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Chronic maxillary atelectasis (including silent sinus syndrome) can present bilaterally

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Abstract

Objective. Chronic maxillary atelectasis is a rare and underdiagnosed condition in which there is a persistent and progressive decrease in maxillary sinus volume secondary to inward bowing of the antral walls. Chronic maxillary atelectasis is typically unilateral. Simultaneous bilateral chronic maxillary atelectasis is extremely uncommon.

Methods. A retrospective review was performed of patient data collected by the senior clinician over a three-year period (2015–2018). A comprehensive literature search was conducted to locate all documented cases of chronic maxillary atelectasis in English-language literature. Abstracts and full-text articles were reviewed.

Results. Three patients presented with sinonasal symptoms. Imaging findings were consistent with bilateral chronic maxillary atelectasis. The literature review revealed at least nine other cases of bilateral chronic maxillary atelectasis. Management is typically via endoscopic middle meatus antrostomy.

Conclusion. Chronic maxillary atelectasis was initially defined as a unilateral disorder, but this description has been challenged by reports of bilateral cases. Further investigation is required to determine the aetiology and pathophysiology of the disease.

Clinical Record

Dr J Ho takes responsibility for the integrity of the content of the paper

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Introduction

Chronic maxillary atelectasis is a rare, acquired condition of persistent and progressive reduction in maxillary sinus volume and antral wall collapse. Although chronic maxillary atelectasis has been reported to occur in children, most patients present in the third to fifth decades of life, with no gender predilection.[1,2] Chronic maxillary atelectasis was first reported in 1964 by Montgomery, who described mucocele-related opacification of a maxillary sinus, associated with orbital floor collapse and enophthalmos.[3] Chronic maxillary atelectasis has since been classified into three separate but progressive stages, according to the degree of wall collapse, as described in Table 1.[1]

The nomenclature used in the literature for this spectrum of conditions is often confusing. Previous authors have used other terms, such as silent sinus syndrome, imploding antrum syndrome and acquired maxillary sinus hypoplasia. Soparkar *et al.* first used the term silent sinus syndrome to describe the presentation of asymptomatic unilateral enophthalmos and hypoglobus in association with ipsilateral maxillary sinus hypoplasia.[4] However, it is now acknowledged that silent sinus syndrome is likely a subtype of stage III chronic maxillary atelectasis, with the only difference in defined criteria being the presence or absence of sinus-related symptoms (Table 1).[5] Furthermore, chronic maxillary atelectasis is an acquired defect and should therefore be distinguished from congenital maxillary sinus hypoplasia.[6]

Correlation between symptoms and severity of chronic maxillary atelectasis is poor.[6] While patients with early-stage chronic maxillary atelectasis may present with non-specific sinonasal symptoms of nasal congestion, rhinorrhoea, facial pressure and pain, and post-nasal drip, those with more advanced disease may develop facial and ophthalmological symptoms such as diplopia, enophthalmos, lagophthalmos, hypoglobus and mid-face asymmetry.[2,7] However, gradual changes, especially in those with bilateral disease or concurrent pathologies (such as thyroid ophthalmopathy), may go unnoticed by patients themselves, and are often only recognised by others or incidentally detected on imaging performed for other purposes.[6,8,9]

Furthermore, while the diagnosis may be suspected clinically, it is confirmed radiologically.[10] Pathognomonic computed tomography (CT) findings include: near or total opacification of the affected maxillary sinus; obstruction of the ostiomeatal complex; and inward bowing of the antral walls, with orbital floor displacement.[11] Lateralisation of the uncinate process is seen, resulting in asymmetric enlargement of the middle meatus when compared to the contralateral side. The collapse of the antral walls results in a reduction of maxillary sinus volume and a concurrent enlargement of orbital volume.[10] This causes inferior displacement of the orbital contents, leading to the clinical finding of hypoglobus, enophthalmos and facial asymmetry.

