

Maxillary Sinus Odontogenic Keratocyst: Role of Radiology and Surgical Approach A Rare Case

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ABSTRACT

Odontogenic keratocyst (OKC) is a benign cystic lesion of odontogenic origin with aggressive and infiltrative behavior. Maxillary sinus involvement is reported rare in OKC with <1% cases and could mimic symptoms of other Sino nasal neoplasm. Recurrence rate was high between 25-60% that attributed to surgical technique. We present a case of a 14-year-old female presented with swelling at lateral side of left nose extended to the left cheek for 3 months. It was tender on palpation without pain nor hyperemic. There was no discharge from both nasal and oral cavity. Computerized tomography (CT) scan showed cystic mass with destruction of maxillary wall suspected as mucocele. Sub labial surgical approach was performed. Large cystic mass seen occupied left maxillary sinus with destruction of its anteroinferior wall. Displacement of several teeth roots was seen with canine root was attached in the lesion. Whole mass was completely enucleated without affected external appearance. Patient has been asymptomatic for 3 months follow up. Patient was referred to the dentist for teeth prosthetic treatment, however patient was felt satisfied with the cosmetic and functional result. Diagnosis of maxillary OKC is challenging. CT scan and/or Magnetic Resonance Imaging (MRI) were important to differentiate the cyst with other sin nasal neoplasms. Panoramic radiography can give important information related teeth involvement. Site and size of lesion, aggressive nature and high risk of recurrence in OKC contributed in choosing the best surgical technique. Surgical approach chosen should ensure the wide surgical field for complete resection while preserving cosmetic appearance.

Keywords: *Odontogenic Keratocyst, Maxillary Odontogenic Keratocyst, Panoramic Radiography, Sublabial Approach, Surgical Resection.*

1. INTRODUCTION

Odontogenic keratocyst (OKC) defined as a benign cystic lesion of odontogenic origin with aggressive and infiltrative behavior. It represents around 10% of odontogenic cyst and around 80% of cases found in the posterior part of mandible. These characteristics believe to be caused by mitotic activity and epithelium growth potential. OKC found slightly more common in males than females and around 60% patients diagnosed in the age of 10 until 40 years [1-4].

Less than 1% case of OKCs involved maxillary sinus. It often misdiagnosed and confused with other cysts and neoplasm due to nonspecific clinical and radiological finding. Radiologic examination commonly used are panoramic radiography, CT scan and MRI. Pre-operative

examination plays important role for treatment plan mainly related to the surgical approach to be used [1,4,5].

Treatment option for OKCs divided as conservative and aggressive. Enucleation, curettage and marsupialization are categorized as conservative therapy. Some authors included peripheral osteotomy and combination with Carnoy's solution as complementary treatments. Aggressive therapy defines as resection with or without loss of jaw continuity.^{6,7} Recurrence rate was reported varied up to 62% and can occur within 2 years after surgery. It is affected by size, site and surgical approach performed for treatment [1,2].

OKCs case was rarely found in our center, and commonly misdiagnosed as another entity. Thus, the aim of this paper is to present the OKC case in the maxillary

sinus, the importance of radiology examination and discussion about best surgical approach to perform.

2. CASE REPORT

A 14-year-old girl presented with swelling at lateral side of left nose extended to the left cheek for 3 months. The swelling area was tender on palpation without pain nor hyperemic. It collapsed the left nasal cavity. Intra oral examination revealed the swelling expanded lower until the left maxillary vestibule from left incisor until premolar area. Patient also complaint of left nasal obstruction without headache nor discharge from both nasal and oral cavity. The lymph nodes were not palpable on both sides. No disturbance of eye sight. Past medical history was unremarkable.



Figure 1 Swelling at the left cheek area

CT scan revealed a radiolucent lesion at left maxillary sinus with destruction of mostly inferior maxillary wall and medial displacement of left nasal cavity that pushed

the nasal septum to the right and narrowing the right nasal cavity. This lesion then diagnosed as mucocele of the left maxillary sinus.

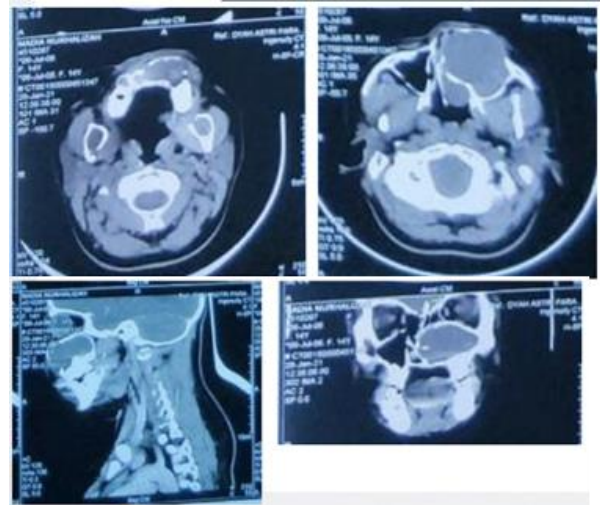


Figure 2 CT Scan showed radiolucent lesion and extension of the lesion

Sub labial surgical approach was performed (Figure 3). Incision was made 0.5 cm above the mucogingival junction. Large cystic mass seen occupied left maxillary sinus with very thin appearance of anterior wall and destruction of inferior wall. Displacement of left incisor tooth roots was seen with canine root was attached in the lesion. Whole mass was completely enucleate as one piece with affected canine and continued with curettage to clear the attached area. The left incisor and 1st premolar teeth were also extracted

