Management of keratocystic odontogenic tumor in the maxillary sinus of 12-year-old children: A case report

Hendy H. Suhandi, Yudy A. Utomo,* Eki Nasuri, Vera Julia

Abstract

Objective: KCOTs are characterized by an aggressive behaviour with a relatively high recurrence rate, especially if they are associated with syndromes. Treatment of KCOTs include marsupialization, saucerization.

Methods: A complete history was taken and clinical examination followed by complete enucleation with sulcular flap from 12 region until 18 regions was performed. The diagnosis was maxillary dentigerous cyst based on histopathological result after the operation until 18 regions was performed. The diagnosis was maxillary dentigerous cyst based on histopathological result after the operation.

Results: The medical history was not significant and patient was in a generally good condition with no fever, nausea, pain, or difficulty in breathing. Extra oral examination showed facial asymmetry and visible lump in the right region of the maxilla.

Conclusion: The diagnosis of maxillary KCOT expanding to maxillary sinus can be achieved with adequate data; history, clinical, and histopathological examinations. Careful surgical approach and proper treatment decision is crucial to avoid complications according to individual case of each patient, especially in children.

Keywords: KCOT, KCOT in children, Treatment of KCOT
DOI: 10.15562/jdmfs.v8i1.1145

Introduction

Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst (OKC) is a benign intraosseous tumor of odontogenic origin, making up the third most common odontogenic cyst. KCOT reflecting infiltrative, aggressiveness and potential for recurrence. This cyst is non-inflammatory and it can be unicystic or multicystic, with a characteristic lining of parakeratinized stratified squamous epithelium and has a potential to become aggressive and infiltrative. KCOT’s arise from the remnants of dental lamina or the basal cells of the overlying oral epithelium. The recurrence of KCOT associated with age, location, size, shape and the surgical technique. The most common sites of occurrence are body of mandible molar region and ascending ramus. KCOTs rarely presents itself in anterior portion of the maxilla, the maxillary third molar area, the maxillary antrum and the canine region is the most common location. KCOTs can occur in any age, with the peak incidence in the third decade of life and a slight male predominance.

Clinically, KCOT is usually asymptomatic in the initial stage such as mild swelling, displacement of teeth, pain and pus discharge can be seen likely due to secondary infection, also invasion of anatomic structures may present themselves in the later stages. KCOTs in maxilla tend more to be secondarily infected than mandibular ones, due to its vicinity to maxillary sinus. KCOTs are characterized by an aggressive characteristic with a relatively high recurrence rate, especially if they are associated with syndromes. Radiographically, KCOTs appear as a well-defined unilocular or multilocular radiolucency bounded by corticated margins which appears like a well-defined peripheral rim, with scalloped border, usually associated with root resorption and displacement of teeth with involvement of impacted tooth. Most KCOTs are unicystic, whereas multicystic variant occurs in approximately 30% of cases, most commonly in the mandible. Histologically, KCOTs are constituted by a cystic space containing desquamated keratin, lined with a uniform parakeratinized squamous epithelium of 5 to 10 cell layers, with cuboidal cells, whose nuclei tend to be vertically oriented. The major variants that have been recognized by many researchers through histological features of the KCOT based on microscopic findings: A cyst with parakeratinized epithelial lining and a cyst with an orthokeratinized epithelial lining.

Treatment of KCOTs include marsupialization, saucerization, Caldwell-Luc technique, Carnoy’s solution, marsupialization and enucleation, depending on the patient’s age, location and size of lesion. The most effective treatment still remains controversial.
Case Report

A 12-year-old child patient was referred to the Specialist Clinic of Oral and Maxillofacial Surgery, Cipto Mangkusumo Hospital with a chief complaint of facial asymmetry caused by painless lump in the right of his maxillary. On physical examination, the patient was apparently healthy. The medical history was not significant and patient was in a generally good condition with no fever, nausea, pain, or difficulty in breathing. Extra oral examination showed facial asymmetry and visible lump in the right region of the maxilla figure 1. The fixed lump with a size of 5x4x3 cm was palpable with tenderness. The color and the temperature of the lump was similar to its surrounding and the skin was intact. The patient mouth opening was 4.4 cm wide.

