

9세 남자 환아에서 급성 부비동염의 드문 원인 : 과잉치가 동반된 감염된 함기성 낭종

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An Unusual Cause of Acute Maxillary Sinusitis in a 9-year-old Child: Odontogenic Origin of Infected Dentigerous Cyst with Supernumerary Teeth

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Acute maxillary sinusitis is a common disorder affecting children. Untreated acute sinusitis can develop into chronic sinusitis, and complications, such as orbital cellulitis or abscess, can occur. Maxillary sinusitis of odontogenic origin is not a well-recognized condition and is frequently missed in children. As an odontogenic source of sinusitis, the dentigerous cyst is one of the most prevalent types of odontogenic cysts, and it is associated with the crown of an unerupted or developing tooth. This report concerns a nine-year-old boy who was diagnosed with maxillary sinusitis originating from a dentigerous cyst with supernumerary teeth. The boy visited our pediatric clinic presenting with rhinorrhea and nasal obstruction and was initially diagnosed with maxillary sinusitis only. With antibiotic treatment, his symptoms seemed to improve, but after 2 months, he came to our clinic with left facial swelling with persistent rhinorrhea and nasal obstruction. Radiographic examinations of the sinuses were performed, and he was diagnosed with maxillary sinusitis originating from a dentigerous cyst with supernumerary teeth. After a surgical procedure involving the removal of the dentigerous cyst with supernumerary teeth, the symptoms of sinusitis gradually diminished. There are only very few cases in the pediatric medical literature that remind us that odontogenic origin can cause maxillary sinusitis in children. Our patient can act as a reminder to general pediatricians to include dentigerous cysts in the differential diagnosis of maxillary sinusitis.

Key Words: Maxillary sinusitis, Dentigerous cyst, Supernumerary teeth, Children

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Introduction

Acute sinusitis is defined as the inflammation of the mucosa of the paranasal sinuses and it is usually developed after upper respiratory tract infection¹⁾. Though paranasal sinuses are developed with the age, maxillary sinus and ethmoid sinus are exist from birth and large enough to cause infection²⁾. Acute sinusitis may present with different symptoms and signs that may not be specific. The diagnosis of acute sinusitis can be based

on clinical criteria alone in children who present with upper respiratory symptoms that are either persistent with no improvement after 10 days or severe for more than 3 days³⁾.

Maxillary sinusitis originating from an odontogenic source has been reported to account for approximately 10-12% of cases of maxillary sinusitis⁴⁾. Despite its prevalence, acute maxillary sinusitis of odontogenic origin is often overlooked in pediatric practice. This is because clinical features in young children are often nonspecific and there are some conceptions that acute maxillary sinusitis of odontogenic origin is rare in children.

Maxillary sinusitis of odontogenic origin occurs when the Schneiderian membrane is violated by conditions such as maxillary posterior teeth, maxillary trauma, pathologic lesions of the jaws and teeth, or iatrogenic causes, such as dental and maxillofacial surgery complications, and infection, which can easily spread into the maxillary sinus, leading to maxillary sinusitis⁵⁾. For this reason, an odontogenic source should be considered in individuals with symptoms of maxillary sinusitis with a history of dental or jaw pain, dental infection, or dental surgery and in those who are resistant to conventional sinusitis therapy⁶⁾.

Maxillary sinusitis originating from dentigerous cysts has rarely been reported in the pediatric medical literature⁷⁾. We report a rare case of infected dentigerous cyst of maxillary sinusitis with radiologic and pathologic features arising from supernumerary teeth and resistance to conventional antibiotic therapy in a 9-year-old boy.

Case Report

A previously healthy 9-year-old boy came to our pediatric clinic with complaints of nocturnally worsening cough and nasal discharge and obstruction. He was initially diagnosed with maxillary sinusitis based on the symptoms and past medical history of allergic rhinitis with allergic conjunctivitis, and he was treated with antihistamines, intranasal corticosteroid spray, and anti-

biotics.

