Maxillary Sinus Pneumocele Presenting as Aesthetic Deformity: A Case Report With Literature Review

Hyun Sang Cho, MD1, Seok Jung Hong, MD1, Hyun Kyu Chae, MD2, and Kyung Soo Kim, MD, PhD2

Abstract
We report an interesting case of maxillary sinus pneumocele that presented with aesthetic deformity and completely treated with Caldwell-Luc approach and thoroughly review all of the past literature focusing on clinical symptoms and surgical approach. Based on our comprehensive review of maxillary sinus pneumocele, we found 2 important characteristics. First, maxillary sinus pneumocele may be asymptomatic but cause various symptoms owing to the displacement of neighboring structures, such as facial symptoms, eye symptoms, and nasal obstruction. Second, there is no standard operation technique for maxillary sinus pneumocele, but surgical approach should be individualized depending on patient’s symptoms and needs. Therefore, more case studies are needed to confirm this.

Keywords
pneumocele, maxillary sinus, Caldwell-Luc approach, aesthetic deformity, facial pain

Introduction
Pneumocele is a rare lesion of the paranasal sinus characterized by abnormal expansion of an air-containing sinus lined with normal mucosa.1 It gradually displaces the surrounding structures as the bony sinus wall becomes thinner. This lesion can occur in any sinus, with the frontal sinus most commonly affected, whereas involvement of the maxillary sinus is an exceedingly rare condition.1,2 Maxillary sinus pneumocele was first described by Noyek et al in 1974, followed by only 10 additional cases.1,3-12 Maxillary sinus pneumocele is usually asymptomatic over long periods but may cause significant clinical symptoms related to compression of adjacent structures such as facial pain and aesthetic deformities cause by the locally gradual expansion.4-6

In this article, we describe the case of maxillary sinus pneumocele presented with both right facial pain and aesthetic deformity with literature reviews focusing on clinical symptoms and surgical approach.

Case Report
A 23-year-old man presented with right facial pain that had not improved by medication for 2 years. The pain was aggravated after nose blowing or flight. He also complained of the change in right facial contours that did not become worse after 2 years ago. The patient denied any history of sinusitis, facial trauma, or prior nasal surgery. On physical examination, there was a hard, nontender, and 2 × 2 cm–sized bulging on the right maxillary area without overlying skin changes (Figure 1A and B). Nasal endoscopy showed no abnormal findings. Facial computed tomography (CT) images revealed the marked expansion of the right maxillary sinus with anteromedial displacement and thinning of the bony wall, but there was no defect (Figure 2A-C).

Based on abovementioned findings, we clinically diagnosed as maxillary sinus pneumocele and decided to perform pneumocelectomy for decompression using Caldwell-Luc approach under general anesthesia. The operative findings were that the bony wall of right maxillary sinus showed the anteromedial protrusion compared to the left side and the overlying mucosa appeared normal after removal of the protruded bony wall (Figure 2D and E). We used titanium mesh to reconstruct the anterior wall of right maxillary sinus (Figure 2F). The patient discharged without postoperative complications. After 2

1 Department of Otorhinolaryngology—Head and Neck Surgery, Veterans Health Service Medical Center, Seoul, Korea
2 Department of Otorhinolaryngology—Head and Neck Surgery, Chung-Ang University College of Medicine, Seoul, Korea

Received: April 1, 2019; accepted: April 22, 2019

Corresponding Author:
Kyung Soo Kim, MD, PhD, Department of Otorhinolaryngology—Head and Neck Surgery, Chung-Ang University College of Medicine, 224-1, Heukseok-dong, Dongjak-gu, Seoul, Korea.
Email: entkks@cau.ac.kr
weeks, facial pain subsided completely and the preoperative facial asymmetry was largely improved (Figure 1C and D). There is no evidence of recurrence during a follow-up 12 months.

**Discussion**

Diseases causing pathological expansion of the paranasal sinuses containing only air include hypersinus, pneumosinus dilatans, and pneumocele, but terms are often used interchangeably. Hypersinus refers to air-filled sinus so that the size of the sinus does not compress the surrounding structures without presenting symptoms. Pneumosinus dilatans denotes expanded sinus beyond the normal boundaries, but the sinus wall thickness is normal without any defect. Pneumocele is similar to pneumosinus dilatans but characterized by the enlarged sinus in which the wall thickness is not uniform and partly thinned with loss of integrity. According to this definition, pneumocele is most suitable for our case in that the bony wall of maxillary sinus lined with normal mucosa is thinned, but not eroded.

Based on previously reported cases, pneumocele of the frontal and sphenoid sinus is frequently described, but only several cases have been reported in the maxillary sinus where the anteromedial portion is often involved. It is thought that the anteromedial wall in the maxillary sinus is the thinnest and most affected when changes in barometric pressure occur. Pneumatization is a physiologic process that occurs in all paranasal sinuses during the growth period. However, pneumatization of the paranasal sinuses shows broad individual differences

**Figure 1.** Preoperative external photographs show a 2 × 2-cm sized protrusion on right maxilla area (A and B). Clinical photographs 6 months after surgery show the improvement of facial contour compared to preoperative state (C and D).

