

Natural progression of bilateral maxillary silent sinus syndrome: A metachronous case report

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Bianca Rullan-Oliver¹ , Edgar del Toro-Diez² 
and Juan C Portela-Arraiza¹

Abstract

Silent sinus syndrome refers to a constellation of spontaneous and progressive enophthalmos and hypoglobus in the setting of asymptomatic ipsilateral maxillary sinus atelectasis. Although its exact etiopathogenesis is not completely understood, obstruction of the ostiomeatal complex appears to be the inciting event. Most of the reported cases of silent sinus syndrome involve one maxillary sinus. Only a handful of true bilateral silent sinus syndrome cases have been reported in the literature. The aim of this report is to present a case of metachronous bilateral maxillary silent sinus syndrome and its natural progression. Also included is a review of the literature on ethmoidal, frontal, and bilateral maxillary silent sinus syndrome.

Keywords

Otolaryngology, bilateral maxillary silent sinus syndrome, imploding antrum syndrome, maxillary atelectasis, enophthalmos, hypoglobus

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Introduction

Silent sinus syndrome (SSS) consists of spontaneous and progressive enophthalmos and hypoglobus in the setting of asymptomatic ipsilateral maxillary sinus atelectasis. Montgomery described the first case of SSS in 1964, and Soparkar et al.¹ coined the term 30 years later. Less than 100 cases of SSS have been published in the literature. Historically, SSS was defined as a unilateral maxillary sinus atelectasis and enophthalmos, but over the last 10 years a handful of true bilateral SSS cases have been reported.

The exact etiopathogenesis of SSS is incompletely understood, but there is a clear consensus that the main inciting event is obstruction of the ostiomeatal complex (OMC).² OMC obstruction leads to hypoventilation and negative pressure on the affected paranasal sinus, subsequently resulting in collapse of its walls and ipsilateral orbital volume changes.³ In the setting of long-standing OMC obstruction, progressive asymptomatic facial asymmetry may ensue over a period of weeks to months.

Patients typically present to the ophthalmologist, and less commonly to the otolaryngologist, due to the chief complaint of ocular asymmetry of 3–8 months of evolution. Average enophthalmos and hypoglobus at presentation are 3

and 3.4 mm, respectively.^{1,2,4,5} Other distinguishing ocular features include a deep superior orbital sulcus and eyelid retraction. Sometimes pseudopneumoorbita may be seen as well, described as air trapped under the superior eyelid and superior conjunctival recess, which gives the appearance of air in the orbit in computed tomography (CT) scans. Trapped air in the conjunctival recess may be noticed as an audible clicking sound when blinking.⁶ Extraocular motion and visual acuity are usually unaffected, although patients with severe hypoglobus may experience diplopia.^{4,7,8} Nasal endoscopy will reveal a lateralized uncinate process, with obstruction of the maxillary natural ostium, and an enlarged middle meatus.⁴

The diagnosis of SSS is suggested clinically, but confirmed with a CT scan, which will reveal partial or complete

¹Department of Otolaryngology Head and Neck Surgery, School of Medicine, University of Puerto Rico, San Juan, Puerto Rico

²School of Medicine, University of Puerto Rico, San Juan, Puerto Rico

Corresponding Author:

Bianca Rullan-Oliver, Department of Otolaryngology Head and Neck Surgery, School of Medicine, University of Puerto Rico, PO BOX 365067, San Juan 00936-5067, Puerto Rico.

Email: brullan6@gmail.com



opacification of the affected sinus as well as inward collapse, and possibly osteopenia, of its walls.² Maxillary sinus atelectasis will inevitably result in progressive hypoglobus and enophthalmos. Treatment consists of endoscopic uncinectomy and maxillary antrostomy to restore sinus ventilation and halt disease progression. Surgery alone can often restore paranasal sinus volume, therefore improving patients' orbital asymmetry.^{9,10} In cases of severe hypoglobus, enophthalmos, or diplopia, oculoplastic reconstruction of the orbital floor should be considered.⁵

Most of the reported cases of SSS involve one maxillary sinus; however, a few case reports on ethmoid and frontal SSS, as well as bilateral SSS, have been described in the literature. Bilateral SSS may be more difficult to suspect than unilateral disease given the absence of obvious facial asymmetry. Only a handful of bilateral SSS cases have been reported in the literature. The aim of this report is to present a case of metachronous bilateral maxillary SSS and its natural progression.

