

Mucocele in the maxillary sinus involving the orbit: A report of 2 cases

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ABSTRACT

Mucocele of the paranasal sinuses is a benign, slow-growing, expansile lesion. Maxillary sinus mucoceles are usually associated with painless bulging of the cheek; however, orbital expansion is rarely observed. Maxillary sinus mucoceles can be classified as primary or secondary according to their etiology. An impediment to sinus ostium ventilation is thought to be the cause of primary mucocele, while sequestering of residual mucosa after surgery in the wound and long-term retention of tissue fluid have been suggested to lead to the formation of secondary mucocele. This report presents 2 cases of primary and secondary mucoceles, with a focus on radiographic features. As primary and superiorly positioned secondary maxillary sinus mucoceles are uncommon and their close proximity to the orbit predisposes the patient to significant morbidity, the authors expect that this report will contribute to a better understanding and diagnosis of maxillary sinus mucocele involving the orbit. (*Imaging Sci Dent* 2022; 52: 327-32)

KEY WORDS: Maxillary Sinus; Mucocele; Orbit

Paranasal sinus mucocele is a benign epithelial-lined cystic structure containing mucus that expands in size and can be locally destructive.^{1,2} The frontal sinus is most commonly affected, followed by the ethmoid sinuses, with reports suggesting that 70%-90% of mucoceles occur in these locations.²

The general mechanism of cyst formation is an accumulation of fluid in a mucoperiosteal-lined cavity resulting from obstructed sinus outflow due to inflammation, trauma, or sinus surgery.^{1,3,4} As mucus accumulates within the cavity, it enlarges gradually, resulting in erosion and remodeling of the surrounding bone.¹

The predisposing etiologies of primary and secondary mucocele seem to be different.⁵ Inflammatory blockage of mucus drainage, secretory duct obstruction, cystic dilation of mucosal glands, and cystic degeneration of polyps have been proposed as possible mechanisms in the pathogenesis of primary mucocele.^{5,6} Contrastingly, sequestration of resi-

dual mucosa after surgery and long-term tissue fluid retention have been suggested as potential triggers for secondary mucocele.⁵⁻⁷ As secondary mucocele tends to occur in sinuses that have previously undergone surgery, such as the Caldwell-Luc operation (CLOP), the maxillary sinus is the most common sinus for secondary mucocele development.⁵

Maxillary sinus mucocele is relatively rare, accounting for fewer than 10% of all paranasal mucocele cases reported in the English-language literature.^{8,9} However, secondary maxillary sinus mucocele has been commonly reported in Korea and Japan because of the preference for surgery related to inflammation of the maxillary sinus. They have been reported as post-operative cheek cysts,¹⁰ postoperative maxillary mucoceles,^{11,12} and postoperative maxillary cysts (POMCs),^{13,14} typically occurring as long-term complications of CLOP.

This report describes the characteristic radiographic features of primary and secondary maxillary sinus mucocele expanding to the orbit in 2 cases. The primary mucocele showed unilocular and spherical expansion. The secondary mucocele, in a patient with a CLOP history, presented as a multilocular lesion with a slow-growing tendency.