After the completion of 2 weeks of treatment, his symptoms seemed to have improved, and a 4-week course of leukotriene-receptor antagonist with intranasal corticosteroid spray was additionally prescribed.

Two months later, the boy was referred to our clinic and admitted because of the abrupt onset of left facial swelling with jaw pain and persistent rhinorrhea with nasal obstruction. Although he had taken antibiotics for 5 days, there had been no improvement. He did not have a fever, and he had been previously healthy and had no history of past illness including admission or trauma to the jaw, except sinusitis with allergic rhinitis and allergic conjunctivitis. A physical examination revealed facial swelling from the left nostril to the nasolabial fold, and the swelling was soft and painful upon light touch. However, there was no warmth, fluctuation, discoloration of the overlying skin, or discharge from the swollen lesion.

On the first day of admission, he had a fever of up to 38.1°C. The paranasal sinus view of the radiographic examination showed maxillary sinusitis with a cystic lesion surrounded by the supernumerary tooth in the left anterior maxilla (Fig. 1). He was immediately examined by oral and maxillofacial surgeons, and cone-beam computed tomography (CT) was performed. In the panoramic view of the cone-beam CT, there was a large, low-density, non-enhancing, well-circumscribed cystic lesion, which is called a dentigerous cyst (Fig. 2). The dentigerous cyst included three supernumerary

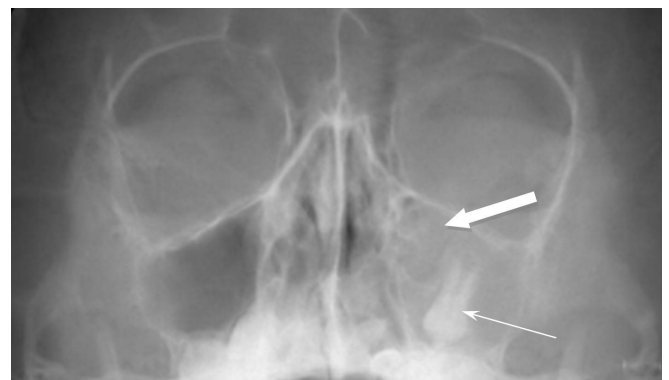


Fig. 1. Paranasal sinus view of dentigerous cyst (thick arrow) with supernumerary tooth (thin arrow) in left maxillary sinus.

teeth (Fig. 3A, B) and caused resorption of the cortical bone, resulting in a 3×5 cm-sized bony defect on the left maxillary anterior wall. On the basis of these clinical and radiographic characteristics, the diagnosis of “maxillary sinusitis with dentigerous cyst associated with impacted supernumerary teeth” was made.

The therapeutic approach was planned to perform the surgical procedure (the Caldwell–Luc operation) with enucleation of the dentigerous cyst and extraction

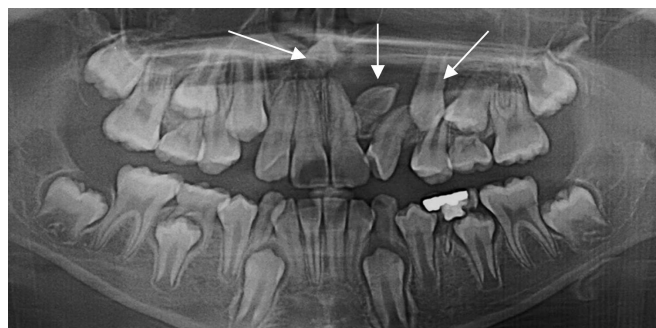


Fig.2. Pre-operative panoramic view showed supernumerary teeth (arrows).

