

# Maxillary Air Cyst

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## ABSTRACT

Pathologic dilatation of the maxillary sinus by air is a rare condition with unclear etiology. We present a case of a 17 year old male with a maxillary air cyst diagnosed by computed tomography. The CT demonstrated air-filled expansion of the maxillary sinus beyond the normal anatomical limits with associated cortical bone thinning. The case report highlights the pathognomonic computed tomography findings of this rare entity and discusses the perplexing nomenclature, proposed etiologies and various treatment options.

## CASE REPORT

### CASE REPORT

A 17-year-old previously healthy male presented to a tertiary rhinology clinic with an eight-month history of progressively worsening left-sided nasal obstruction and facial pressure, having failed appropriate antibiotic therapy for presumed acute rhinosinusitis. Facial asymmetry developed without diplopia or sensory deficits. On examination, the patient demonstrated mild left facial protrusion and upward deviation of the left eye without proptosis or enophthalmos (Figure 1). Cranial nerve exam was normal with no evidence of ophthalmoplegia or visual changes. Rigid nasal endoscopy demonstrated a narrowed left nasal cavity with a medialized middle turbinate and no purulent or polypoid change.

Sinus computed tomography (CT) was performed which demonstrated significant left maxillary sinus expansion with cortical bone thinning anteriorly, medially and superiorly (Figure 2). The sinus protruded into the left nasal cavity, through the anterior left maxillary wall and compressed the inferior orbital wall. The sinus was completely air-filled with no mass or mucosal thickening. Surrounding soft tissues, including the orbits and other sinuses, were normal.

The patient was diagnosed with a symptomatic and cosmetically deforming left maxillary air cyst, which required surgical intervention. Prior to the surgery, the patient was instructed to avoid dental procedures or contact sports and was prescribed intranasal corticosteroids to decrease inflammation.

The patient successfully underwent a left endoscopic limited medial maxillectomy with an elevated opening pressure of 24mmHg above atmospheric, as documented by an arterial line setup. The maxillary sinus appeared dilated on direct visualization with thinning of the orbital floor, and there was no inflammation or mass (Figure 3). In short term post-operative follow up, the patient had subjective improvement in symptoms, now being able to breathe through his nose, and very subtle improvement in the left facial protrusion. Endoscopic assessment showed a well healed ethmoid and maxillary cavity.

### DISCUSSION

Pathological expansion of paranasal sinuses with air is a rare condition, with only 36 prior reported cases involving the maxillary sinus in the English literature [1]. Radiologists must recognize this condition as the diagnosis is made radiographically and the appearance is pathognomonic.

The nomenclature surrounding sinus air cysts remains controversial. Most recent authors follow the strict modern definitions of Urken et al to separate pathological aeration of the sinuses into hypersinus, pneumosinus dilatation, and pneumocele [2]. Hypersinus is an asymptomatic expansion of the sinus beyond the normal anatomical limits, while pneumosinus dilatation denotes an expansion surrounded by

normal cortical bone thickness resulting in facial symptoms or cosmetic deformities. A pneumocoele refers to expansion with thinning of the overlying bone (focally or diffusely) with local symptoms and/or cosmetic deformity. According to this definition, the above case falls into the category of a pneumocoele. Most experts agree that the three conditions represent a continuum with considerable overlap. However, the original definition put forth by Benjamins in 1918 is still used by some; it describes pneumosinus dilatans as a slow forming dilatation involving the whole sinus with the sinus wall intact [3,4]. Benjamins used six criteria to differentiate pneumosinus dilatans from a pneumocoele. In pneumosinus dilatans, in contrast to a pneumocoele, there is no air outside the bony cavity of the sinus, no change with pressure, the bones are intact, the expansion is slow in development, the large sinus cavity contains air only, and no air is in the soft tissues. Given the varying definitions and descriptions, pneumosinus dilatans and pneumocoele are often mistakenly used interchangeably in the literature. It has been proposed to use the umbrella term air cyst to describe the entire spectrum of these lesions as the precise definition does not alter management [1]. Management remains dictated by symptomatology.

