

# Obstructive Mucocele of the Maxillary Sinus in an 18 Year Old Cambodian Female

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## Abstract

**Objective:** Sinus mucoceles are true cysts that develop secondary to obstruction of sinus ostia and can be associated with significant morbidity. Study Design. We present a case of obstructive-type sinus mucocele of the maxillary sinus, including clinical, radiographic, histologic findings, as well as treatment.

**Results:** An 18 year old female presented for evaluation and management of maxillary sinus swelling. A diagnosis of sinus mucocele was rendered after clinical, radiographic, surgical and histopathologic examinations. Recurrence developed within four months of the initial surgical intervention.

**Conclusions:** Sinus mucoceles may be associated with significant morbidity and are currently managed most often by endoscopic nasal surgery.

## Introduction

Sinus mucoceles are true cysts lined by pseudostratified respiratory epithelium and have a mucous or mucopurulent content [1]. They are produced by the accumulation of mucous secretions within tissue or the sinus cavity, secondary to obstruction of the sinus ostia (obstructive type). By definition, if there is partial obstruction of ducts of seromucous glands within the sinus wall, such true cysts are called retention cysts of the maxillary sinus. While retention cysts are typically asymptomatic and incidental findings, the obstructive type sinus mucocele is capable of significant expansion and destruction [2]. Chemical mediators, such as prostaglandin E2 and collagenase, are released at the capsule of the mucocele, possibly causing bony destruction and allowing the mucocele to expand into adjacent structures [3]. They can lead to exophthalmos, limited eye movement and headaches in patients. Rarely, ethmoid sinus involvement may induce oculomotor nerve paralysis. The frontal sinus is most commonly affected, whereas involvement of sphenoid, ethmoid, and maxillary sinuses are unusual sites of involvement [4]. We present a case of obstructive type sinus mucocele of the left maxillary sinus in an 18 year old female.

## Case Report

An 18 year old Cambodian female presented to Children's Surgical Center at the National Rehabilitation Center in Phnom Penh, Cambodia for evaluation of left maxillary sinus swelling and discomfort (Figure 1). In addition to notable facial asymmetry, the enlargement was associated with elevation of the left orbit. No visual disturbances, however, were noted. A computerized tomography (CT) scan without contrast was ordered and showed a cystic process of the left maxillary sinus (Figure 2) and treatment was planned for February 2014. An 18-gauge needle was chosen for aspiration through an existing bony defect apical to teeth 12-14, returning a mucoid material. Subsequent surgical access was gained in the same area and, upon entry into the sinus, the membrane appeared normal. The lumen of the sinus was entirely filled with a gray, viscous, mucoid material. This, along with a portion of the sinus lining, was removed and submitted for histopathologic examination. An antrostomy of the sinus was completed prior to closure. Recovery and healing were uneventful.

Histologic examination of the submitted specimen showed fragments of fibrous and fibromyxoid stroma with small blood vessels and foci of chronic inflammation. Foamy histiocytes were seen in isolated portions of the specimen. While the majority of the specimen was rather paucicellular, foci of plump, angular and spindle-shaped cells were seen. Immunohistochemical stains for S100 protein were negative. The histologic findings, in combination with those of the CT imaging and surgical exploration, were consistent with a sinus mucocele, obstructive type. Within four months of surgery, the patient returned for follow-up with lesion recurrence, necessitating further intervention. A second surgical procedure was completed and follow-up is ongoing.

## Discussion

Langenbeck first described mucoceles as hydatids in 1820. Later, in 1909, Rollet coined the term mucocele. They are found most often in the frontal sinus, with those of the maxillary sinus generally considered rare [5,6]. While mucoceles are benign processes, they behave like space occupying lesions, causing erosion of surrounding bone and displacement of adjacent structures. Their proximity to vital structures can be a cause of significant morbidity if left untreated [4]. Obstructive type sinus mucoceles occur secondary to the obstruction of the natural orifices and are most often associated with chronic inflammation, allergy, surgery or trauma. They distend the bony walls of the sinuses as the intraluminal pressure increases and eventually erode the bone by