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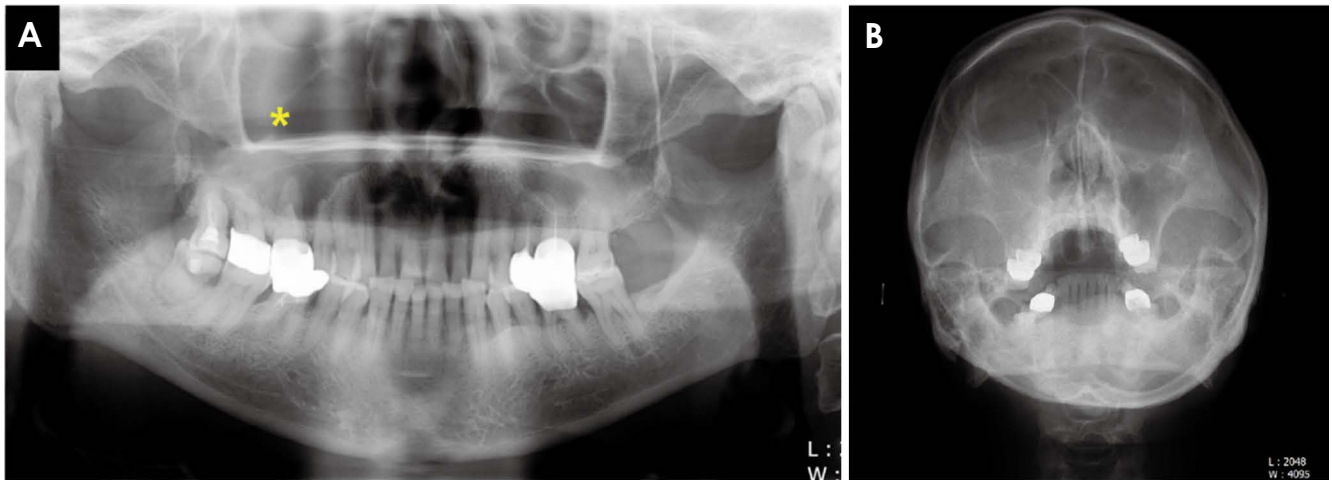


Fig. 1. A. A panoramic radiograph shows a well-defined expansile cystic lesion (asterisk) in the right maxillary sinus. Downward displacement of the maxillary sinus floor and upward displacement of the orbital floor are observed. B. A Waters' radiograph reveals opacification of the entire right maxillary sinus.

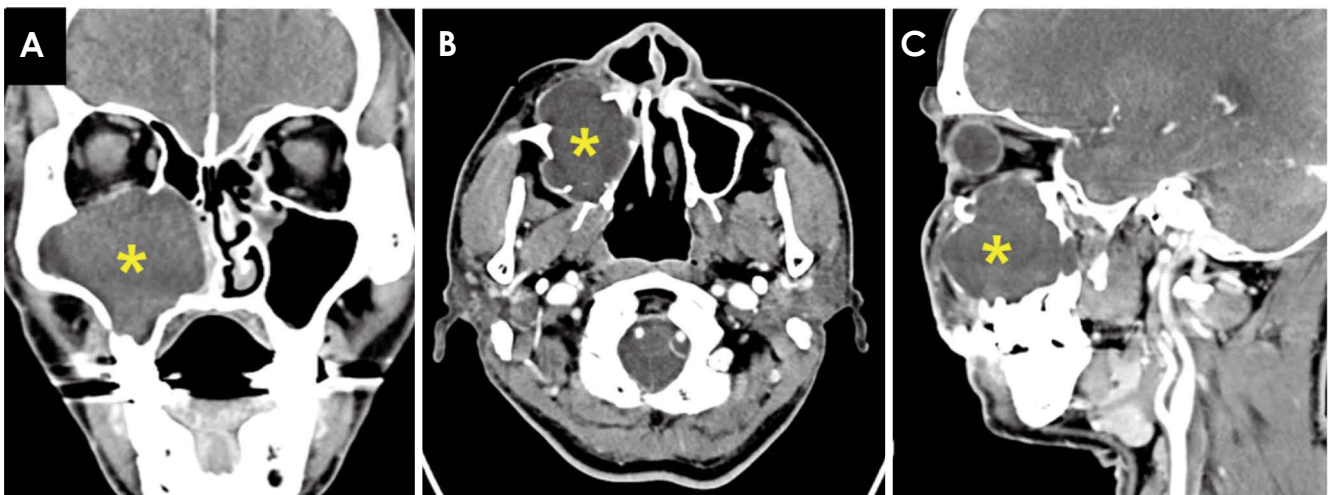


Fig. 2. A. An enhanced computed tomographic coronal image shows a spherical, unilocular, expansile lesion (asterisk) in the right maxillary sinus, elevation and thinning of the orbital floor, and displacement of the orbital contents. B. An axial image shows bony erosion and remodeling of the surrounding walls. Contrast enhancement is not observed. C. A sagittal image shows expansion of surrounding walls and elevation of the orbital floor.

