

Odontogenic Keratocyst Associated with an Ectopic Tooth in the Maxillary Sinus – A Report of Two Cases and a Review of the Literature –

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Received: March 26, 2010

Accepted: April 13, 2010

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*Hyukil Kwon, YoungJong Ko, and InAe Kim are supported by the second stage of Brain Korea 21.

Odontogenic keratocysts are benign intraosseous tumors of odontogenic origin that occur most commonly in the jaw. In particular, they have a predilection for the angle and ascending ramus of the mandible. In contrast, odontogenic keratocysts arising in the maxillary sinus are relatively rare. Two such cases are reported herein. In addition, the English literature that concerns odontogenic keratocysts of the maxillary sinus is reviewed.

Key Words: Odontogenic keratocyst; Maxillary sinus

Tooth development involves complicated, multistep interactions between the oral epithelium and the underlying mesenchymal tissue. Ectopic teeth can arise when these tissue interactions during development are affected¹ by developmental disturbances, iatrogenic activity, or pathological conditions, such as the presence of a tumor or a cyst.^{2,3}

Odontogenic keratocysts (OKC) have high recurrence rates, mitotic counts and epithelial turnover rates, and are the most aggressive of the odontogenic cysts in the oral cavity. In addition, unlike most cysts, usually thought to grow solely due to osmotic pressure, the epithelium in the OKC appears to have innate growth potential, which is consistent with a benign tumor. Given these features, not observed in common cysts, such as radicular and dentigerous cysts, the World Health Organization reclassified OKC as a keratocystic odontogenic tumor (KCOT) in 2005. We chose the term OKC instead of KCOT in this

report, because most of references yet contain the former term.

The clinical and radiographic features of OKC are unspecific: while some may be associated with pain, swelling, or drainage, most are asymptomatic, and radiography reveals a well-defined radiolucent area, which is also characteristic of dentigerous cysts, radicular cysts, or residual cysts.⁴ Thus, while the clinical and radiographic features can often be highly suggestive, they are not diagnostic. This means that OKC is often misdiagnosed as an ordinary cyst and is therefore undertreated, resulting in unnecessary recurrences. The aim of this paper is to report two cases of OKC associated with an ectopic tooth in the maxillary sinus, which is an unusual location. In addition, the English literature concerning this condition over the past 15 years was reviewed by searching data recorded on PubMed between 1995 and 2010, using the key words “odontogenic keratocyst” and “maxillary sinus.”

CASE REPORTS

Case 1

An 18-year-old female patient was referred with the chief complaint of a supplementary tooth, with a cyst in the left maxilla. An intraoral clinical examination did not reveal any remarkable features, such as palpable organomegaly, tenderness, edema, or

pain. Computed tomography scans and Water's view revealed an expansile cystic mass ($4.5 \times 3.1 \times 5.4$ cm) in the left maxilla, with an ectopic tooth that obstructed the ostium of the left maxillary sinus, with thinning of the left maxillary anterior, posterior, superior, and inferior walls (Fig. 1). The preoperative diagnosis was dentigerous cyst in the left maxillary sinus. Under general anesthesia, the patient underwent cyst enucleation via the Caldwell-Luc approach. In the cystic lesion, thickening of the left