Table 1. Staging of chronic maxillary atelectasis[1,5]

Parameter	Chronic maxillary atelectasis			Silent sinus syndrome
	Stage I	Stage II	Stage III	
Deformity	Membranous	Bony	Clinical	
Characteristic features	Lateralised maxillary fontanelle	Inward bowing of 1+ osseous walls (anterior, superior &/or posterolateral bony walls) of maxillary antrum	Enophthalmos, hypoglobus &/or midfacial deformity	
Enophthalmos?	Absent	Absent	Present	
Sinusitis symptoms or facial pain?	Absent or present	Absent or present	Absent or present	Absent

Chronic maxillary atelectasis has traditionally been described as a unilateral condition.[1,2] Bilateral simultaneous chronic maxillary atelectasis is extremely uncommon, with only a handful of reports in the literature. This paper reviews three cases of bilateral chronic maxillary atelectasis seen in the same institution, between 2015 and 2018, which challenge the traditional unilateral definition of this disease.

Materials and methods

Ethics approval was obtained from the Western Sydney Local Health District Human Research Ethics Committee. A retrospective review was performed of data collected prospectively between January 2015 and April 2018. The data were from patients of the senior author (NPS), a rhinologist at Westmead Hospital (a tertiary institution based in Sydney, Australia). Patients with bilateral chronic maxillary atelectasis (diagnosed on imaging) were identified. A case note review was conducted of these patients using data retrieved from the hospital health information database. Consent was obtained from all patients included in this series. Data collected included demographics, presenting symptoms, imaging findings, operative details and follow-up outcomes.

A comprehensive search of published literature was conducted, using Medline and Embase databases, to identify all documented cases of chronic maxillary atelectasis, published in English-language from January 1964 to December 2017. The search terms 'chronic maxillary atelectasis' and 'silent sinus syndrome' identified 283 articles for review, which were then narrowed to 153 after the removal of duplicates. The abstracts of all search results were examined to locate relevant articles, and the full text of these articles was then reviewed by JPKH. Another 10 appropriate records were found by hand-searching the reference lists of relevant articles. A total of nine full-text articles were included in the qualitative analysis of bilateral chronic maxillary atelectasis.

Results

Patient one

A 32-year-old male presented with a 6-week history of bilateral nasal congestion, and malar, periorbital and frontal facial pain. His symptoms were preceded by a significant upper respiratory tract infection, for which he had taken multiple courses of oral antibiotics.

Nasendoscopy demonstrated a right-sided septal deviation and mucopurulent discharge from both middle meatus. Allergen-specific immunoglobulin E and skin prick testing was positive for house dust mite and cockroach allergens. A CT scan of the paranasal sinuses demonstrated bilateral opacification and hypoplasia of the maxillary antra (Figure 1). There was lateralisation of the posterior maxillary fontanelle and uncinate process bilaterally, and partial opacification of the bilateral ethmoid and sphenoid sinuses.

The patient was diagnosed with stage I chronic maxillary atelectasis bilaterally, in addition to septal deviation, chronic rhinosinusitis and allergic rhinitis. In view of the recent history, maximal medical therapy was trialled (oral and intranasal steroids, macrolide antibiotics, and saline irrigation), without resolution.

A bilateral uncinectomy and maxillary antrostomy was performed, in addition to a septoplasty and bilateral inferior turbinoplasty. At nine months' follow up, the patient remained symptom-free, with patent antral ostia bilaterally.

Patient two

A 48-year-old male was referred for management of daytime somnolence, and severe snoring causing bed-partner disturbance. Other than bilateral nasal congestion, there were no other sinonasal symptoms.

Examination revealed right anterior septal deviation and lateralisation of the lateral nasal walls. Sleep study demonstrated an apnoea hypopnea index of 19.4 events per hour, with a minimum oxygen saturation of 78 per cent. Skin prick testing was strongly positive for house dust mite allergen. Cone-beam CT scanning of the sinuses revealed right anterior septal deviation, complete opacification of both hypoplastic maxillary sinuses, and lateralisation of the uncinate processes (Figure 2).

The findings were consistent with a diagnosis of stage I bilateral chronic maxillary atelectasis, right anterior septal deviation, allergic rhinitis and moderate obstructive sleep apnoea.