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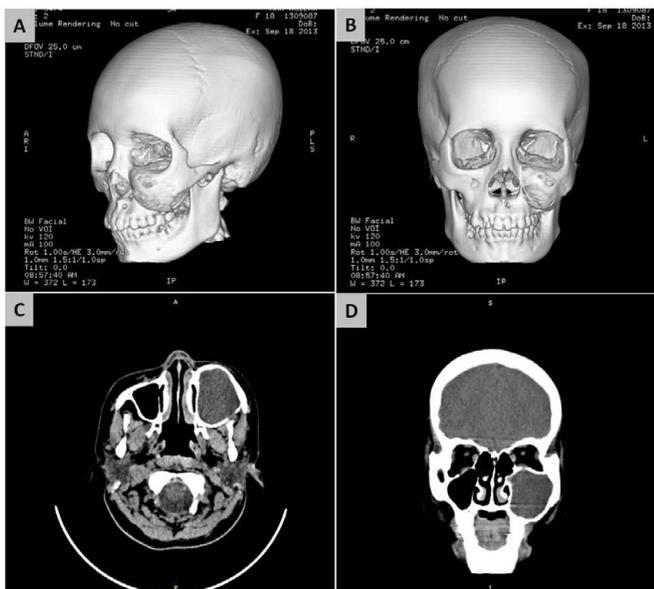
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**Figure 1:** Clinical image showing swelling of the left maxillary sinus region.



**Figure 2:** A: Three dimensional computed tomography demonstrating expansive lesion of the left maxillary sinus – lateral view; B: Frontal view; C: Computed tomography showing homogenous, expansile view of the left maxillary sinus – axial view; D: Coronal view.

pressure resorption [6]. Occasionally, bone dyscrasias, such as fibrous dysplasia or osteopetrosis, can cause obstruction of the ostia and lead to sinus mucoceles.

Typically, sinus mucoceles are diagnosed in adults during the 3<sup>rd</sup> and 4<sup>th</sup> decades. However, patients as young as 4 have been reported [1]. There is a slight male predilection [7]. Clinical symptoms may include headache, diplopia, visual impairment, nasal obstruction, and a variety of other symptoms related to involvement of the various cranial nerves and the pituitary gland, depending upon the location of the individual mucocele. Primary antral mucoceles usually occur as swellings of the cheek or mucobuccal fold, with displacement of the dentition and occasional nasal obstruction or exophthalmos. Poorly localized pain or tenderness often accompanies the swelling [6].

While diagnosis is often by rhinoscopy, CT scans of the paranasal sinuses or magnetic resonance imaging (MRI) with gadolinium contrast

may be helpful [8]. MRI may be particularly useful in differentiating mucoceles from neoplastic processes [4]. CT scans of maxillary sinus lesions typically show a uniformly expansile mass within the antrum and thinning of the bone, mostly of the anterior wall [9].

Infection of sinus mucoceles (mucopyocele) with pyogenic bacteria may result in the accumulation of pus within the lumen and increased morbidity. The most frequent organisms include staph aureus, alpha-hemolytic streptococci (six isolates), *Hemophilus* spp. (five isolates), and Gram-negative bacilli (six isolates). The predominant anaerobes are *Peptostreptococcus* sp., *Prevotella* sp., *Fusobacterium* sp., and *Propionibacterium acnes*.

On microscopic examination, obstructive type mucoceles within paranasal sinuses are lined by pseudostratified ciliated columnar epithelium. Squamous metaplasia and reactive bone formation can appear in areas adjacent to cystic epithelium. Occasionally, there is activation of T<sub>h2</sub> lymphocytes due to increased expression of IL-12 and IL-2 which can lead to an increased inflammatory response [7].

The treatment can be either radical or conservative. Conservative surgical care involves marsupialization of the mucocele with maintenance of adequate sinus drainage, which relieves the symptoms and prevents recurrence. This is now done most often by endoscopy using medial maxillectomy with mucosal flap [7,10,11]. With Endonasal sinus surgery, Bockmuhl and colleagues [5] reported that 98.4% of 185 patients were long-term disease free. Previously, mucoceles have been removed through external approaches, such as a Lynch-Howarth incision, Caldwell-Luc approach, or an osteoplastic flap. Surgical excision is the treatment of choice and early intervention is indicated to prevent visual compromise or the development of a mucopyocele.

## Conclusion

Sinus mucoceles are characterized by the potential for significant morbidity when not treated promptly or adequately. We have presented a case involving the maxillary sinus in an 18 year old female, which recurred after the initial surgical procedure.

## Conflict of interest

Authors have no conflict of interests to declare

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