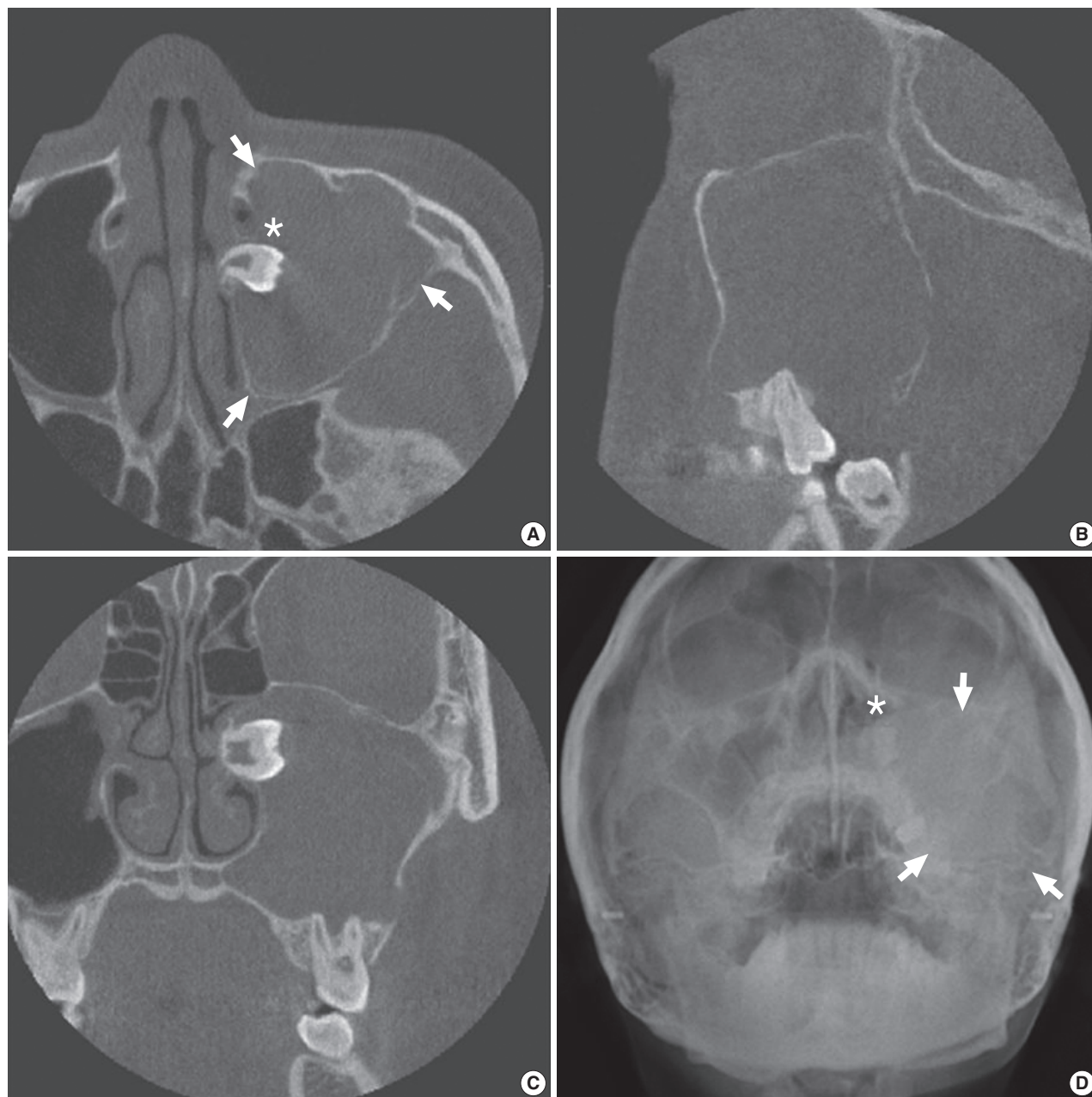


Fig. 1. (A) Axial image of the patient's left maxilla. (B) Sagittal image of the patient's left maxilla. (C) Coronal image of the patient's left maxilla. (D) Water's view of the patient. *indicates the ectopic tooth; arrows indicate the boundary of the cystic lesion.

maxillary mucosa and bony septum formation were detected. The cystic wall in the left maxillary sinus was removed *in toto* by surgical curettage, and the supplementary tooth was extracted using a kelly. Microscopically, the cyst wall consisted of six to eight epithelial layers along with a distinct basal cell layer, consisting of both palisading columnar and cuboidal cells, with polarized and hyperchromatic nuclei; the luminal surface of the cyst had a parakeratinized, corrugated appearance (Fig. 2A). There was extensive chronic inflammation along the epithelial lining of the cyst, which had infiltrated into the cyst wall and

consisted of lymphocytes, plasma cells, polymorphonuclear cells, and many foam cells (Fig. 2B). The diagnosis of OKC was made on the basis of these histological features. The patient has been asymptomatic for 4 months after the operation.