Bilateral endoscopic maxillary antrostomy, inferior turbinoplasty and septoplasty were performed. At five weeks' postoperative follow up, nasendoscopy demonstrated a widely patent nasal airway and patent sinus ostia, with no complications.

Patient three

A 46-year-old male presented with a 2-year history of right anterior septal ulceration, resulting in bleeding and crusting. He also noted right-sided nasal obstruction and occasional headaches, although he denied other sinonasal symptoms.

On nasendoscopy, there was a 1.5 cm area of ulceration over the right anterior septum. *Staphylococcus aureus* was

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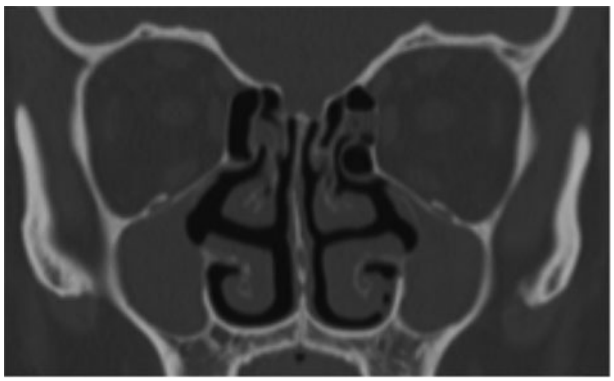

Fig. 1. Coronal computed tomography scan of patient one, demonstrating complete bilateral maxillary sinus opacification and hypoplasia, and uncinate process lateralisation.

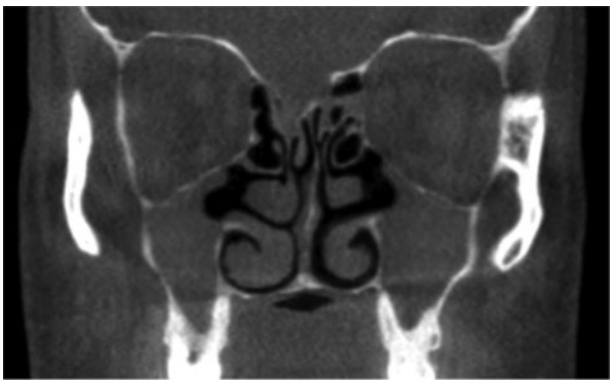

Fig. 2. Coronal cone-beam computed tomography scan of patient two, demonstrating complete opacification of bilateral hypoplastic maxillary sinuses and lateralisation of uncinate processes.

grown from a swab taken of this area. The rest of the examination, including visualisation of the nasal vestibule and postnasal space, was unremarkable. The results of an autoimmune screen were normal. The patient was diagnosed with *S aureus* septal infection secondary to digital trauma.

The patient responded to mupirocin ointment and avoidance of digital trauma. At six weeks' follow up, there was complete resolution of the ulceration. A CT scan of the sinuses, however, demonstrated septal deviation to the right and bilateral stage I chronic maxillary atelectasis (Figure 3).

The patient underwent a bilateral endoscopic maxillary antrostomy, as well as septoplasty and bilateral inferior turbinoplasty, with complete resolution of symptoms.

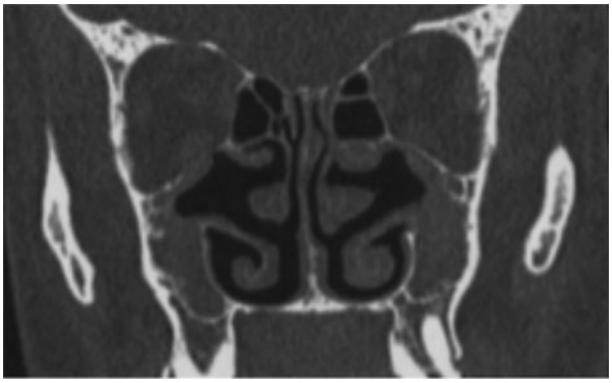

Fig. 3. Coronal computed tomography scan of patient three, demonstrating complete opacification of both maxillary antra and lateralised posterior maxillary fontanelles.