Case report

This is the case of an otherwise healthy 47-year-old female with history of well-controlled allergic rhinitis, who was referred by Neurology to our Otolaryngology clinic due to an incidental left "maxillary sinusitis" found on magnetic resonance imaging (MRI) performed for a chief complaint of headaches. The patient was asymptomatic at the moment of presentation and denied history of facial trauma and surgery. Physical examination revealed a mild left enophthalmos and hypoglobus, and the diagnosis of left maxillary SSS was confirmed on a CT scan (Figure 1).

Surgery was offered at the time of initial presentation, but the patient was lost to follow-up and returned 2 years 5 months later with a new CT scan that revealed bilateral maxillary SSS (Figure 2). The patient continued to be asymptomatic at the time of her second presentation, and the diagnosis of metachronous bilateral SSS was made. Figures 3 and 4 depict the temporal development of right-sided enophthalmos secondary to maxillary sinus atelectasis from SSS. The patient underwent bilateral endoscopic sinus surgery, and postoperative endoscopic examination revealed patent and clear maxillary sinuses with reestablishment of adequate ventilation. The patient had a prompt recovery after surgery and has remained asymptomatic, without surgical complications or clinical progression of disease.

Discussion

Silent sinus syndrome, also known as imploding antrum syndrome, is a rare phenomenon consisting of enophthalmos and hypoglobus secondary to asymptomatic maxillary sinus atelectasis, in the absence of prior trauma, surgery, and sinonasal inflammatory disease. SSS typically presents between the third and fifth decades of life, consistent with

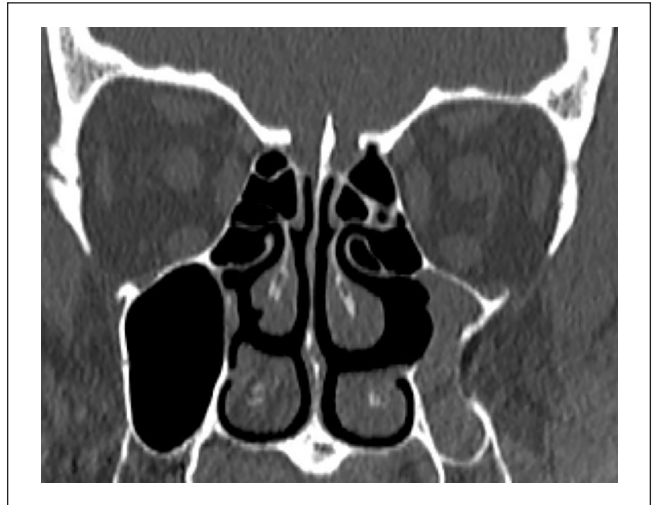


Figure 1. Left maxillary SSS. This coronal CT scan performed on December 2014 reveals a fully opacified left maxillary sinus with marked collapse of the lateral and medial walls and lateralization of the uncinate and lateral nasal wall. It also suggests the presence of downward collapse of the maxillary roof, with subsequent development of subtle hypoglobus.



Figure 2. Bilateral maxillary SSS. This is a follow-up coronal CT scan performed on May 2017 that illustrates the incidental development of right maxillary SSS, characterized by complete opacification as well as inward collapse of its walls and hypoglobus.

the presenting age of our patient.^{1,2} Review of all 84 reported cases of SSS by Numa et al.⁷ revealed that the average age of presentation is 39 years. It affects males and females equally and has no racial predilection. There seems to be a slight right-side predominance (57%).¹¹ Patients classically present with an otherwise asymptomatic, slowly progressive, enophthalmos and hypoglobus over a course of weeks to months,² with the mean duration of ocular symptoms before presentation being 3 to 6 months.^{1,12,13} Naik et al.¹³ reported a mean

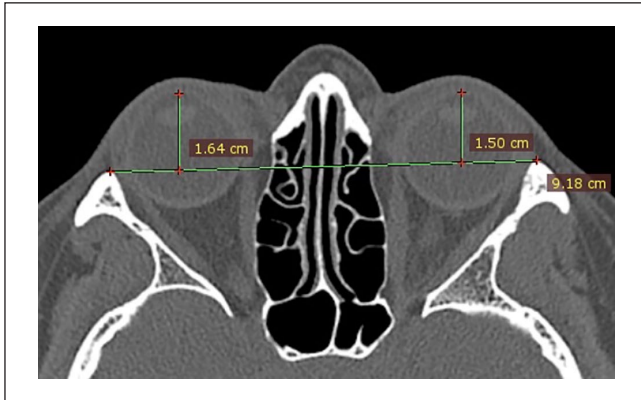


Figure 3. Unilateral left maxillary SSS. This is an axial image of the patient's first paranasal sinus (PNS) CT scan, performed on December 2014. The measurements reading 1.50 and 1.64 cm represent the anterior projection of the left and right orbits, respectively, in relation to the interzygomatic line (9.18 cm). Mild left enophthalmos is appreciated in comparison with the unaffected right eye.