Case Report

This study was approved by the Institutional Review Board of Wonkwang University Dental Hospital (IRB number: WKDIRB202106-04). Written informed consent was obtained from the following 2 patients to publish this case report and any accompanying images.

Case 1

A 69-year-old man visited Wonkwang University Dental Hospital complaining of swelling in the right infraorbital

region. The symptoms had persisted for the past 2 months. The patient had no significant medical history, and no surgery, including CLOP, had been performed. No symptoms were observed in the nasal or dental region.

A panoramic radiograph showed a well-defined expansile lesion involving the entire right maxillary sinus (Fig. 1A). The sinus floor deviated downward, and the posterior wall showed a scalloped margin. The orbital floor deviated superiorly. A Waters' radiograph revealed complete opacification of the right maxillary sinus and superior deviation of the orbital floor (Fig. 1B).

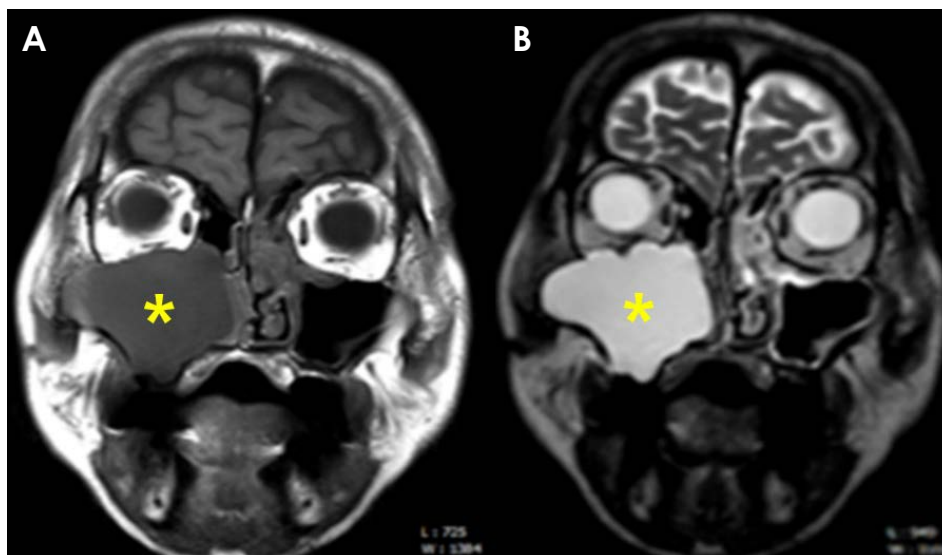


Fig. 3. Magnetic resonance imaging shows a well-defined homogeneous lesion in the right maxillary sinus (asterisk). A. A T1-weighted magnetic resonance image shows an expansile hypointense lesion. B. A T2-weighted magnetic resonance image shows hyperintensity of the lesion. Marginal lobulation can be observed on the superior and inferior borders of the cystic lesion. A slight upward displacement of the eyeball can be observed.

Due to the wide extent of the lesion, multidetector computed tomography with contrast medium injection was performed to determine the exact extent of the lesion and whether the content of the lesion was solid or liquid. The images obtained showed a unilocular, remarkable expansile lesion in the right maxillary sinus (Fig. 2). Bony expansion, probably due to sinus ostium obstruction, was prominent in the lateral, medial, orbital, and downward directions with a spherical shape. The lesion severely extended superiorly, eroded the orbital floor, and contacted the inferior rectus muscle. As there was no enhancement inside the lesion, it could be observed that the lesion was filled with fluid. As the surrounding bone was severely displaced and eroded, an additional magnetic resonance imaging (MRI) examination was performed to closely observe changes in the surrounding soft tissues, including the eyeball and peri-orbital muscles (Fig. 3). MRI revealed a well-defined homogeneous hypointense T1 and hyperintense T2 signal cystic lesion occupying the right maxillary sinus. Marginal lobulation was detected at the superior and inferior borders of the lesion. Pathological changes in the eyeball and peri-orbital muscles were not evident. Considering all the radiological and clinical data, the imaging diagnosis was primary maxillary sinus mucocele.

