Dentigerous Cyst Over Maxillary Sinus: A Case Report and Literature Review

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Abstract

Dentigerous cyst (DC) is a common odontogenic cyst developed abnormally around unerupted maxillary or mandibular teeth. It is often asymptomatic and can be found incidentally on dental radiography with delayed eruption of teeth. However, it can be large and cause symptoms related to expansion and impingement on contiguous structures. Pain and swelling may be the major complains of patients. However, DC seldom caused head and neck inflammation or infection. Here, we described a 20-year-old male, who was found of a DC arising from right maxillary third molar involved the maxillary sinus and with sinusitis. We also reviewed articles to discuss the differential diagnosis of DC from other odontogenic cysts or odontogenic tumors.

Key words: dentigerous cyst, sinusitis, maxilla.

Introduction

Dentigerous cyst (DC) is a common oral lesion formed by fluid accumulation between the fully formed tooth crown and the reduced enamel epithelium. It is considered a developmental abnormality arising from the reduced enamel epithelium around the crown of an unerupted tooth. The predilection site of DC is the mandibular third molar. Other frequent sites include maxillary canines, maxillary third molars, and mandibular second premolar. It is always associated with any unerupted teeth, usually attached to the tooth at the cemento–enamel junction. But rarely involves unerupted deciduous teeth1.

Radiographically, the DC typically shows a unilocular radiolucent shadow with a well-defined sclerotic border associated with the crown of an unerupted tooth, but an infected cyst will show ill-defined borders2. Here, we will describe a case of DC arising from right maxillary third molar and involving the maxillary sinus with sinusitis.
**Case report**

A 20-year-old young male soldier was referred to the Division of Oral and Maxillofacial Surgery, Kaohsiung Armed Force General Hospital with a history of painful sensation and swelling over right maxilla. He was also suffered with yellow-green pus discharges from nasal cavity. On physical examination, pus discharged from right maxillary second molar distal gingival crevice area was noted. The right maxillary third molar cannot be seen. The patient had recurrent sinusitis for six years with medical treatment but in vain. He had no systemic disease or drug and food allergy history. Family history did not show any contribution.

The panoramic radiography showed a well-defined radiolucent lesion with sclerotic margin. The lesion pushed the impacted maxillary third molar upward to the roof of the maxillary sinus. The right maxillary sinus was not clear in the radiography. Root resorption of upper right second molar was also noted (Fig. 1). Computed Tomography revealed an expansive mass in the right maxillary sinus associated with the radioactive intensity similar to soft tissue. There is no bone destruction (Fig. 2).

The patient was arranged to receive operation under general anesthesia. Cyst enucleation was done with Caldwell-Luc’s procedure. Full thickness mucoperiosteal flap was reflected at the right mucobuccal fold from right fist premolar to second molar. A bony window was made to approach the maxillary sinus (Fig. 3). The cyst was totally removed and the surrounding bony structures about 1 to 2 mm thickness were also shaved by Stryker. The lesion was about $4 \times 3 \times 2$ cm in size with firm in consistency. The impacted tooth was also removed. Delayed closure of the wound was done. The opening was packed with sterilized petrolatum gauze. Oozy discharges were found on the following 2 days. On the third day, the mucoperiostum was sutured with 4-0 silk. Postoperative antibiotics were prescribed for 7 days.

Microscopically, the epithelium was composed of non-keratinizing stratified squamous epithelium with plenty chronic inflammatory cells infiltration (Fig 4, 5). There is no evidence of malignant change or other odontogenic cysts differentiation. A dentigerous cyst was confirmed.

One month latter, the patient was free from sinusitis after receiving cyst enucleation. No cyst recurrence was noted after an 18-months follow up.

**Discussion**

The DC is the second most common odontogenic cyst, with periapical cyst being found more commonly. It presents mostly in the second or third decade of life in the maxillary or mandibular third molar or maxillary canine regions. It can originate from any tooth, including supernumerary tooth. The DCs are mostly asymptomatic and may be found on routine dental radiographic check-up. They may also cause symptoms like pain or swelling with the enlargement of the cyst size. Several researchers reported the pathologic fracture of the mandible caused by the huge of DC. The outgrowth of the cyst may also cause the resorption of adjacent tooth. According to Eliasson’s report, roughly 1% of impacted maxillary third molars will subsequently become involved with a DC.

Radiographically, the typical DC showed a well-defined radiolucency with sclerotic border associated with the crown of an unerupted
Fig. 1. Unclear sinus image with distal root resorption of upper right 2nd molar were seen. The impacted tooth was pushed to the roof of maxillary sinus by the cyst. The cyst extended from upper right premolar to retromolar area and maxillary sinus roof.

Fig. 2. A cystic lesion associated with an impacted teeth was seen at right maxillary sinus. There was no bony destruction at sinus wall.
Fig. 3. The lesion was removed.