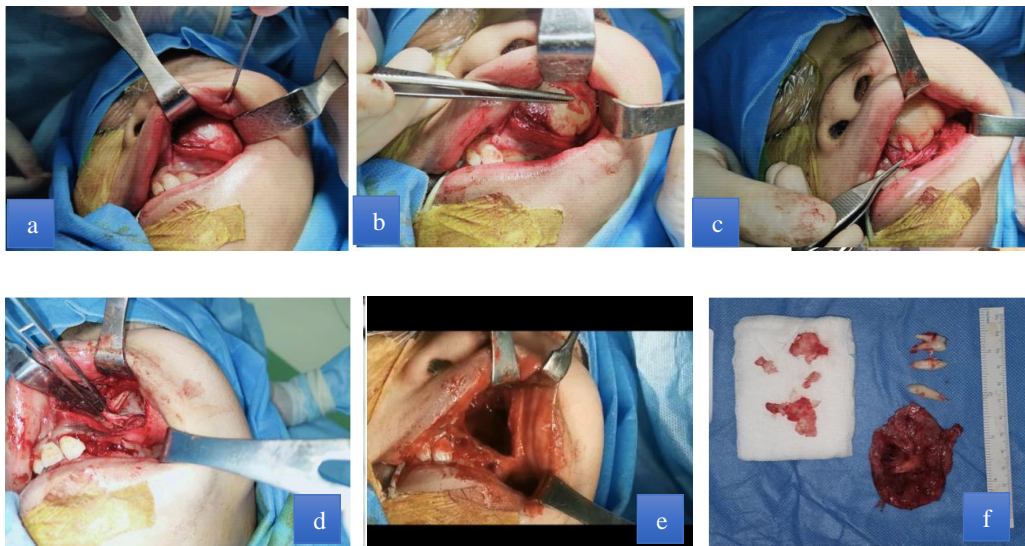


Figure 3 Cystic mass seen in the left maxillary sinus (b) very thin anterior wall of maxillary sinus (c) Canine root seen attached in the mass (d) Extraction of involved tooth and extraction of the mass (e) The maxillary sinus after curettage (f) extracted mass, parts of maxillary sinus anterior wall and teeth.

After surgery, Patient was consulted to the dentist for prosthetic denture treatment. Patient didn't find difficulties in eating nor talking and felt satisfied with the cosmetic result. Patient has been asymptomatic for 3 months after the operation (Figure 4).



Figure 4 Post- surgery follows up (a) 2 days (b) 2 weeks (c) 3 months

3. DISCUSSION

Around 60% of OKCs found between 10 until 40 years old and the incidence is slightly higher in male, meanwhile this case reported OKCs in 14-year-old female. Involvement of maxillary sinus as reported in this case is found rare in this entity. Some authors suggested the entrapment of odontogenic epithelium within the sinus could presumably give rise to OKCs. Close proximity between dental lamina, primordium of canine to the floor of sinus also reported in several studies. These anatomy conditions can contribute to 11-12% of maxillary sinusitis case and lead to maxillary sinus pathology. Approximately 30% of OKCs associated with at least one unerupted tooth and most commonly the third molar. Yet, common affected site in maxilla is between canine and lateral incisor with or without alveolar bone and unerupted teeth association. It rarely associated with impacted molar area. This accordance to our case with involvement of lateral incisor and canine area and not related with unerupted teeth [1,2,4,5,8].

In many cases, patients are asymptomatic until the cyst reached a large size, involve nearby structures and may present with swelling, pain or discharge [9]. These symptoms are relative similar to other sinonasal neoplasm. Diagnosis is also challenging as maxillary imaging do not offer characteristic features due to overlapping of various structures. Pre-operative assessment is very important to determine the prompt treatment and surgical approach to perform. Involvement of maxillary sinus must be carefully assessed because the risk of orbital damage and infection spreading that could lead to the local and/or systemic complication. Lesion can easily expand bigger in the maxillary sinus due to the less dense structure [8]. This could lead to underdiagnosis and improper treatment and resulting high chance of recurrences [2,4].