Surgical excision and histopathological examinations were performed. The surgical site revealed a cystic mass protruding into the anterior wall of the right zygomatic bone, which was clearly detached from the surrounding wall.

Histopathologically, a wall with thick edematous submucosal tissue was observed beneath the pseudostratified ciliated cylindrical epithelium (Fig. 4). Based on both the

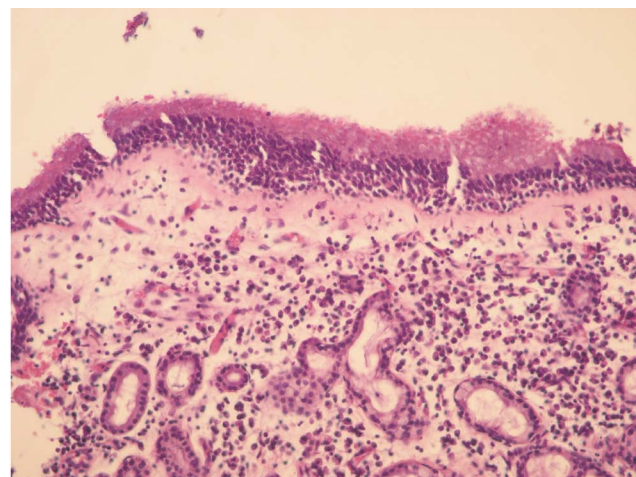


Fig. 4. A histopathological image shows a wall with thick edematous submucosal tissue beneath the pseudostratified ciliated cylindrical epithelium.

histopathological and radiographic findings, the final diagnosis was primary maxillary sinus mucocele.

There was no evidence of recurrence during the 1-year follow-up period. Normal bone healing was observed without pathological findings.

Case 2

A 53-year-old man visited the Department of Otolaryngology at Wonkwang University Hospital complaining of difficulty in opening his right eye for the past 6 months. He first visited a private ophthalmology clinic and was referred to the hospital because radiographs revealed a cystic mass in the infraorbital region.

The patient had a previous history of CLOP approximate-

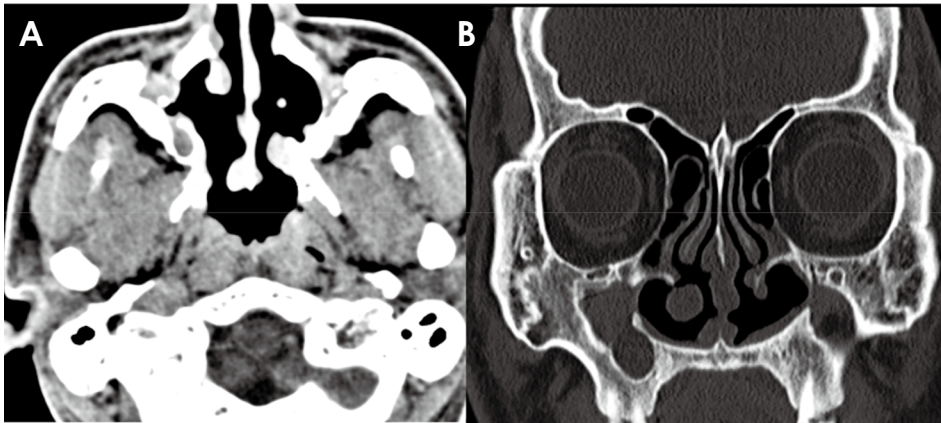


Fig. 5. A. An axial computed tomographic image (the first visit, taken 8 years previously) shows bilateral collapse of the right maxillary sinus, suggesting a previous Caldwell-Luc operation. B. Computed tomographic coronal image shows a region surrounded by remodeled bone and aeration loss in the right maxillary sinus, but no distinct expansion can be detected.

