

# Bilateral maxillary silent sinus syndrome: A case report and literature review

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## Abstract

Silent sinus syndrome refers to a spontaneous enophthalmos caused by maxillary sinus collapse without any symptomatic sinonasal illnesses. Its prevalence is almost entirely unilateral. The authors report a patient with a bilateral silent sinus syndrome managed successfully by middle meatal antrostomies. This case brings attention to recognizing bilateral silent sinus syndrome. Because of its bilateral involvement, the facial disfigurement might be recognized late due to the symmetrical presentation. This article highlights the pathophysiology and reports the detailed course of such a rare disease. The balloon dilatation is promising as a newly described treatment modality in a patient with silent sinus syndrome, although more long-term data on its outcome is needed.

## Keywords

Bilateral, silent sinus syndrome, maxillary sinus

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## Introduction

Silent sinus syndrome (SSS) is almost entirely reported as a unilateral disease in the literature. It is described as a contracture of the maxillary sinus without any symptomatic sinonasal disease that results in spontaneous enophthalmos.<sup>1</sup> Being a bilateral presentation is an extremely rare disease and may go unnoticed or delayed in the diagnosis due to the symmetrical enophthalmos. Early identification and management are important in preventing long-term facial disfigurement.<sup>2</sup>

In this article, we describe in detail the clinical course as well as the successful antrostomies in the management of such a rare entity.

## Case report

An obese 21-year-old male presented to the clinic complaining of facial pain and nasal blockage. No history of trauma, previous sinonasal surgery, tumors, or radiation. Patient's height, weight, and body mass index are 184 cm, 158 kg, and 46.7 kg/m<sup>2</sup>, respectively. The trial of intranasal corticosteroid and nasal douche did not improve his complaints. A lateralized uncinate process, blockage of the maxillary natural ostium, and an expanded middle meatus were seen on nasal endoscopy. CT scan showed (Figure 1) the finding of

bilateral SSS, concha bullosa that was large on the left side, left frontoethmoiditis, and deviated nasal septum. An inferior bowing of the roof of maxillary sinuses was seen on CT with an increased orbital volume. The score of a preoperative 22-item sinonasal outcome test (SNOT-22) was 46/110; (Figure 2: The dotted line/square marks). The patient underwent bilateral middle meatal antrostomy, concha bullosa resection, left anterior ethmoidectomy, left frontal sinus surgery, and septoplasty (Figures 3 and 4).

After 6 weeks of postoperative follow-up, endoscopic examination indicated patent maxillary sinuses, with sufficient ventilation restored.

The patient recovered quickly following surgery and has remained asymptomatic, with no surgical complications or

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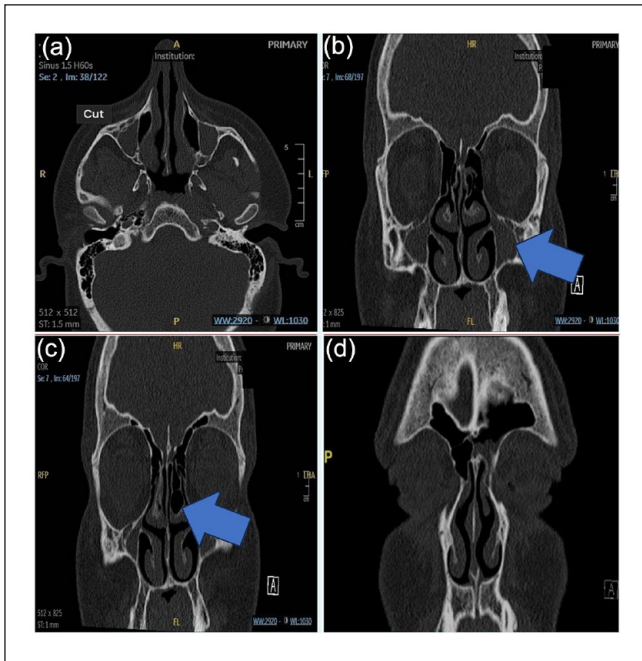
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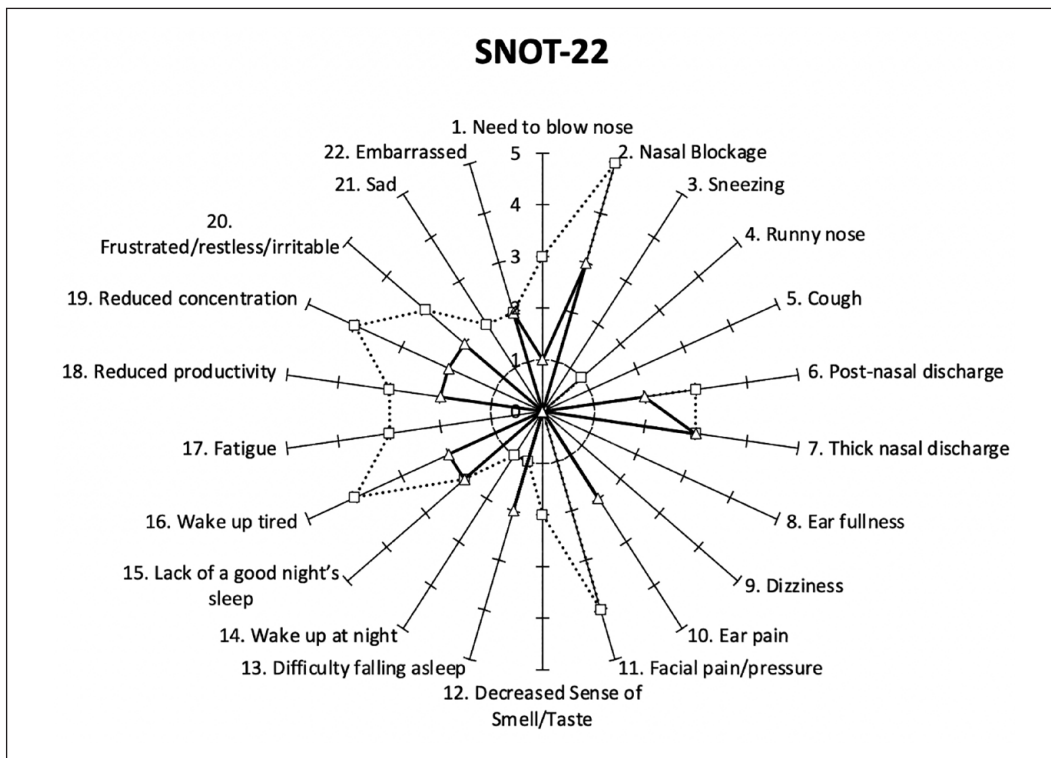
**Figure 1.** CT scan (Axial (a) and Coronal cuts (b–d)) of the paranasal sinus showing opacified and contracted maxillary sinuses (a–c) lateralized uncinate processes (b) concha bullosa (c) partial opacity along the left frontal sinus drainage pathway (d).