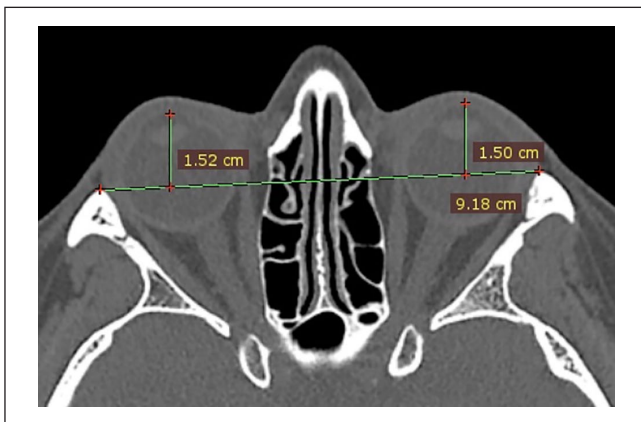


Figure 4. Metachronous bilateral maxillary SSS. This axial PNS CT scan illustrates the development of mild right enophthalmos 2 years 5 months later in the same patient, as evidenced by a 1.2 mm decrease in anterior projection of the right eye, now 1.52 cm from 1.64 cm. The projection of the left eye (1.50 cm) and the interzygomatic line distance (9.18 cm) remained unchanged.

duration of symptoms of 6.52 months. SSS is acquired; it is neither congenital nor preceded by sinus symptoms, trauma, or surgery. Nonetheless, approximately 36% of patients have a remote history of sinus disease in the childhood.¹

Patients generally present with spontaneous facial or orbital asymmetry. All patients have enophthalmos and hypoglobus, which typically range from 2 to 5 mm and 2 to 6 mm, respectively. At presentation, enophthalmos averages 3 mm and mean hypoglobus is 3.4 mm.^{1,2,4,5} Patients exhibit a deep superior orbital sulcus and eyelid retraction. Ocular motility and visual acuity are rarely affected, but patients with severe hypoglobus may develop diplopia. When present, patients

Table 1. Clinical signs of silent sinus syndrome.

Signs of silent sinus syndrome	Frequency
Enophthalmos	Present
Hypoglobus	Present
Orbital asymmetry	Common
Deepening of superior sulcus	Common
Lid lag and lagophthalmos	Common
Gaze restriction	Uncommon
Sinking or pulling sensation of the eye	Uncommon
Diplopia	Uncommon
Ptosis	Uncommon
Audible clicking sound with blinking due to air being trapped in the conjunctival recess (Pseudopneumoorbita)	Uncommon
Malar depression	Uncommon

usually exhibit vertical diplopia due to dysfunction of the inferior rectus muscle, which is attached to the orbital bone and is therefore affected when the globe is displaced inferiorly from its natural position secondary to downward collapse of the orbital floor.^{4,7,8} Superior oblique muscle dysfunction, contralateral exophthalmos, eyelid retraction, lagophthalmos, ptosis, gaze restriction, and pseudopneumoorbita have also been reported.⁵ Patients may rarely report malar depression due to severe collapse of the anterior maxillary wall.¹⁴ The presenting signs of SSS may vary depending on the severity of the atelectasis, but a constant clinical finding is enophthalmos and hypoglobus (Table 1).

