

Bilateral Chronic Maxillary Atelectasis with a Unilateral Accessory Ostium

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Abstract

Chronic maxillary atelectasis (CMA) is an underdiagnosed condition that can occur bilaterally and may lead to significant complications. An accessory maxillary ostium (AMO) is a defect in the posterior fontanelle that is associated with maxillary sinus pathology. We present a case of a 47-year-old man with sinonasal symptoms. Nasendoscopy demonstrated bilateral lateralized uncinate processes. Radiological findings were consistent with a diagnosis of bilateral stage II CMA, with the left side less atelectatic than the right and only partially opacified. The patient was treated surgically with bilateral uncinectomy and maxillary antrostomy. Intraoperatively, an AMO was visualized on the left. The main theory regarding the etiology of CMA is sustained obstruction of the ostiomeatal complex resulting in negative intra-sinus pressures and subsequent atelectatic remodeling of the antral walls. Our patient had an AMO on the left side, which was also the side of reduced severity of both symptoms and radiographic findings. We propose that the patient initially had bilateral CMA, with subsequent development of a left AMO, which halted further progression of the disease on this side. This case suggests that an AMO can develop or enlarge and supports the notion that CMA is caused by negative intra-sinus pressures. Further research is required to establish the detailed etiology of CMA.

Keywords: Maxillary sinus, maxillary sinusitis, paranasal sinuses, sinusitis

INTRODUCTION

Chronic maxillary atelectasis (CMA) is an underdiagnosed, acquired condition of persistent and progressive reduction in maxillary sinus volume that results in antral wall collapse. The first known report of CMA in the literature was in 1964, when Montgomery described a mucocele-related opacification of a maxillary sinus associated with orbital floor collapse and enophthalmos (1). The diagnosis of CMA can be made when the following criteria are fulfilled: (1) sinus opacification on imaging of at least 3 months' duration and/or the intraoperative finding of tenacious mucus secretions filling the antrum of the maxillary sinus, and (2) lateral displacement of the medial infundibular wall and fontanelle, with or without inward bowing of the osseous walls (2). CMA has been classified into three distinct but progressive stages according to the degree of wall collapse (2).

Active mucociliary transport in the maxillary sinus is directed toward the natural ostium, which typically opens through the anterior portion of the posterior fontanelle into the nasal cavity (3). An accessory maxillary ostium (AMO) is an anatomic variant where there is a dehiscence in the fontanelle, resulting in an additional communication between the maxillary sinus and nasal cavity. It has been suggested that the existence of an AMO may promote the development of maxillary sinusitis. Furthermore, it has not been established whether AMO is a congenital or an acquired structure (3).

We present a case of bilateral CMA in the presence of a unilateral (left) AMO. This case provides support for the notion that CMA is caused by sustained negative pressure and that an AMO, in some instances, may be acquired pathology.

CASE PRESENTATION

A 47-year-old male with a long history of nasal obstruction was referred to us for evaluation. He noted that the nasal obstruction was worse on the right side and was associated with right-sided malar and periorbital facial pain and frontal headaches. The patient reported no other sinonasal symptoms and there was no additional significant past medical history.

Examination revealed no enophthalmos or hypoglobus and no discernible mid-facial deformities. Flexible nasendoscopy revealed a right anterior septal deviation and marked lateralization of both uncinate processes. Computed tomography (CT) of the paranasal sinuses demonstrated a right septal deviation, lateralization of both uncinate process-

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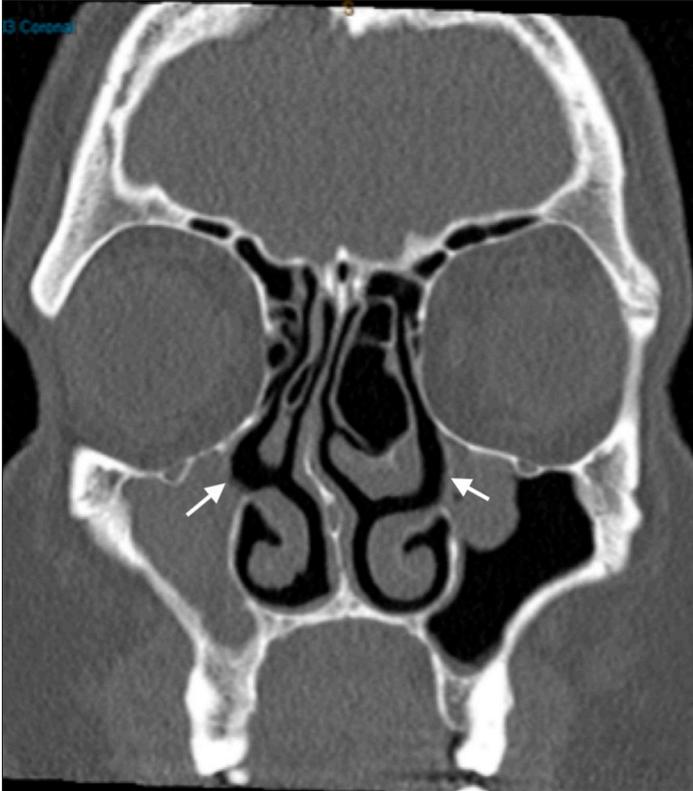