**Figure 2.** Facial CT scan shows enlarged right maxillary sinus (arrows) expanding anteromedially with thinned bony wall; axial view (A), coronal view (B) sagittal view (C). Operative findings show protruded anterior maxillary wall before the osteotomy by Caldwell-Luc approach (D). After the removal of bony wall (E), the defect was reconstructed with titanium mesh (F; asterisk: infraorbital bundle, arrow head: mini-screw). CT indicates computed tomography.
in dimension not only in dependence on the patient's age. The pathogenesis of pneumocele remains unclear, but the presence of a 1-way valve is known as the most convincing mechanism. Nasal polyps, redundant mucosa, inflammation and anatomic variation which caused obstruction of the natural orifice act as a 1-way valve. This allows air to enter the sinus with cough or nose blowing (chronic air trapping) but prevents subsequent pressure equalization and produces air trapping (intrasinus pressure disequilibrium). The increased intrasinus pressure leads to chronic expansion of the sinus wall (hyperpneumatization). In our patient, there was no evidence of obstructive lesion around the maxillary sinus ostium at first visit, but considering that the patient complained of changing facial contour 2 years ago, pneumocele might be occur when the 1-way valve of the maxillary sinus ostium was present.

We reviewed 11 maxillary sinus pneumocele cases that selected patients of 11 retrieved articles from PubMed (Table 1). The mean age was 29.1 years, ranging from 9 years to 62 years with a female to male ratio of 6:5. All 11 patients had unilateral lesion with right side preponderance; the involved side was on the right side in 9 patients and on the left side in 2 patients.

### Table 1. Reported 11 Cases of Pneumocele of the Maxillary Sinus.

| Author and Year | Case Number (Publication Year) | Age, Years | Sex | Side | Aggravating Factors | Simple X-Ray Findings | CT Findings | Treatment | Recurrence (F/U) |
|-----------------|---------------------------------|------------|-----|------|--------------------|-----------------------|-------------|-----------|------------------|
| Braveman5       | 14 F                            | Right      | Traveled by airplane | ND               | Extremely large sinus, medial wall reached the nasal septum, caused an obstruction of nasal passage | Endoscopic surgery | None (12 months) |
| Dillard and Sillers7 | 26 M                           | Right      | ND  | ND   | Extensive pneumatization with medial displacement of medial wall | Endoscopic surgery | ND |
| Flanary and Flanary6 | 9 M                         | Left       | ND  | ND   | Enlarged sinus with erosion of bony orbital floor and lamina papyracea | Endoscopic surgery | None (18 months) |
| Knapp and Klenzner4 | 49 F                        | Right      | Trip to higher altitudes | ND               | Marked enlargement of sinus with thinning of sinus walls | Microscopic endonasal infundibulotomy | ND |
| Meyers and Burtschi8 | 25 M                        | Right      | ND  | Hyperlucent, expanded sinus with thinned bony walls, bony dehiscence (+), displaced medial wall | ND | C-L op with nasoantral window | None (ND) |
| Morrison et al9 | 45 M                           | Left       | Traveled by airplane | Extremely large sinus, expanding in every direction (medial expansion is the most), erosion (+) | C-L op with nasoantral window | None (4 months) |
| Noyek and Zizmor3 | 35 M                           | Right      | Flight, sneezing | Pneumatized sinus, bone destruction, erosion, or dehiscence (+) | ND | C-L op with nasoantral window | None (8 weeks) |
| Omnell and Rohlin10 | 27 F                        | Right      | ND  | Expansion of inferomedial part with medial displacement of medial wall | ND | Wait and see | None (9 years) |
| Vines et al11 | 62 F                           | Right      | ND  | Markedly hyperlucent sinus with generalized expansion of sinus borders, erosion (+) | ND | C-L op with nasoantral window | None (3 months) |
| Wolfensberger and Herrmann1 | 15 M                     | Right      | Nose blowing | Hyperlucent, expanded sinus with thinned bony walls | Hyperlucent, expanded sinus with thinned bony walls | Canine fossa approach with nasoantral window | None (2 years) |
| Zizmor et al12 | 13 F                           | Right      | NA with flights or nose blowing | Hyperlucent, expanded sinus with thinned bony walls | ND | C-L op with nasoantral window | ND |

Abbreviations: CT, computed tomography; C-L, Caldwell-Luc; F, female; F/U, follow-up; M, male; NA, not associated; ND, not described.
Table 2. Descriptive of Patients With Maxillary Sinus Pneumocele of Current Literature Review.

| Total Number | N = 11 |
|--------------|--------|
| Sex          | M:F = 6:5 |
| Mean age, years (range) | 29.1 (9-62) |

**Symptoms**

- Facial symptoms
  - Pressure/numbness | 4 |
  - Pain (cheek, dental), swelling/bulging | 3 (each) |
- Eye symptoms
  - Exophthalmos | 3 |
  - Increased scleral show | 2 |
  - Laze eye, ptosis, propotis; eyeball upward displacement; increased lacrimation | 1 (each) |
- Nasal obstruction | 4 |
- Others
  - Headache, ear pain, ear fullness, hearing loss | 1 (each) |

**Treatment**

- Endonasal approach
  - Endoscopic surgery | 3 |
  - Microscopic endonasal infundibulotomy | 1 |
- External approach
  - C-L Op + Nasoantral window | 5 |
  - Canine fossa approach + nasoantral window | 1 |
  - Wait and see | 1 |

Abbreviations: C-L Op: Caldwell-Luc operation; F, female; M, male.