The etiopathogenesis of SSS is not completely understood. However, the main inciting event is obstruction of the OMC, which leads to hypoventilation of the affected sinus and negative intra-sinus pressure that results in the collapse of its walls, resulting in downward displacement of the orbit.^{2,3} Possible mechanisms of acquired anatomic obstruction of the OMC include a lateralized middle turbinate, hypermobile or lateralized medial infundibular wall, inflamed mucosa, infraorbital ethmoid (Haller) cells, inspissated mucus, mucocele, or a nasal polyp occluding the antrum or ostium of the affected sinus, resulting in sinus outflow obstruction and subsequent hypoventilation of the affected sinus.^{11,15} Ipsilateral nasal septum deviation is observed in 50%–71% of patients, possibly contributing to the development of SSS.^{1,5,10} Other theoretical etiologies described in the literature include traumatic nasal intubation or nasogastric tube placement, and barotrauma following scuba diving.^{14,16} In addition, gender and sinus mucosa morphology may also be influencing factors.¹⁷

The treatment of choice of SSS consists of restoration of sinus ventilation via an endoscopic uncinctomy and maxillary antrostomy. A Caldwell-Luc approach was used in the past, but has become supplanted by endoscopic sinus surgery. Surgery alone halts disease progression and frequently restores paranasal sinus volume, improving patients' orbital

asymmetry.^{9,10} Orbital floor reconstruction is controversial but should be considered in cases of severe hypoglobus, enophthalmos, diplopia, or little improvement of these symptoms after maxillary antrostomy.⁵ The orbit may be repaired via a transconjunctival or subciliary approach with placement of a titanium mesh or autogenous cartilage, which may be harvested from the nasal septum or auricular concha. The timing of repair is also controversial, with some authors proposing concomitant functional endoscopic sinus surgery and orbital floor reconstruction, while others advocate delaying the orbital floor repair for 2–6 months given that enophthalmos may improve or even resolve in some patients after maxillary antrostomy. A close follow-up is important to ensure SSS progression has ceased and refer selected patients for oculoplastic reconstruction.

Most of the cases of SSS described in the literature involve the maxillary sinus, unilaterally. However, a handful of cases of ethmoidal and frontal SSS, as well as of bilateral SSS, have been recently reported. Braganza and Khooshabeh described the first case of SSS involving the ipsilateral ethmoid air cells in 2005, and McArdle and Perry published the first case of a solitary ethmoidal SSS in 2010, in the absence of maxillary sinus atelectasis.^{15,18} In 2012, Naik et al.¹³ described the first case of a frontal SSS due to a large obstructive Type III Kuhn cell. Ethmoidal and frontal SSS may be explained by the same pathophysiology as maxillary SSS, OMC obstruction, given their common drainage pathway to the middle meatus.

Bilateral SSS is extremely rare, with only four reported cases in the literature.^{5,9,19,20} Three patients were diagnosed incidentally with synchronous bilateral maxillary SSS upon initial presentation. The other case, reported by Ferri et al.,⁹ was that of a 27-year-old female who developed right maxillary SSS 4 months after functional endoscopic sinus surgery for the original left maxillary SSS. Similarly, we illustrate the case of a 49-year-old female patient who developed metachronous right maxillary SSS over 2 years after she was incidentally diagnosed with left maxillary SSS. The patient did not undergo surgery, despite it having been offered at the time of diagnosis, and she returned to our clinics with a new CT scan 2 years 5 months after her initial presentation, which revealed the incidental and rare development of contralateral maxillary SSS. This suggests that there may be a temporal relationship in the development of bilateral SSS and that bilateral SSS occurs in a staggered manner. In addition, the CT scans depict the slowly progressive nature of this condition and suggest that, if left untreated, SSS will not resolve on its own.

Conclusion

Contralateral SSS can be diagnosed in a synchronous or metachronous pattern. In this report, we illustrate the potential delay in manifestation of contralateral maxillary SSS. Our patient developed bilateral SSS within 2 years

5 months of her original presentation, to our knowledge the longest time-interval described in the literature. Our findings not only illustrate the natural progression of ipsilateral SSS when left untreated, but also demonstrate that bilateral SSS may develop within years of the initial presentation, mandating adequate follow-up of these patients.

Further research is needed to better understand the precise etiology of SSS, recognize the triggers of OMC obstruction, identify patients at risk for delayed contralateral manifestation, and therefore prevent disease progression and disfiguring facial asymmetry prior to its onset. We understand that this may be a challenging task given that the diagnosis of SSS is often incidental, but believe that a comprehensive understanding of this condition may allow for early diagnosis and prevention of potentially permanent facial asymmetry, or even visual disturbances.

Declaration of conflicting interests

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.



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Informed consent

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ORCID iDs

Bianca Rullan-Oliver  <https://orcid.org/0000-0002-9341-0126>
 Edgar del Toro-Diez  <https://orcid.org/0000-0002-0545-4314>

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