Imaging is important in located the lesion, evaluating the extent and effect to adjacent structures. Panoramic

radiography, CT scan and MRI are most commonly used in OKCs [1]. OKCs appears as unilocular or multilocular well-defined radiolucency [5,10]. Some authors believe that CT is superior since it able to display the important features such as size, shape, margins, bony changes, and extension of the lesion [1,2]. Panoramic radiography represents the curved surfaces of dental arches, involvement of alveolar bone and adjacent structures, tooth displacement and root resorption [1]. MRI is useful to provide better demonstration of the internal features and soft tissue involvement and useful in discriminating between OKCs and ameloblastoma [1]. Study by Cardoso LB et al [11] showed that CT has lower sensitivity and specificity was slightly higher compared to the panoramic radiograph. There was a significant decrease in the accuracy of the diagnosis with the use of CT compared to panoramic radiograph [11]. MRI was reported superior in assessment of recurrent lesions which might not always be outlined by bony tissues. But when the aim is to make a differential diagnosis, combination of panoramic and CT increases the diagnostic ability [12]. Imaging examination in this case was proceed with CT scan only and lead to pre-operative underdiagnoses. Some study performed aspiration of cyst for diagnosis assessment. Squamous and abundant eosinophilic material commonly found which was suggestive to keratocyst [5].

The management of OKCs aims to reduce the chance of recurrences due to its aggressive behavior while minimalizing the morbidities. Several factors believed to be considered to reach the successful treatment. Factors form lesion features such as; size, location, uni- or multilocularity, soft tissue involvement, cortical perforation, patients' characteristics such as patients age, and also adequate surgical procedure and follow up [1,2,6]. Conservative treatment suggested by several authors as the main treatment. Enucleation and curettage can provide a complete specimen for histopathologic analysis [6]. Karaca et al [13] showed that enucleation with peripheral osteotomy resulted minimal morbidity and could be preferred over aggressive treatment [13]. One-piece enucleation and curettage was performed to this case with sublabial approach. The ideal treatment for OKC would be enucleation or curettage. If the tooth located in the maxillary ostium endoscopic approach can be chosen [10]. When the lesion occurs in the maxillary sinus, Carnoy's solution is not recommended since it can penetrate the bone to a depth of 1.54 mm. Sublabial incision approach can provide direct view to the sinus ensuring the clear enucleation and curettage. One-piece enucleation with eradication of whole epithelium will give lower recurrence compared to several pieces [8].

Lowest recurrence rate can be achieved by resection. It also can be chosen in large mass with size 45 cm³, multilocularity and affected adjacent soft tissue. It also recommended for recurrent lesion in difficult anatomic location. However, resection related with morbidities

such as facial asymmetry and the loss of jaw continuity. Thus, this aggressive treatment is not recommended as the primary treatment in most of the cases [1, 13, 14]. Systematic review by Johnson et al showed that enucleation is associated with the highest recurrence rate followed by marsupialization mostly if the lesion located in mandibular angle due to difficult access [1,6]. Recurrences can happen due to incomplete removal gives rise to new cyst formation (microcyst/daughter cyst), development of new keratocyst from epithelial off-shoots of the basal layer, and continuous cyst formation in patients with basal cell nevus syndrome. Some authors reported that recurrence not only related with types of treatment but also influenced by biological nature of OKCs. It has a very thin and friable cyst wall, expression of proliferative markers such as Ki-67 and great mitotic activity and growth potential found in epithelium, remnant dental lamina and epithelial islands [4,15,16].

OKC related to ectopic tooth reported as 8 cases [10]. There was no statistically different between the recurrence rate and the treatment protocol of the teeth. Canine tooth was enucleated in this case along with the cyst and other involved teeth around the cyst were also extracted. Satellite cyst possibly can rise in the left alone teeth. Recurrent can also happen in the endodontically treated teeth. The preservation of tooth involved in the lesion has been reported to increase the risk of recurrence and extraction of the affected tooth in case of root involvement has been suggested [13].

Recurrences can occur within the first 2 years after treatment and some authors have been reported more than 10 years. For this reason, periodic radiographic monitoring and long term follow up is very essential of the treatment plan. Functional and cosmetic appearance should also be evaluated for the better quality of life of the patient [1]. We plan to perform panoramic radiography for six-month evaluation in this case and will proceed to MRI if there is a sign of recurrences.

4. CONCLUSION

Diagnosis of maxillary sinus OKC is challenging. CT scan and/or Magnetic Resonance Imaging (MRI) were important to differentiate the cyst with other Sino nasal neoplasms. Panoramic radiography can give important information related teeth involvement. Site and size of lesion, aggressive nature and high risk of recurrence in OKCs contributed in choosing the best surgical technique. Surgical approach chosen should ensure the wide surgical field for complete resection while preserving cosmetic appearance.

CONSENT FOR PUBLICATION OF THE IMAGE

Author declare that the patient or guardian agreement to published her images on this journal.

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