Case 2

A 19-year-old male patient was referred to the oral and maxillofacial surgery department, because of nasal obstruction, rhinorrhea, and sneezing. His medical history showed that he had

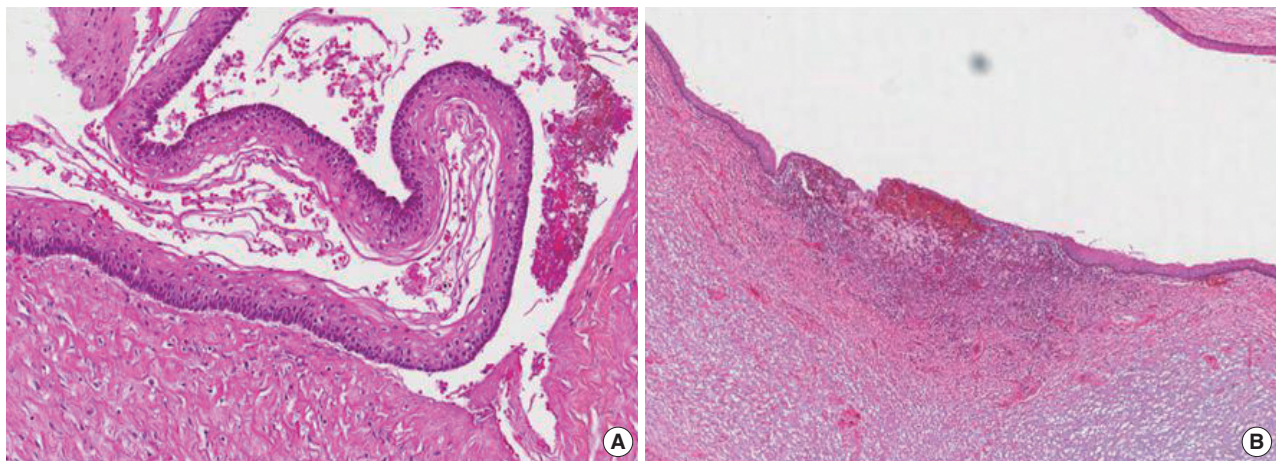


Fig. 2. Histological view of the odontogenic cyst wall. (A) The epithelial lining has a hyperchromatic and palisaded basal cell layer, is 6 to 8 cells thick, has a corrugated parakeratotic surface, and is detached from the connective tissues. (B) The characteristic microscopic features of the KCOT are lost in the central area of this portion of the cystic lining, due to inflammatory cell infiltration.