Causes of maxillary sinus dilatation can be divided into benign and malignant tumors, which are common, and air trapping or mucous trapping, which are rare [5]. Any patient presenting with pain and facial asymmetry should be assessed by CT to make the diagnosis of air cyst and, importantly, to rule out an underlying tumor. If an isolated air cyst is demonstrated on CT, no further imaging is required; if a tumor is identified, magnetic resonance imaging may be helpful for further characterization. Benign tumors arising in the sinus, such as ameloblastoma, myxoma and pseudotumor, demonstrate a central mass surrounded by the expanded sinus. If arising from the bony cortex, they will often leave a peripheral rim of air. Malignant tumors, such as primary and secondary squamous cell tumors, are soft tissue masses which expand to fill the sinus. They are identified by their extensive bony damage causing an irregular sinus and loss of normal architecture, although this may also uncommonly be seen with benign tumors. Mucocoeles on non-contrast CT demonstrates the presence of a mucous containing homogeneous mass isodense to brain parenchyma with no infiltration of adjacent structures, but associated osteolysis of the sinus structures [6]. Contrast CT of a mucocoele demonstrates no variation in the density of the homogeneous mass and a regular hyperdense rim on the orbital and intracranial side. Conversely, air trapping is identified by the presence of a dilated sinus with no mass. Common anatomical variants surrounding the maxillary sinus may also be diagnosed by CT. These include the pneumatization of the middle turbinate (concha bullosa), or the presence of extra mural ethmoidal air cells within the inferior orbital wall, also named maxilloethmoidal cells or Haller cells [7].

A recent review by Teh et al. identified 36 cases of maxillary sinus expansion secondary to air [1]. The average age of presentation of the patients was 27 (range 9-62 years) with a male to female ratio of 1.8:1. Patients with pneumosinus dilatans or pneumocoele often present with symptoms of nasal

obstruction, facial deformities, and worsening visual acuity [8]. Patients may experience diplopia and proptosis if the dilatation is severe enough to impinge the orbital contents.

The etiology of pathologic dilatation of a sinus with air is poorly understood with multiple proposed hypotheses. The most popular etiology is a one-way valve mechanism involving the sinus ostium [9]. Positive pressure events, such as nose blowing, cause air trapping within the affected sinus due to the one-way valve mechanism. Elevated opening pressure in the maxillary sinus, as documented in this patient (24mmHg while normal is near zero), support the one-way valve hypothesis [10]. Other hypotheses not strongly supported by evidence include spontaneous drainage of a mucocoele leaving an air filled cavity, the presence of a gas-forming organism within the sinus although no organism has yet to be isolated, congenital abnormalities or endocrine disorders.

Asymptomatic air cysts are not treated, but surgical management is indicated for symptomatic or deforming air cysts. Multiple treatment modalities exist with goals of correcting the cosmetic deformity and establishing an equal pressure system between the sinus and the nasal cavity. A middle meatus antrostomy achieves equal pressure with little morbidity [8]. A treatment algorithm proposed by Choi et al assesses the antral wall thickness and extent of anterior wall expansion to decide between three surgical procedures for cosmesis [11]. With normal thickness, the easiest way to decrease maxillary convexity is with electrical burring. With thinning but no expansion, antral wall turnover is used by performing surrounding osteotomies and repositioning the bony fragment inside out. The presence of wall thinning and expansion was treated with greenstick downward fractures. All three methods give satisfactory cosmetic result. The prognosis for an air cyst is excellent, with resolution of symptoms and no sequelae following surgery. Bony remodelling with resolution of the cosmetic deformity may be accomplished via limited medial maxillectomy, but cosmetic improvement for significant deformities may require more invasive surgery as described above [8].