Discussion

Chronic maxillary atelectasis is thought to be uncommon amongst the general population, although the incidence remains unknown.[1] Chronic maxillary atelectasis has been defined as a unilateral condition.[12] While most cases of chronic maxillary atelectasis are indeed unilateral, this definition has been challenged by sporadic reports of bilateral cases (Table 2).[1,7–9,13–17] To date, there has not been a case series on bilateral chronic maxillary atelectasis. Furthermore, while Ferri et al. presented their case as bilateral silent sinus syndrome, the two maxillary sinuses were affected at different times.[7]

The aetiology and pathogenesis of chronic maxillary atelectasis remain controversial. The two main theories regarding the aetiology of chronic maxillary atelectasis are the obstruction of outflow and the mechanical theory.[18] It is thought to be a gradual and progressive disorder, with changes occurring over a course of weeks to months.[12] The three patients in our series presented with a variety of symptoms; however, all three complained of nasal obstruction. Gunaratne *et al.* had previously reported patient one's case as the first known report of bilateral chronic maxillary atelectasis in which the inciting event was identified and a rapid progress of disease followed.[6] It was therefore suggested that chronic maxillary atelectasis might, in some cases, have more of an acute progression than the name implies.

The most widely agreed-upon hypothesis describes sustained maxillary ostiomeatal complex obstruction, resulting in resorption of sinus mucosal gas and the development of negative pressure within the sinuses. This negative pressure then causes remodelling and demineralisation of the bone, with subsequent thinning and inward bowing of the maxillary sinus walls.[19,20]

Manometric studies performed by Kass, Salman and Montgomery, in 1996, revealed an average pressure of −8.4 ± 2.6 centimetres of water (cmH_2O) in those with chronic maxillary atelectasis affected maxillary sinuses, compared to the control (isobaric) pressures of the contralateral non-affected sinus and those with chronic rhinosinusitis.[20] Although only five affected patients were involved in this study, it demonstrated that those with chronic maxillary atelectasis may indeed have negative pressure within the maxillary sinus.[20] This pressure gradient appears to decrease, approaching atmospheric pressures with progressive chronic maxillary atelectasis disease. This was demonstrated by a pressure of −13 cmH_2O in the patient with stage I disease, and −6 and −8 cmH_2O in the two patients with stage III disease. It has been postulated that the ongoing osseous remodelling and inward displacement of the antral walls is due to chronic inflammation and mucus retention.[11,20]

Rose and Lund proposed that severe nasal inflammation with marked engorgement of a turbinate in a narrow airspace may cause prolonged impairment of antral drainage.[21] The retained secretions may be associated with sub-atmospheric pressures, causing secondary collapse of the antral walls. This collapse, or 'implosion', will continue to progress until the limits of remodelling are reached or until the sinus re-aerates as the acute intranasal inflammation settles.[21]

However, the exact causative factor leading to ostial occlusion remains unclear. It has been proposed that a deficiency in

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Table 2. Literature review of bilateral chronic maxillary atelectasis

Study	Gender, age (years)	Presentation	Chronic maxillary atelectasis stage	Treatment
Kass *et al.*[1]	M, 64	Sinonasal	I	Unknown
	F, 30	Sinonasal	I	Unknown
Ando & Cruz[13]	F, 36	Orbital	III	Endoscopic MMA & OFR
Liss *et al.*[14]	M, 56	Orbital	III	Endoscopic MMA & OFR
Ferri *et al.*[7]	F, 27	Orbital	III	Endoscopic MMA & OFR
Suh *et al.*[8]	M, 29	Incidental	III	Endoscopic MMA
Sun & Dubin[15]	M, 52	Sinonasal & orbital	III	Endoscopic maxillary ostium balloon dilatation
Kohn *et al.*[16]	Unknown	Unknown	Unknown	Unknown
Lin *et al.*[17]	Unknown	Unknown	Unknown	Endoscopic MMA
Shieh *et al.*[9]	F, 68	Orbital	III	Endoscopic MMA & orbital decompression
Ho *et al.* (current study)	M, 32*	Sinonasal	I	Endoscopic MMA
	M, 48	Sleep-disordered breathing	I	Endoscopic MMA
	M, 46	Sinonasal	I	Endoscopic MMA