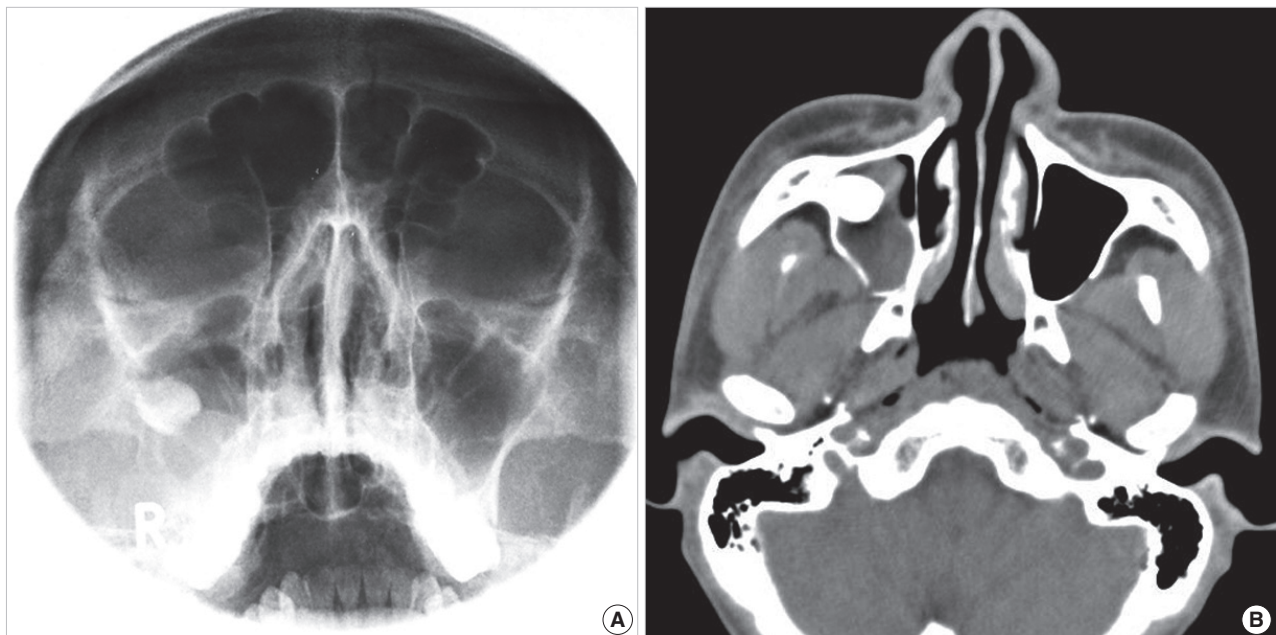


Fig. 3. (A) Water's view of the patient. (B) Axial image of the patient's right maxilla.

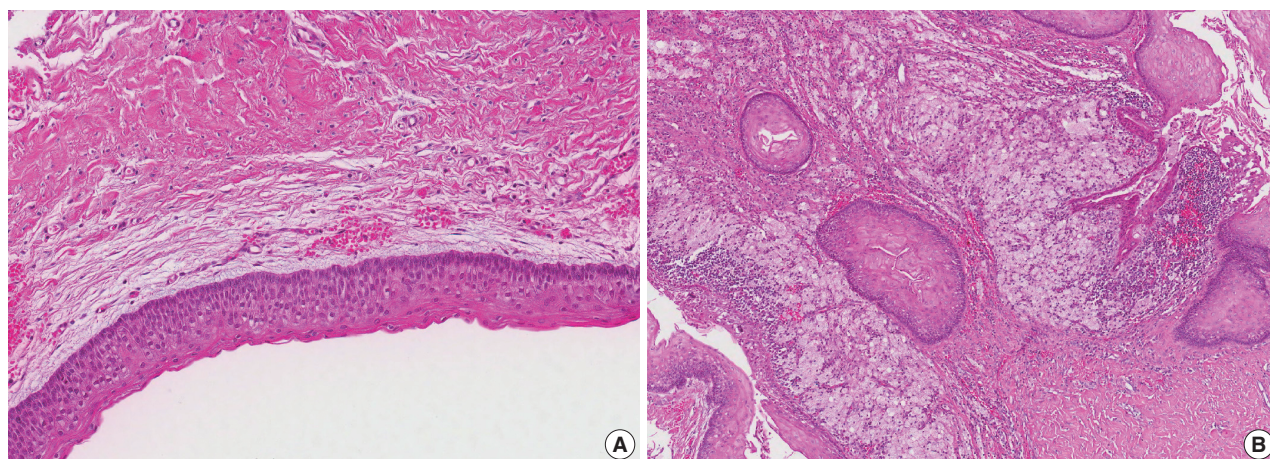


Fig. 4. (A) Characteristic odontogenic keratocystic lining and (B) variously sized daughter cysts.