#### TEACHING POINT

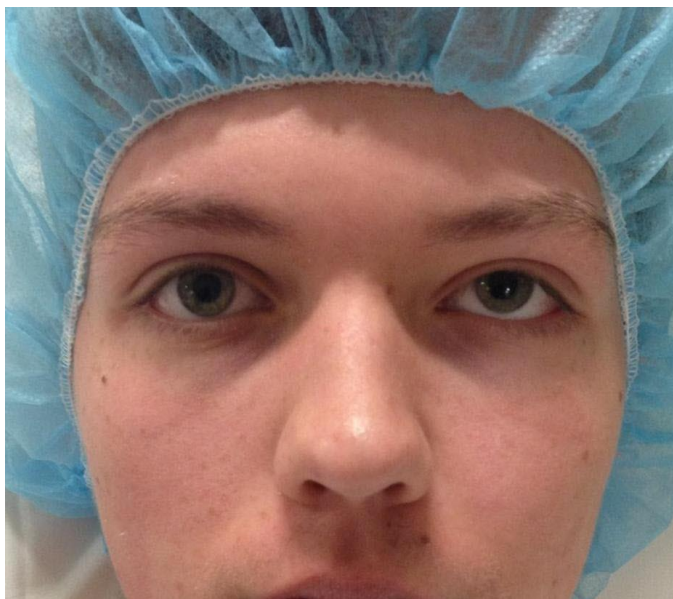
Pathological expansion of paranasal sinuses is a rare diagnosis made radiographically given the pathognomonic CT appearance. The expected findings on CT are an air-filled expansion of a sinus beyond the normal anatomical limits with or without associated cortical bone thinning. Although nomenclature of this condition is complicated, a simplified approach of naming all the lesions as air cysts is preferred because management is dictated by symptomatology.