Upon intraoral examination, the patient’s oral hygiene was fair. A fixed lump in the buccal region of the right maxilla was visible from region 17 to region 13 figure 2. The lump has a soft consistency with the same temperature and color as its surrounding and is palpable with tenderness. The skin was also intact.

From the complete blood count result, it was found that the patient had an abnormal leukocytes count (9810 cell/mm), creatinine (0.70 mg/dL), and blood sugar count (106 mg/dL). No abnormalities were found in the thorax radiograph. CT scan of face conducted in RSCM without contrast showed an image of bilateral maxillary dentigerous cyst with extended defect to the lateral wall of right maxillary sinus figure 3. The CT scan also showed signs of right maxillary sinusitis with cyst retention in the left maxillary sinus figure 4. From patient’s chief complaint, history of illness, extra oral and intra oral examination, radiological, and histopathological examination, a diagnosis of maxillary Odontogenic keratocyst was made. Surgical enucleation of the cyst using sulcular flap from 12 region until 18 regions to create an opening at the anterior wall of sinus maxillary surgically was chosen as the treatment of choice figure 5A, followed by teeth extraction in the 17, 16, 15, 14, and 13 regions figure 5B. The surgery was performed under general anesthesia. The patient was prescribed with antibiotic for prophylaxis (cefotaxim 1g intravenous, half an hour before procedure) and Omeprazole 2caps 20mg to reduce ulcer stress. The patient was instructed to go on fasting for 6-hour period before the procedure and to maintain good oral hygiene. During the procedure, the cystic sac was identified and dissected from the walls and mucosa sinus of maxilla. The wound was closed and the specimen was sent for histopathologic examination to confirm the diagnosis of odontogenic keratocyst.

Discussion

Keratocystic odontogenic tumor (KCOT) can occur in any age with the peak occurrence in the third
A CASE REPORT

Figure 4. CT scan and MRI confirmed the infiltration of maxillary dentigerous cyst to the maxillary sinus. CT scan of face conducted in RSCM without contrast showed an image of bilateral maxillary dentigerous cyst with extended defect to the lateral wall of right maxillary sinus. The CT scan also showed signs of right maxillary sinusitis with cyst retention in the left maxillary sinus.

Figure 5. A. Surgical enucleation of the cyst using sulcular flap from 12 region until 18 regions to create an opening at the anterior wall of sinus maxillary surgically was chosen as the treatment of choice, B. Extraction of teeth in the 17, 16, 15 regions. The cystic sac was identified and dissected from the walls of maxilla. The wound was closed and the specimen was sent for histopathologic examination to confirm the diagnosis of dentigerous cyst.

decade of life. Its frequency in children is relatively low. In the present case, the cyst was associated with the unerupted teeth of 12-year-old children. The prevalence of mandibular KCOT is more common than maxillary KCOT, and KCOT is rarely associated with an unerupted tooth.

Since there are two major variants of KCOT: parakeratinized and orthokeratinized, the comparison between them based on the occurrence, parakeratinized more commonly than orthokeratinized. Also based on the recurrence, parakera-

tinized (42%) has a higher recurrence rate compared to orthokeratinized (2.2%). Residual epithelial cells with stimulation are common features in development of any cyst. In case of KCOT, the epithelial cells implicated are from the rest of Serres or Malassez or from the reduced enamel epithelium. Epithelial cells proliferate into enlarging mass gradually. During the enlargement, the cells in the core mass are driven farther from the blood supply and eventually die creating the lumen. The lumen later becomes hypertonic, creating an osmotic gradient and resulting in an increased hydrostatic pressure that causes bone resorption and subsequent expansion of the cyst. Residual epithelial cells are sloughed into the lumen as the cyst increases in size and the cycle reoccur. The adjacent connective tissue is compressed with the continuations of enlargement. Maturation of the lining then forms a basement membrane and also contributes to the enlargement of the cyst. If no treatment is conducted during the initial stages, the enlargement will continue and eventually become symptomatic.

Diagnosis of KCOT can be made by careful clinical, radiological, and histological investigations. Radiological imaging such as computed tomography (CT) and magnetic resonance imaging (MRI) plays an important role in the diagnosis and management of KCOTs.