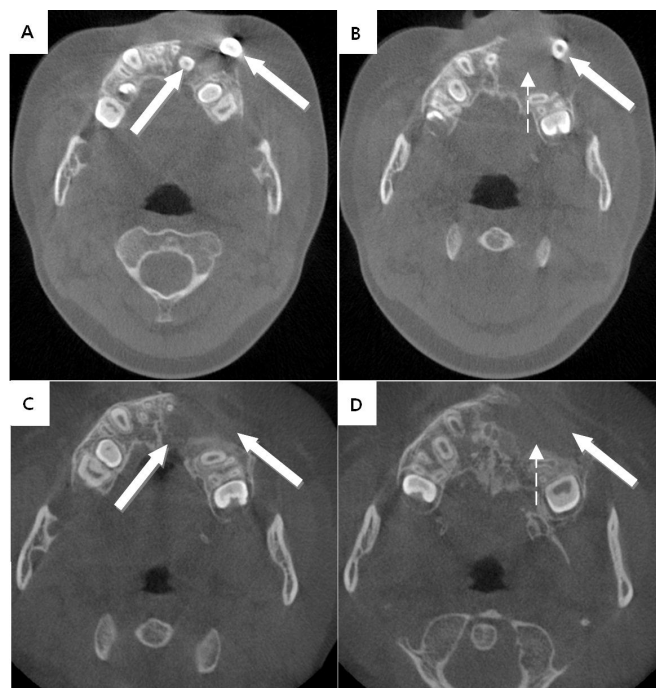


Fig. 3. Total of three supernumerary teeth cyst (thick arrows) and dentigerous cyst (dotted arrow) in initial cone-beam computed tomography (A, B). Supernumerary teeth (thick arrows) and dentigerous cyst (dotted arrow) are completely disappeared in repeated cone beam CT after 3 months (C, D).

of the supernumerary teeth, and surgical intervention was conducted on the fifth day of admission. A removed dentigerous cyst was measured to be 5.0×6.0×0.5 cm, and the impacted supernumerary teeth were conical in shape. The histopathological examination of the cystic lining was performed to differentiate the diagnosis from those of radicular cyst, odontogenic keratocyst, and odontogenic tumor, and no evidence of malignant changes was noted. The fluid from the swollen lesion was aspirated for bacterial culture, and viridans group streptococci was reported. Consequently, the cyst was diagnosed as a dentigerous cyst arising from supernumerary teeth called mesiodens.

The boy’s postoperative period was uneventful. He was discharged from hospital 3 days after the surgical procedure. The facial swelling had decreased considerably, and he was symptom-free on clinical follow-up after 2 weeks.

Three months later, repeated cone-beam computed tomography showed no recurrence of the dentigerous cyst (Fig. 3C, D) and the boy had no symptoms of maxillary sinusitis. Although there was a complete resolution of the facial swelling, the left anterior maxillary wall defect had been unchanged. Because there was a possibility of spontaneous bone regeneration, he was only advised to correct problems with lack of dentition for orthodontic and prosthetic rehabilitation.

Discussion

Sinusitis is a common illness in childhood and adolescence and is associated with significant acute and chronic morbidity as well as the potential for serious complications. Acute sinusitis usually occurs as a late complication of upper respiratory tract infections and chronic sinusitis is caused when acute sinusitis is repeated or it is not appropriately treated¹⁾. In children and adolescents with sinusitis, nonspecific symptoms such as stuffy nose, unilateral or bilateral purulent rhinorrhea, fever, or cough may appear, and sometimes complications such as foul breath, decreased sense of smell, swelling around the eyes may be accompanied.

Rarely, headache, facial pain or tenderness and maxillary teeth region discomfort can appear⁸⁾.

Sinusitis of odontogenic origin, such as dentigerous cysts, accounts for approximately 10–12% of all maxillary sinusitis cases⁴⁾, but it is not a well-recognized condition in children. This is because the second and third decades of life are common ages for the occurrence of dentigerous cysts⁹⁾, and they are extremely uncommon in childhood.