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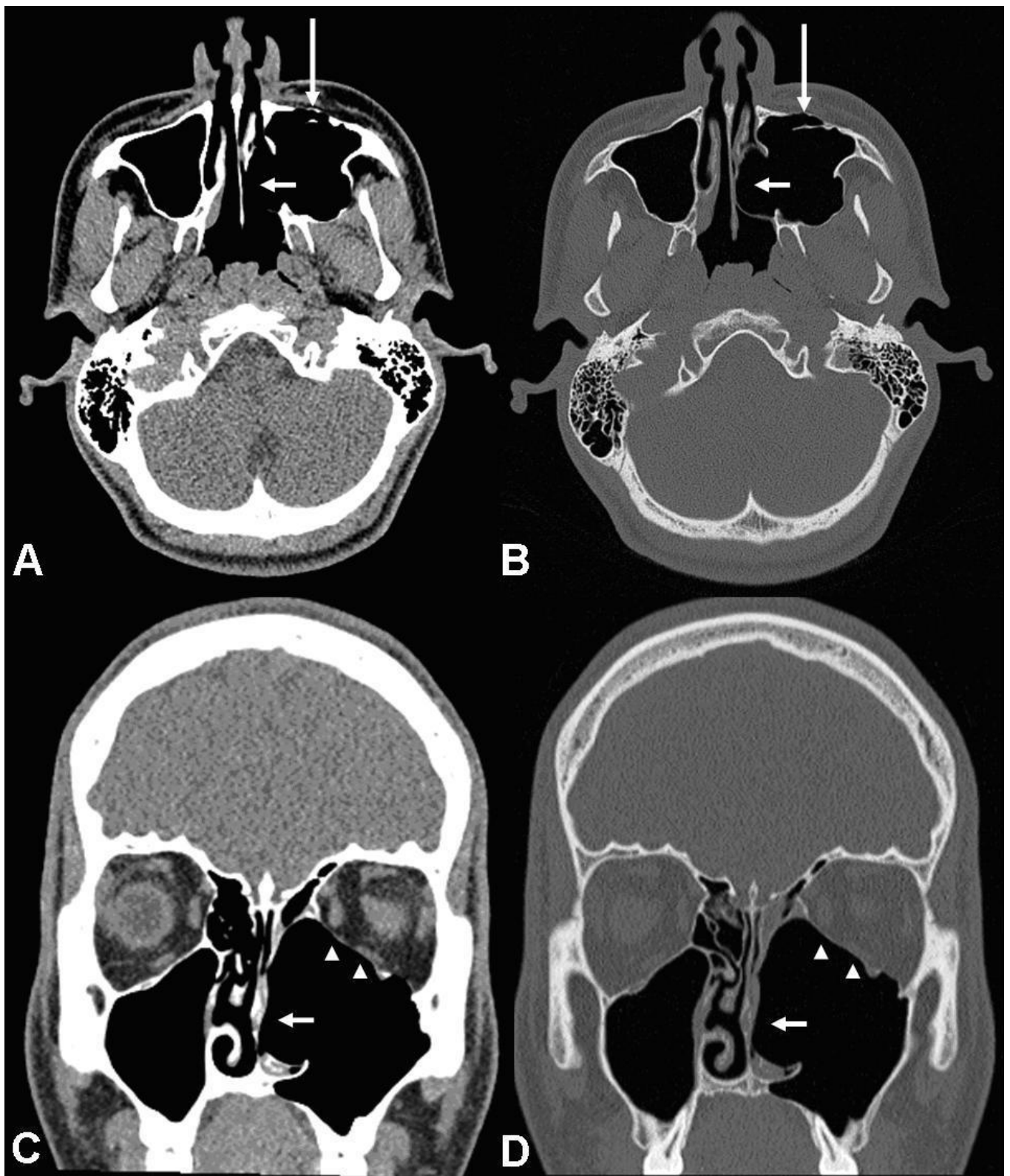
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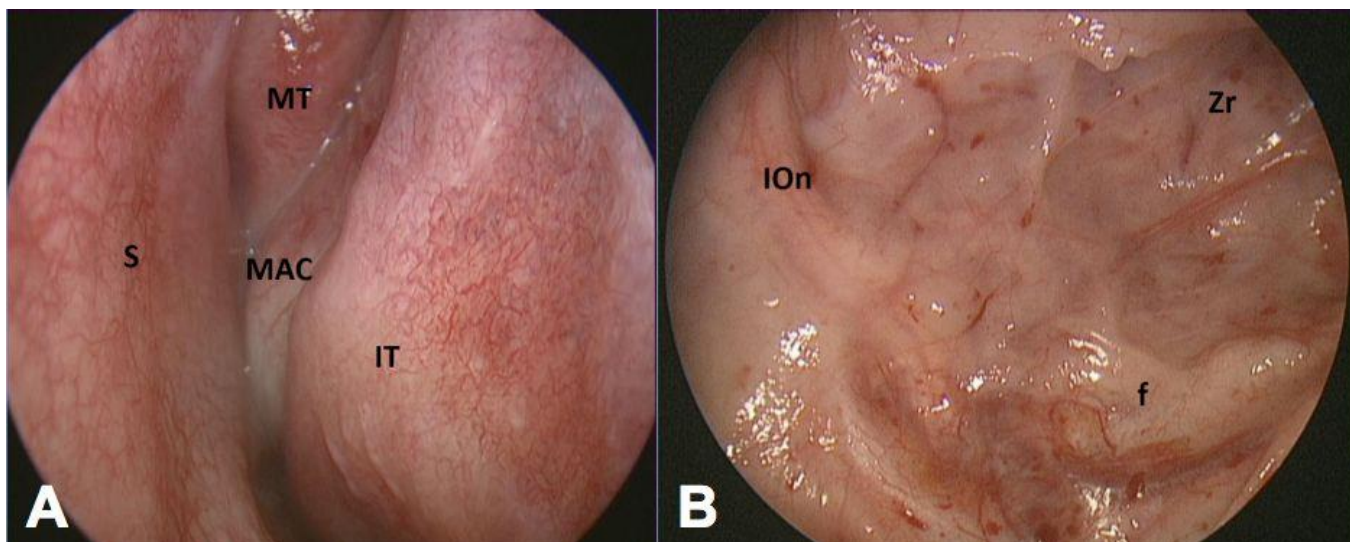
#### FIGURES



**Figure 1 (left):** Pre-operative picture of the 17 year old male diagnosed with a left maxillary air cyst demonstrating slight left facial protusion and slight upward deviation of the left eye.