In clinical examination for KCOT there are some regions that need to be confirmed include maxillofacial region, facial, intraoral, neck and thorax-abdomen-extremity. In histological investigation can be conducted through biopsy either an incisional or excisional biopsy depending on the size of lesion. Before incising the lesion, aspiration is important to rule out a vascular lesion. If the aspiration of bright red blood alerts the surgeon to a high flow vascular lesion which could result in uncontrollable hemorrhage. If the aspiration of clear fluid is the characteristic of cystic lesion, while the absence of any aspirate may be seen with a solid mass (tumor). A smaller cystic lesion can be completely excised while for the larger lesion need an incisional biopsy to guide the final therapy.

CT scans of the mandible and neck are recommended when a large lesion is found. CT scans are valuable because they provide more information about the proximity of the surrounding structures (e.g., mandibular canal), integrity of cortical plates, and presence of perforation into the adjacent soft tissue. The accuracy of CT scans helps the surgeon to assess the size of lesions and demonstrate the additional anatomic details (or lesions) that are not covered by panoramic radiographs. Evaluation of osseous structures mostly informed using non-

contrast-enhanced CT scans. Through MRI, three-dimensional volume-reconstructed scans can be obtained. This imaging reconstructs the bony framework of the facial skeleton and can be helpful when large intraosseous pathology is found. KCOTs have been demonstrated that T2-weighted magnetic resonance imaging (MRI) can detect KCOT with recognized pattern.

KCOTs are often asymptomatic because they often grow along the anterior-posterior direction within the jaws, with little evidence of cortical expansion in the initial stages. KCOTs occasionally cause swelling, pain, discharge, teeth mobility, and invasion of adjacent structures. If KCOTs involve the maxillary sinus, they can easily expand to an enormous size due to the less dense structure of the sinus, rarely causing nasal obstruction, root erosion, and paresthesia.

Diagnosis of such lesions is challenging as maxillary radiographs do not offer characteristic features due to the overlapping of various structures. Plain radiographic images in such situations may be interpreted, and CT can provide information on the extent of these lesions, contributing to diagnosis and preoperative preparation. Differential diagnosis for KCOT includes dentigerous cysts, lateral periodontal cysts, radicular cysts, ameloblastomas, adenomatoid odontogenic tumors, odontogenic myxomas, simple bone cysts, central giant cell granulomas, arteriovenous malformations, and a number of fibro-osseous lesions.

Surgical excision and pathologic analysis of the lesion is essential for the definitive diagnosis. In the maxillary sinus, the use of carnoy’s solution and resection are not recommended. The standard treatment for KCOT involves surgical enucleation and extraction of the cyst-associated impacted or unerupted tooth, marsupialization, the Caldwell-Luc operation, and endoscopic sinus surgery. After diagnosis was made, treatment of dentigerous cyst can be performed surgically, either by enucleation or marsupialisation. Following enucleation of the cyst and extraction of the teeth involved, long term follow-up must be done to detect any recurrence associated with the lesion when it occurs in the maxillary sinus. The decision whether to enucleate or marsupialise the cyst depends on careful consideration of various patient factors. Enucleation will alter the normal tooth development.

The KCOT as explained before has clinical features that include potentially aggressive and high recurrence rate. Because the recurrence is a major concern, the surgeon varies in their surgical approach or techniques. The basic mechanism for recurrence has been postulated to be incomplete removal of cystic wall or new primary cyst formation from additional activated rests. KCOT have been reported to undergo transformation into ameloblastoma and squamous cell carcinoma although this occurrence is rare. Other postsurgical complications include inferior alveolar nerve paresthesia, postoperative infection, and pathological mandibular fracture in larger lesion (2 weeks after enucleation).

**Conclusion**

Although KCOTs are rare in the first decade of life, they can form in young children. For this reason, the proper treatment choice with the early clinical diagnosis of these lesions may increase the rate of success. A long-term follow-up is important for the diagnosis of the recurrence as well.

**Acknowledgment**

I express my gratitude to Yudy A. Utomo, DDS., OMFS, Vera Julia, DDS., OMFS., PhD and Lilies D. Sulistyani, DDS., OMFS., PhD for their invaluable assistance and advice in this study.

**Conflict of Interest**

The authors report no conflict of interest.

**References**