Maxillary sinus pneumocele may be asymptomatic but cause a variety of symptoms described in Table 2. It may usually be presented as facial symptoms including slowly changing facial contours over long periods and dull maxillary cheek pain or pressure on the affected area. Other associated symptoms are included in eye symptoms such as exophthalmos and increased scleral show and nasal symptoms, especially unilateral nasal obstruction owing to the displacement of neighboring structures. Maxillary cheek pain or pressure may be caused by irritation of the maxillary division of trigeminal nerve (trigeminal irritation) due to the mass effect or direct pressure on the nerve. The pain is exacerbated with sudden changes in atmospheric pressure, such as in trips to the mountains, flying, diving, or when blowing the nose by the mechanism described earlier.

Although the only early symptom of maxillary sinus pneumocele is pain accentuated by nose blowing, whether increased antral pressure after nose blowing is indeed responsible for the expansion of the maxillary sinus may still be open to debate. The diagnosis should be clear with CT scans after the associated symptoms have presented. Computed tomography scans show a hyperpneumatization of the sinus with bony thinning. Nasal endoscopy shows normal mucosa without secretions. Pneumocele requires differential diagnosis with mucocele and benign or malignant neoplasms.

In conclusion, we report an interesting case of maxillary sinus pneumocele that presented with aesthetic deformity and completely treated with Caldwell-Luc approach. Also, based on our comprehensive review of maxillary sinus pneumocele, we found 2 important characteristics. First, maxillary sinus pneumocele may be asymptomatic but cause various symptoms owing to the displacement of neighboring structures, such as facial symptoms, eye symptoms, and nasal obstruction. Second, there is no standard operation technique for maxillary sinus pneumocele, but surgical approach should be individualized depending on patient’s symptoms and needs. Therefore, more case studies are needed to confirm this.

**Authors’ Note**

Written consent was obtained for publication.

**Declaration of Conflicting Interests**

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

**Funding**

The author(s) received no financial support for the research, authorship, and/or publication of this article.

**ORCID iD**

Hyun Sang Cho, MD [https://orcid.org/0000-0003-3411-544X](https://orcid.org/0000-0003-3411-544X)

Kyung Soo Kim, MD, PhD [https://orcid.org/0000-0003-2637-0555](https://orcid.org/0000-0003-2637-0555)
References

1. Wolfensberger M, Herrmann P. The pathogenesis of maxillary sinus pneumoceles. Arch Otolaryngol Head Neck Surg. 1987;113(2):184-186.
2. Jarvis JF. Pneumocele of the frontal and sphenoid sinuses. J Laryngol Otol. 1974;88(8):785-793.
3. Noyek AM, Zizmor J. Pneumocele of the maxillary sinus. Arch Otolaryngol. 1974;100(2):155-156.
4. Knapp FB, Klenzner T. Pneumocele as a rare differential diagnosis in trigeminal irritation. Am J Otolaryngol. 2003;24(4):236-238.
5. Braveman I. Pneumocele of the maxillary sinus with orbital and trigeminal nerve involvement: case report and review of the literature. J Otolaryngol Head Neck Surg. 2009;38(2):E35-38.
6. Flanary CJ, Flanary VA. Maxillary sinus pneumocele. Otolaryngol Head Neck Surg. 1998;119(5):518-520.
7. Dillard ML, Sillers MJ. Maxillary sinus pneumocele causing orbital displacement. Am J Otolaryngol. 1999;20(4):250-251.
8. Meyers AD, Burtschi T. Pneumocele of the maxillary sinus. J Otolaryngol. 1980;9(4):361-363.
9. Morrison MD, Tchang SP, Maber BR. Pneumocele of the maxillary sinus. Report of a case. Arch Otolaryngol. 1976;102(5):306-307.
10. Omnell KA, Rohlin M. Pneumocele of the maxillary sinus: a case report and literature review. Dentomaxillofac Radiol. 1994;23(3):172-175.
11. Vines FS, Bonstelle CT, Floyd HL. Proptosis secondary to pneumocele of the maxillary sinus. Neuroradiology. 1976;11(2):57-59.
12. Zizmor J, Bryce M, Schaffer SL, Noyek AM. Pneumocele of the maxillary sinus. A second case report. Arch Otolaryngol. 1975;101(6):387-388.
13. Adams WM, Jones RI, Chavda SI, Pahor AL, Taifa KT. Pneumosinus dilatans: a discussion of four cases and the possible aetiology. Rhinology. 1998;36(1):40-42.