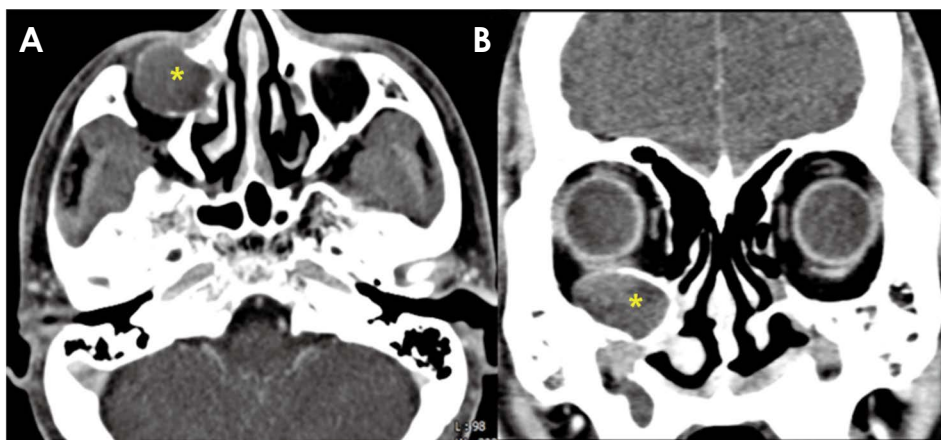


Fig. 6. A. An axial computed tomographic image (second visit) shows a well-defined, enlarged multilocular cystic lesion extending superiorly to the orbit (asterisk). The Hounsfield units inside the lesion are similar to those of fluid but not soft tissue, indicating that the lesion is filled with fluid. B. A coronal computed tomography image shows erosion of the right orbital floor and displacement of the right eyeball.

ly 30 years prior. Therefore, a computed tomography (CT) scan performed 8 years previously, taken for long-term follow-up after surgery, was available as an imaging record (Fig. 5). It showed bilateral surgical defects and collapse of the maxillary sinus as postoperative changes following CLOP. In the right maxillary sinus, there was a region surrounded by remodeled bone and loss of aeration, but there was no distinct expansion.

CT was performed to evaluate the cause of the difficulty in opening the right eye (Fig. 6). It showed a well-defined, enlarged, multilocular cystic lesion extending superiorly to the orbit. The orbital floor was eroded, and the right eyeball was displaced by the lesion. The lesion showed compartments due to a bony septum. The low attenuation inside the lesion was consistent with the fluid level and not the soft tissue, indicating that the lesion was filled with fluid. Considering all radiological and clinical information, the preoperative diagnosis was secondary maxillary sinus mucocele.

CLOP combined with inferior meatal antrostomy was performed. The lesion contained a large amount of chocolate-colored pus. The lesion was finally diagnosed as sec-

ondary maxillary sinus mucocele based on clinical, radiographic, and operative findings. A histological examination was not performed.

There was no evidence of recurrence during the 1-year follow-up.

Discussion

In 1978, Canalis et al. defined mucocele as a chronic, expansile, and cyst-like lesion of the paranasal sinuses.¹⁵ It contains sterile mucoid secretions and is surrounded by the mucosa of the affected cavity. The expansile nature of mucocele may cause bone erosion owing to compression and consequent bone resorption.

Maxillary sinus mucoceles are usually asymptomatic or found incidentally.¹ The symptoms of mucoceles are related to their expansion and subsequent pressure on the surrounding anatomic structures. They commonly present as painless bulges in the cheek.² Medial expansion of the maxillary sinus into the nasal cavity often causes nasal obstruction. Superior expansion into the orbit can cause

displacement of the orbital contents and sometimes visual changes. Downward displacement into the alveoli may cause teeth loosening.¹⁶

Among the various types of secondary maxillary sinus mucocele, postoperative maxillary mucocele has been introduced as a synonym for postoperative cheek cysts and POMC. These secondary mucocèles arise due to the entrapment of mucosal remnants following surgery, such as CLOP. POMC shows postoperative changes, which include a contracted maxillary sinus, mainly due to depression of the surrounding walls, and a thickened bony wall of the maxillary sinus accompanied by a heterogeneous lumen.^{17,18} Moreover, bony defects in the anterior or medial wall of the maxillary sinus can be observed.¹⁷