**Figure 2:** 17 year old male diagnosed with maxillary air cyst. Axial and coronal reconstructions of a non-contrast CT of the sinuses in soft tissue (A&C) and bone (B&D) windows demonstrating dilatation of the left maxillary sinus with protrusion and cortical bone thinning into the left nasal cavity (short arrows), expansion through the anterior maxillary wall (long arrow), and expansion into the inferior orbital wall (arrowheads). No associated mass, mucosal thickening or involvement of surrounding soft tissues is present. (Siemens Somatom Definition AS, 60 mAs, 120 kV, 1 mm slice thickness, non-contrast).



**Figure 3:** Endoscopic images during limited medial maxillectomy of maxillary air cyst on a 17 year old male. A: Endoscopic view of left nasal cavity demonstrating maxillary air cyst between the middle and inferior turbinate. B: Lateral endoscopic view of maxillary air cyst with no inflammation or mass visible. S: septum, MT: middle turbinate, IT: inferior turbinate, MAC: maxillary air cyst, IO n: infraorbital nerve, f: floor of maxillary sinus, Zr: zygomatic recess.

<b>Etiology</b>	Many etiologies proposed, with the most convincing evidence supporting a one-way valve mechanism.
<b>Incidence</b>	Rare; 36 cases reported in the English literature.
<b>Gender ratio</b>	Approximately male to female ratio 1.8:1.
<b>Age predilection</b>	Average age 20 to 40, very rare in childhood.
<b>Risk factors</b>	Unknown.
<b>Treatment</b>	Simplest method to relieve elevated sinus pressures is by performing an endoscopic maxillary antrostomy. Electrical burring, antral wall turnover and greenstick downward fractures have all been described with equal cosmetic success.
<b>Prognosis</b>	Excellent.
<b>Findings on imaging</b>	CT demonstrates air-filled dilation of the maxillary sinus beyond the normal anatomical limits

**Table 1:** Summary table of maxillary air cyst

	<b>CT findings</b>
<b>Hypersinus</b>	Asymptomatic air-filled expansion of a sinus beyond the normal anatomical limits with normal cortical thickness.
<b>Pneumosinus dilatans</b>	Symptomatic/cosmetic air-filled expansion of a sinus beyond the normal anatomical limits with normal cortical thickness.
<b>Pneumocoele</b>	Symptomatic/cosmetic air-filled expansion of a sinus beyond the normal anatomical limits with cortical wall thinning.
<b>Mucocoele</b>	Mucous containing homogeneous mass isodense to brain parenchyma with associated osteolysis of the sinus structures but no infiltration of adjacent structures.
<b>Benign tumor</b>	Central mass surrounded by the expanded sinus; possible peripheral rim of air if arising from the bone.
<b>Malignant tumor</b>	Irregular soft tissue mass rapidly expanding to fill the sinus causing extensive bony damage and loss of normal sinus architecture.
<b>Concha bullosa</b>	Common anatomical variant represented by pneumatization of the middle turbinate.
<b>Haller cell</b>	Common anatomical variant composed of extra mural ethmoidal air cells within the inferior orbital wall.

**Table 2:** Differential diagnosis table of a dilated maxillary sinus

**ABBREVIATIONS**

CT: Computed Tomography

**KEYWORDS**

Air cyst; maxillary sinus dilatation; pneumocoele

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