Fig. 4. H&E staining showed non-keratinizing stratified squamous epithelium lining with plenty of chronic inflammatory cell infiltration (X100).
Fig. 5. Same picture from Fig.4 H&E staining showed plenty of inflammatory cells infiltration with hemorrhage (X200).

tooth\textsuperscript{1}. Three varieties of the cyst-to-crown relationships can be seen on radiographic examination. They are central variety, lateral variety and circumferential variety\textsuperscript{1}. In the case presented here, the cyst-to-crown relationship was classified to a circumferential variety. The expansion of the cyst caused the resorption of the distal root over upper right second molar. The cyst growth in this case was quite extensive. The tumor extended from the mesial aspect of upper first and second premolar, to the retromolar area, and superiorly to the roof of the maxillary sinus. This large cyst made the patient had symptoms such as pain, swelling and sinusitis. In the mandible, the DC may grow in to the ramus and caused mandible expansion\textsuperscript{7}. In our case, antral obliteration was seen from computed tomography. Generally speaking, a panoramic radiology was sufficient for evaluation. The computed tomography may be useful for evaluating the extent of bony involvement. Since DCs may contain fluid, in the magnetic resonance imaging (MRI), the cyst fluid may be seen as low intensity on T\textsubscript{1}-weighted and high intensity on T\textsubscript{2}-weighted images\textsuperscript{8}.

Patients with DCs over maxillary sinus might present nasal symptoms such as sinusitis. In addition, ophthalmologic symptoms might be present such as proptosis, diplopia, ptosis, epiphora but rarely affected visual acuity. Fractured of the orbital bone caused by DC have been reported\textsuperscript{9}. Spontaneous remission of the lesion without surgical removal may happen, but cases are few\textsuperscript{10}.

The differential diagnosis of DC includes odontogenic keratocyst (OKC), adenomatoid
odontogenic tumor (AOT), calcifying epithelial odontogenic cyst (COC), calcifying epithelial odontogenic tumor (CEOT), and unicystic ameloblastoma (UAs). In addition to the histopathologic differences between the feature of the epithelium of OKC and DC, the differential diagnosis can also include the development and the recurrence tendency of these cysts. About 40% unilocular OKC contain impacted tooth. The OKC is more aggressive with higher recurrence risk than DC and may be associated with nevoid basal cell carcinoma syndrome. Recently, researches showed mutation of PTCH gene and overactivated of Shh signaling may be associated with the clinicopathological expression of OKCs. BMP-4 may be a useful biochemical marker to differentiation of OKC and DC. BMP-4 is expressed more intensive in OKC compared with DC, and is more intensively expressed in the recurred cases. The AOT and COC generally are more frequently seen in maxillary anterior area with some degree of calcification within the cyst cavity, which may be observed from radiography. Histopathologically, the COC may present keratinized epithelial cell so-called ghost cells in the cavity. The AOT different from the DC, its predilection in female with epithelial cells syncytially arranged in rosettes or duct-like structure. The CEOT may be differentiated from DC by its honeycomb pattern radioluency with foci of radiopacity in radiographs. Microscopically, it shows large polygonal epithelial cells with variation in size and shape with amyloid materials, which contained concentric calcified deposits (Liesegan rings). Although the unicystic ameloblastoma is rare, its clinical expression and microscopic feature can be deceptive and may be confused with DC. The UAs is more locally aggressive than DC with several immunohistochemical markers enhanced, such as Bcl-2, Bcl-xL, FGF, MMPs, Ki-67 and PCNA. Recently, calretinin was suggested to be a specific immunohistochemical biomarker for neoplastic ameloblastic epithelium and may serve as a diagnostic tool for differentiating cystic odontogenic lesions from ameloblastoma.

The best treatment of the UAs is radical though enucleation is sufficient in subtype 1 and 2. Considering the similarity of UAs and DC clinically, we enucleated the lesion and also trimmed the surrounding bony structures to about 1 to 2 mm in thickness. Hopefully, the histopathological report confirmed the diagnosis of DC.

The DC may cause head and neck infection with a prevalence of 2.1%. The cyst can undergo carcinomatous transformation into ameloblastoma or squamous cell carcinoma but is rare. In cases where mucous cells are present in the epithelium, the intraosseous mucoepidermoid carcinoma may be ruled out. Enucleation of the cyst contents with extraction of the associated tooth is sufficient for DC. For extremely large lesions, or in cases when the involved tooth is desired to keep in the arch, marsupialization may be done. With decompression, the involved tooth may erupt spontaneously by orthodontically into occlusion. When other odontogenic tumors are highly suspected, radical removal of the lesions or removal of the cyst with surrounding bony structures is suggested.

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上顎竇之含牙囊腫：病例報告與文獻回顧

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摘 要

含牙囊腫是一種常見於上下顎阻生齒周圍發育異常的齒源性囊腫。由於通常並無症狀，故患者常因牙齒延遲萌發，於接受放射檢查時而發現。然而，此囊腫若持續擴大，侵犯擠壓到鄰近組織，便可能產生不適。腫大與疼痛是大多數病人的主要抱怨的症狀，但含牙囊腫鮮少引起頭頸部發炎或感染。本文提出一位20歲男性因右上顎第三大臼齒侵上顎竇導致鼻竇炎的個案報告，同時藉由回顧文獻，討論對於含牙囊腫與其他齒源性囊腫的鑑別診斷。

關鍵詞：含牙囊腫，鼻竇炎，上顎。