Although there have been some previously reported cases,^{13,19,20} secondary maxillary sinus mucocèles that affect the orbit are rare because of the location of the surgical procedure. Many cases of extended cystic lesions in the posterolateral portion of the maxillary sinus have been reported.²¹

In contrast, primary maxillary sinus mucocele commonly involves the entire antral lumen in nonsurgical cases.²¹ The etiology of primary mucocele is not well known, but it is presumed to be related to ostium obstruction.

In the early stages, mucocele appears as a nonspecific, uniformly clouded, fluid-filled lesion with normal bony walls on radiographs. In the later stages, the sinus cavity expands, and the walls are thinned and eventually eroded.^{1,2,22} The continuing expansion of mucocele is a characteristic feature attributed to the retention of secretions. Subsequently, pressure necrosis and subsequent bony erosion of the surrounding walls can be observed.³

In the first case, the downward expansion of the inferior border of the lesion was clearly observed in the panoramic view, and opacification of the entire maxillary sinus was present on the Waters' radiograph. This case showed an aggressive tendency owing to the spherical expansion pattern. Maxillary sinus mucocele is sometimes difficult to distinguish from malignant lesions based on plain radiographic evidence alone.⁴ However, in this case, because the lesion had a well-corticated margin and showed expansion rather than destruction, it could be distinguished from a malignant lesion. Furthermore, MRI showed that the lesion had marginal lobulation with a definite border, but no infiltration tendency was observed. Marginal lobulation suggested the expansile nature of this lesion and helped exclude a malignant process.

Another lesion that must be considered in the differential

diagnosis is antral pseudocyst of the maxillary sinus. However, maxillary sinus mucocele often appears spherical, unlike the dome-shaped appearance of antral pseudocysts.²³ Primary mucocele develops from sinus ostium obstruction, and the cavity becomes completely filled with mucus produced by the sinus mucosa.^{6,18} In contrast, antral pseudocysts do not fill the cavity completely.

We presumed the etiology of this case to be ostium obstruction and subsequent inflammatory blockage of mucus drainage.

Case 2 showed a multilocular cystic lesion of the maxillary sinus extending superiorly to the orbit. The patient had a history of CLOP, and POMC was incidentally discovered approximately 22 years after CLOP. The initial radiologic imaging (Fig. 5A and B) revealed a cystic portion without prominent expansion. However, the lesion had expanded and appeared as a multilocular lesion, with another cystic lesion in the infraorbital region (Fig. 6A and B, second visit, 8 years later). This finding suggests the progressive expansile nature of secondary mucocele.

In secondary mucocele, multilocular patterns with a bony septum are usually observed, whereas primary mucocele is most often a unilocular expansile lesion. This multilocular pattern occurs because of simultaneous changes in the sinus due to surgery and cystic expansion by the trapped epithelium.

Secondary mucocele following sinus surgery generally develops as a delayed complication, typically 10-30 years after CLOP.¹⁰ Recently, secondary mucocèles have shown a decreasing prevalence.^{7,8} The reason for this is the wide use of functional endoscopic sinus surgery (FESS) as a surgical method instead of CLOP. FESS has played a dominant role by focusing on improving sinus outflow tract function by restoring sinonasal physiology, whereas CLOP aims to remove diseased mucosa rather than to improve sinus drainage.^{10,24}

In conclusion, this report presents in detail 2 representative cases of maxillary sinus mucocele with a focus on characteristic radiographic features. Primary and superiorly positioned secondary mucocèles are uncommon, but their close proximity to the orbit predisposes the patient to significant morbidity. The authors expect this study to contribute to a better understanding and diagnosis of maxillary sinus mucocele involving the orbit. As only 2 cases were analyzed in this report, it will be helpful to analyze more cases in future studies.

Conflicts of Interest: None

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