taken methylprednisolone 60 mg and solondo 7.5 mg for Henoch-Schonlein purpura on his upper and lower extremities. Water's view radiography revealed an ectopic tooth on the lateral wall of the maxillary sinus (Fig. 3A). This was confirmed by computed tomography of the paranasal sinus, which revealed a hazy mass in the right maxillary sinus that had the density of bone (Fig. 3B). The preoperative diagnosis was dentigerous cyst in the right maxillary sinus. Under general anesthesia, the patient underwent cyst enucleation via the Caldwell-Luc approach. After the operation, a biopsy was performed. Microscopic analysis revealed uniform cystic wall, with a basal cell layer consisting of cells with palisading pattern (Fig. 4A). The cystic wall also included variously sized daughter cysts (Fig. 4B). These histological findings established the diagnosis of OKC. The patient has been asymptomatic for 3 months after operation.

DISCUSSION

In 1956, Philipsen⁵ first described an odontogenic cyst with a keratinous epithelial lining and in 1962, Pindborg and Hansen⁶ described the typical histological features of OKC as follows. The lining epithelium is usually very thin and uniform in thickness, with little or no evidence of rete ridges. There is also a well-defined basal cell layer and a thin spinous layer, that often directly adjoins the palisaded arrangement. The cells of the spinous layer frequently exhibit intracellular edema. Keratinization is predominantly parakeratotic, but can also be orthokeratotic. The keratin layer is often corrugated. The fibrous cystic wall is generally thin and is usually not inflamed.⁷

The growth and biological behavior of OKC differ from

those of common cysts, such as dentigerous, residual, and radicular cysts. Thus, OKC may originate from the cell rests of the dental lamina, in both the mandible and maxilla. In contrast, common odontogenic cysts are thought to arise from fragments of epithelium from the crown, remnants of the epithelial rests of Malassez (derived from the development of tooth root sheaths), remnants of the epithelial rests of Seres (derived from the development of primary tooth lamina), and fragments of tooth germ cells.⁴ Some investigators have also suggested that OKC may grow due to active proliferation of the epithelial lining, whereas dentigerous and radicular cysts develop as a result of increasing osmotic pressure within the lumen.⁸

It is quite unusual for OKC to occur in the maxillary sinus, as its most common location is either the posterior portion of the mandible or the mandibular ramus.⁸ In a review of 429 patients, Valter *et al.*⁹ reported that 70.2, 12.4, 12.8, and 4.7% of the OKCs occurred in the mandible, maxilla, soft tissues, and the maxillary sinus, respectively. It has been shown that an unerupted tooth is involved in the lesion in 25–40% of the cases.^{4,10}

OKC usually presents in the second to third decade of life, but a wide range of 5 to 80 years has been reported.⁴ With regard to OKC associated with an ectopic tooth, there have been only eight case reports to date (Table 1).^{11–17} The age range of these cases was 8 to 39 years, and the mean age was 21.4 years. Four patients were under the age of 20. For the two cases reported in the present paper, the mean age was 18.5 years. Of the eight previous case reports, there was an equal incidence among women and men (i.e., four each). Nevertheless, all cases exhibited some histological variations: ciliated changes were detected in two cases, limited cysts in a sinus were detected in

Table 1. Summary of available clinical data of all reported cases of odontogenic keratocyst associated with an ectopic tooth

Reference	Age (yr)/ Gender	Concerned composition	Symptom	Treatment	Variant feature	Outcome
Hunter <i>et al.</i> ¹¹	9/F	Bilateral ectopic tooth, orbital floor, pterygomaxillary space	Swelling	Midfacial degloving	Unknown	Unknown
Makowski <i>et al.</i> ¹²	34/M	Third molar, anteromedial wall	Pain, swelling	Caldwell-Luc	SCC	NED for 42 mo
Yamazaki <i>et al.</i> ¹³	39/F	Second premolar, inferior portion	Swelling	Unknown	Ciliated change	Unknown
Tanimoto <i>et al.</i> ¹⁴	8/F	Second premolar, sinus roof	Swelling	Marsupialization	Impacted tooth eruption	NED for 5 yr
Vencio <i>et al.</i> ¹⁵	27/F	Second molar, sinus floor	Pain	Resection by curettage	Ciliated change	NED for 5 yr
Costa Carvalho Silva <i>et al.</i> ¹⁶	17/M	Third molar, lateral portion	Headache	Enucleation	Limited in sinus	NED for 8 yr
<i>et al.</i> ¹⁶	14/M	Second molar, upper posterior portion	Bad taste	Caldwell-Luc	Limited in sinus	NED for 5 yr
Cakur <i>et al.</i> ¹⁷	23/M	Third molar, lateral wall	Pain, swelling	Caldwell-Luc	-	NED for 6 mo