Figure 1. Coronal view of a CT scan of the paranasal sinuses demonstrating septal deviation to the right, bilateral lateralization of uncinate processes, a large left concha bullosa, bowing of the superior osseous walls (right more pronounced than left), opacification of the right maxillary sinus and an early retention cyst in the left maxillary sinus

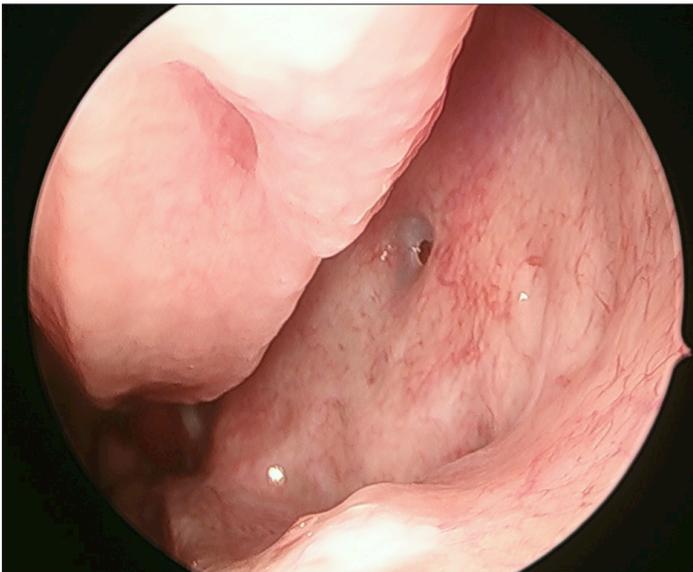


Figure 2. Intraoperative endoscopic view of left middle meatus showing lateralization of uncinate process and the accessory maxillary ostium (AMO)

es, and bowing of the superior and posterolateral osseous walls of both maxillary antra. There was complete opacification of the right maxillary sinus and partial opacification of the left, with a small mucous retention cyst. An accessory maxillary ostium was identified on the left side. Additional findings included a large left-sided concha bullosa and minimal

mucosal thickening in the ethmoid air cells. Findings were consistent with a diagnosis of bilateral stage II CMA (Figure 1).

The patient underwent septoplasty, bilateral inferior turbiniplasty, bilateral uncinectomies, and bilateral maxillary antrostomies. Intraoperatively, a left-sided AMO was identified in the posterior fontanelle (Figure 2) and was incorporated into the antrostomy.

The patient had an uneventful recovery and reported complete resolution of symptoms at six weeks post-operatively.

DISCUSSION

CMA has traditionally been described as a unilateral condition. However, in recent years there have been multiple reports of bilateral pathology; this has challenged the traditional definition of this disorder, and has raised questions with regard to the underlying factors that predispose patients to this condition (4).

The etiology and predisposing factors underlying the development of CMA remain unknown. The theory that is accepted most widely suggests that the inciting factor is sustained obstruction of the ostiomeatal complex. Ongoing mucosal resorption of sinus gas and accumulation of thickened mucus lead to the development of negative pressure within the maxillary sinuses. This process triggers remodeling and inward bowing of the maxillary sinus walls, which can be easily visualized both endoscopically and radiologically (5, 6).

Similarly, the role of an AMO in maxillary sinus pathology remains controversial. Previous studies have demonstrated an association between an AMO and maxillary sinusitis, rhinitis, and the development of mucus retention cysts (3, 7, 8). These features may be associated with mucus recirculation between the nasal cavity and maxillary sinus through both the natural and accessory ostia (3). Additionally, it remains uncertain whether an AMO is an acquired or congenital anomaly. Genc et al. (9) demonstrated that an AMO can develop after experimental induction of rhinogenic sinusitis in rabbit models; this result suggests that an AMO may develop secondary to tissue pathology. The fontanelle of the nasal cavity is an area of the medial wall of the maxillary sinus that is composed of two mucosal layers and no bone. The development of an AMO may be secondary to drainage of maxillary contents (i.e., pus or mucus) into the middle meatus via a perforation in the fontanelle. This would be akin to the perforation of the tympanic membrane in the middle ear following acute otitis media (10).

Our patient had bilateral stage II CMA, as demonstrated by radiographic findings that included inward bowing of one or more of the osseous walls of the maxillary antra. Interestingly, maxillary sinus opacification and symptoms were present only on the right side. Furthermore, the radiographic findings were less pronounced on the side with the AMO. The presence of a congenital, patent AMO may have prevented the initial build-up of negative pressure in the sinus. As such, we would not anticipate the development of a lateralized uncinate process and inward bowing of the maxillary osseous walls as seen on the CT images. Therefore, we postulate that the patient initially developed CMA bilaterally from obstruction of the ostiomeatal complex. The patient subsequently developed a left-sided AMO, or had a pre-existing obstructed or small AMO that became patent, which then halted the progression of the disease on the left side by equalizing the pressure differential. This possibility supports that idea that some AMOs may be acquired anomalies secondary to sinus pathology.

CONCLUSION

CMA is an underdiagnosed condition that can occur bilaterally and cause significant sinonasal symptoms. We discuss the case of a patient who presented with bilateral CMA with a unilateral AMO. The side with the AMO had less severe atelectasis and only partial opacification, suggesting that CMA was halted on this side, following AMO formation. This case suggests that some AMOs may be acquired defects and supports the notion that CMA is caused by negative intra-sinus pressure.

Informed Consent: Written informed consent was obtained from the patient who participated in this case.

Peer-review: Externally peer-reviewed.

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