A dentigerous cyst is defined as a “benign expansive lesion derived from hydrostatic expansion of a dental follicle and surrounds the crown of an unerupted tooth”¹¹⁾. Although the pathogenesis of these cysts is unknown¹²⁾, typical dentigerous cysts are believed to originate from the enamel organ and the follicle of the unerupted tooth¹³⁾ after amelogenesis has finished. Dentigerous cysts are also associated with unerupted teeth and most commonly involve the mandibular third molar and maxillary canine and rarely other teeth^{5,11,13)}, but approximately 95% of these cysts involve permanent dentition and only 5% are associated with supernumerary teeth¹⁴⁾.

These cysts are initially completely asymptomatic. Dentigerous cysts progress slowly, may exist for several years without being noticed^{11,13)} and are usually found during a routine radiographic examination⁷⁾. As dentigerous cysts in the maxillary bone or sinuses may increase in size or become infected, many complications can occur. Patients usually complain of nasal obstruction, purulent rhinorrhea, foul odor or taste, external nasal deformity, and facial swelling¹⁵⁾. The similarity between symptoms of odontogenic maxillary sinusitis and other causes of sinusitis can obstruct early diagnosis, and misdiagnosis can cause permanent symptoms that are resistant to medical and surgical therapies.

Unlike most other forms of maxillary sinusitis, treating odontogenic maxillary sinusitis requires a combination of medical and surgical management. Medical management includes the use of antibiotics, decongestants, and moisturizing sprays in a manner similar to the treatment of routine maxillary sinusitis, and surgical management involves eliminating the source of the

infection to prevent recurrence of the condition and performing a pathologic examination to rule out other more aggressive lesions^{16,17)}.

The patient in our case was diagnosed with maxillary sinusitis originating from a dentigerous cyst with supernumerary teeth. He presented with rhinorrhea and nasal obstruction and was diagnosed with maxillary sinusitis only. He was initially treated with antibiotics, and his symptoms seemed to improve. After 2 months, he came to us with facial swelling and persistent rhinorrhea with nasal obstruction in spite of 5 days of antibiotic treatment. Radiologic examinations were performed, and we found a dentigerous cyst with supernumerary teeth causing maxillary sinusitis. The patient was transferred to the oral and maxillofacial surgeons, and therapeutic surgery was conducted with enucleation of the dentigerous cyst and extraction of the supernumerary teeth.

When a patient with sinusitis is resistant to standard medical treatment, the odontogenic origins of the infection must be considered. We also have to consider radiographic examinations, because a differential diagnosis of sinusitis is needed. Early diagnosis and proper treatment will reduce the incidence of complications and improve the quality of life in children with maxillary sinusitis. For these reasons, although it is a rare cause of sinusitis in children, dentigerous cysts should be included in the differential diagnosis for the cause of persistent or recurrent sinusitis.

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요약

급성 상악동염의 원인으로는 급성 상기도 감염이 가장 흔하며 함치성 낭종 등의 치성원인에 의한 경우는 상악동염의 10-12%를 차지한다. 함치성 낭종은 주로 치과에서 우연한 방사선학적 검사에 의해 발견되며 소아에서는 드물게 보고되고 있다. 함치성 낭종이 과잉치와 동반되었을 때 주위 상악골의 파괴와 치근의 흡수를 야기하거나 침범된 치아의 변위를 유발할 수 있으므로, 조기 진단과 적절한 치료가 중요하다. 본 증례에서 콧물과 코막힘으로 부비동염으로 진단받았던 9세 남자 환아가 2개월 뒤 좌측 안면부 연부조직염으로 내원하여 시행한 방사선학적 검사에서 좌측 상악동에 과잉치와 동반된 함치성 낭종이 관찰되었으며, 치료로 외과적 절제술을 시행하였다. 저자들은 소아에서 부비동염의 원인으로 과잉치를 동반한 감염된 함치성 낭종을 경험하였으며, 부비동염의 진단과 치료에 있어 항생제 치료로 호전되지 않거나 증상이 지속될 시 단순 상기도감염 합병증 외에 다른 질환의 감별을 고려할 것을 당부하는 바이다.