F, female; M, male; SCC, squamous cell carcinoma; NED, no evidence of disease.

two cases, squamous cell carcinoma was found in one case, and impacted tooth eruption was observed in one case. The teeth involved in these OKC cases were the third molar (three cases), the second molar (two cases), the second premolar (two cases), and bilateral ectopic teeth (one case).

Most OKCs are treated similarly to other odontogenic cysts. According to Schmidt and Pogrel,¹⁸ the ideal treatment for the OKC would be enucleation or curettage, followed by treatment of the cavity with an agent, such as liquid nitrogen or Carnoy's solution, to kill the epithelial remnants or satellite cysts. As presented in Table 1, in the rare cases of OKCs associated with an ectopic tooth in the maxillary sinus, the most common treatment was the Caldwell-Luc procedure.^{12,16,17} Although the traditional Caldwell-Luc procedure provides a direct view of the maxillary sinus, this procedure is associated with increased morbidity, compared to endoscopic sinus surgery.¹⁹ If the tooth is small and sited near the maxillary ostium, transnasal extradiation of the tooth may be attempted. In our first presented case, the tooth was located in the maxillary ostium, but it was oversized and therefore we could not use the maxillary ostium approach.²

The ectopic eruption of teeth in regions other than the oral cavity is rare, although there have been reports of teeth in unusual locations, such as the nasal cavity, mandibular condyle, coronoid process, and palate. One of the non-dental sites in which an ectopic tooth has been observed is the maxillary sinus. The etiology of ectopic eruption has not yet been completely clarified, but many possible etiologies have been suggested. They include trauma, infection, developmental anomalies, and pathological conditions, such as odontogenic cysts. With regard to the latter, it is known that, as the growth of an odontogenic cyst continues, the cyst encroaches on the space of the sinus and displaces its borders: it may be that the displacement of teeth buds by this expansion of a cyst results in the "ectopic" erup-

tion of a tooth.¹⁰ Indeed, radiography of case 1 reported here revealed the ectopic tooth impacted in the ostium of the left maxillary sinus, which is a rare location for an ectopic tooth, and also showed an extremely narrow sinus that had been pushed by an expansile cystic mass.

In case 1, the cyst also exhibited chronic inflammation, which had infiltrated the cystic lining. Where inflammation was present, the cystic lining lost the characteristic patterns described above and resembled the non-keratinized, stratified squamous lining of other inflammatory cysts. This demonstrates that OKC associated with inflammatory infiltration may be readily misdiagnosed. Indeed, de Paula *et al.*²⁰ reported that inflammatory cells increase the number of cycling epithelial cells, and that this increased epithelial cell proliferation may disrupt the typical structure of the OKC lining layers.

Since it is rare to find an OKC in the maxillary sinus, its detection in this site by radiography may result in misdiagnosis. Computed tomography can provide further information regarding the extent of these lesions, which can aid both diagnosis and preoperative preparation. Nevertheless, it is difficult to diagnose OKC because of its relative lack of specific clinical and radiographic characteristics. Since OKC has features similar to those of both dentigerous cysts and ameloblastomas, which are the most common provisional diagnoses for OKC, the best way to diagnose OKCs may be to combine accurate clinical, radiographic, and trans-surgical observations with a biopsy specimen examination; this approach will help to determine the most effective treatment, thereby preventing recurrences.

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