*Patient initially reported in Gunaratne *et al.* (2016).[6] M = male; F = female; MMA = middle meatus antrostomy; OFR = orbital floor reconstruction

the posterior attachment of the uncinate process to the inferior turbinate, and a lateralised or hypermobile medial infundibular wall, can result in a flap-valve occlusion of the ostium.[1,22] Other proposed mechanisms include occlusion by inspissated mucus, mucosal changes from adjuvant therapy, and other predisposing anatomical factors, such as a hypoplastic maxillary antrum leading to decreased ostial diameter and resulting in more frequent occlusion.[14,23]

The argument against the obstruction of flow theory is that there are many more patients with chronic sinusitis and ostial occlusion than there are with chronic maxillary atelectasis. A further explanation that may account for this anomaly is the mechanical theory.[24] This theory hypothesises that masticatory muscle contraction creates negative pressure in the infratemporal and pterygopalatine fossa. If a small communication exists, either congenital or post-traumatic, between the infratemporal or pterygopalatine fossa and the antrum, and the antral ostium becomes obstructed, the transmitted negative pressure may be sufficient to cause antral wall collapse.[24]

There are two distinct goals in the management of chronic maxillary atelectasis. The first goal is to re-establish aeration to the affected maxillary sinus through marsupialisation into the nasal cavity. The second goal is to restore the normal orbital architecture, if required.[13] Most authors agree that the method of choice to achieve the sinonasal goal is endoscopic uncinectomy and maxillary antrostomy. However, it should be noted that the risk of orbital content injury is much greater in chronic maxillary atelectasis patients because of the more inferior positioning of the globe and orbital floor. More recently, some authors have proposed the use of balloon sinus dilatation surgery for chronic maxillary atelectasis.[15,25,26] Although balloon sinus dilatation may be more cost-effective, further studies need to be performed prior to the routine implementation of this procedure.

Early reports suggested that any defect in the orbital floor required routine correction. Subsequent reports recommended that a two-stage repair should be used to correct the sinonasal and orbital defects. A volumetric analysis performed by Lin *et al.* demonstrated a reduction in orbital volume and re-expansion of the maxillary sinus on the diseased side following corrective endoscopic sinus surgery.[17] Reports have shown that patients may have spontaneous resolution of the enophthalmos and hypoglobus following only re-ventilation of the maxillary sinus, without addressing the orbit.[13] Furthermore, disease progress stops after surgical aeration of the maxillary sinuses.[27] In view of these findings, it is agreed by most authors that immediate orbital floor reconstruction is not required in the majority of non-diplopic patients.[27–29] Clinicians should consider a minimum delay period of six months following initial antrostomy before re-evaluating the need for orbital floor reconstruction.[29]

- Chronic maxillary atelectasis is a rare condition involving a progressive decrease in maxillary sinus volume
- In addition to sinonasal symptoms, disease progression can result in ophthalmological and cosmetic deformities
- It has traditionally been defined as a unilateral condition
- This paper challenges the unilateral definition of this condition
- The paper presents a case series of three patients with bilateral chronic maxillary atelectasis

As illustrated in our paper, bilateral chronic maxillary atelectasis (including silent sinus syndrome), while being a rare clinical entity, does occur. As the clinical signs are often subtle and may be symmetrical in bilateral disease, rhinologists should reserve a high degree of suspicion.

Conclusion

Although a rare condition, chronic maxillary atelectasis can lead to significant sinonasal symptoms, as well as ophthalmological and facial deformities in the later stages. It is often underdiagnosed or mistaken for other pathologies. We present

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a case series of three patients with bilateral chronic maxillary atelectasis, which challenges the current inclusion of unilateral disease in the definition. Further investigation is required to determine the aetiology and pathophysiology of the disease.

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Competing interests. None declared

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