves to provide extent of the lesion which helps in better delineation and perioperative planning and treatment. Extension of the tumour into the orbital compartment, nasal cavity, zygomma or any palatal erosion can also be identified using computed tomography guiding the surgeon in treatment planning for resection and rehabilitation. However histopathological examination remains the gold standard for diagnosis of KCOT. There are two variants of the KCOT histologically which are parakeratotic and orthokeratotic. It has generally been agreed that tumors with orthokeratinized lining have a lower incidence of recurrence and are less aggressive than the parakeratinized version. The variant of KCOT in the present case is a parakeratotic. There are two modes of surgical management first being conservative approach which includes simple enucleation and marsupialization. The second approach involves aggressive treatment which range from enucleation with peripheral osteotomy and chemical cauterization using Carnoy’s solution and resection.[9] The lesion in the present case was treated by enucleation and curettage along with Carnoy’s solution. The antral bony defect was reinforced using a 1.5mm stainless steel plate. The healing
was satisfactory and follow up was done by serial radiographic evaluation along with clinical examination. The patient was under regular follow up for 6 years with no indication or sign of recurrence.

**Conclusion:**
Pathologies in the maxillary sinus with abstract symptoms require definitive radiographic evaluation. The presence of an aggressive odontogenic tumour like KCOT in the maxillary antrum is rare, its radiographic image in such situation needs to be evaluated for identification of any tooth structure, extent of the lesion, involvement of neighboring anatomical spaces. Computed tomography thus contributes to provisional diagnosis and treatment plan. However OKC has clinical diagnostic difficulties due to relative lack of specific clinical and radiographic characteristics.[10] There are no specific and characteristic clinical and radiographic features of KCOT which aid in the diagnosis. The radiographic similarity of OKC to a dentigerous cyst and clinical resemblance to an ameloblastoma makes it difficult for the surgeon to rely on clinical and radiological examination alone for adequate treatment. The histopathological examination of the biopsy specimen along with accurate clinical, radiographic, intra operative surgical observation are essential to determine the most effective treatment for a KCOT to prevent recurrence at any location.

**References**