clinical progression of the disease, and the SNOT-22 score improved to 25/110 (Figure 2: The solid line/triangle marks).

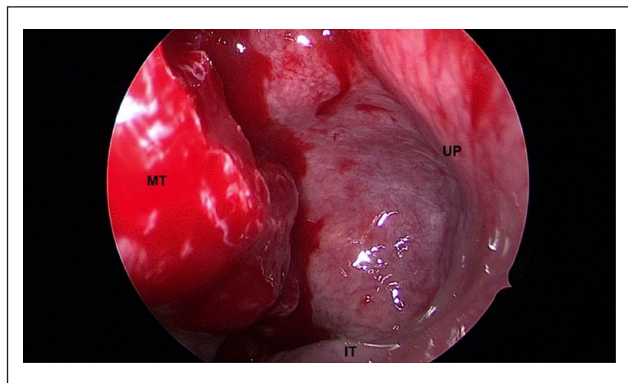
**Discussion**

SSS is defined as a spontaneous, progressive enophthalmos, and hypoglobus in the setting of asymptomatic maxillary sinus atelectasis. It is a rare phenomenon that its specific etiopathogenesis is unknown. However, there is broad agreement that the key initiating factor is ostiomeatal complex blockage (OMC). This blockage causes hypoventilation and negative pressure on the affected sinus, causing its walls to collapse and ipsilateral orbital volume alterations.<sup>3</sup>

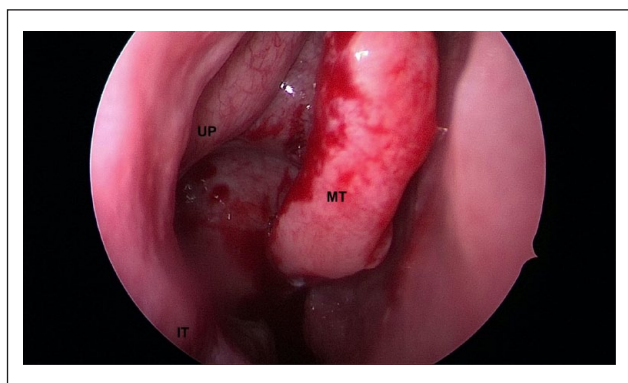
Specific anatomical variants can lead to OMC obstruction. Concha bullosa, a deviated nasal septum, and an acute or chronic infection are all potential SSS risk factors. These risk factors have the potential to impede the ostiomeatal complex. As a result, the affected maxillary sinus experiences hypoventilation and negative pressure, resulting in maxillary wall collapse and ipsilateral orbital volume alterations.<sup>4</sup> Similar to our patient, his nasal blockage and pain were explained by the findings of the deviated nasal septum, concha bullosa, and frontoethmoiditis. A trial of nasal, topical medication started with no improvement, for which a CT



**Figure 2.** SNOT-22, the dashed line/square marks represent the preoperative total score (46/110) while the continuous line/triangle marks represent the total postoperative score (25/110). SNOT-22: 22-item sinonasal outcome test.



**Figure 3.** Intraoperative view (using a 30° rigid endoscope) after the left concha bullosa lateral lamella resection. Left middle meatus is deep and showing a lateralized unciniate process. MT: Middle turbinate; IT: Inferior turbinate; UP: Uncinate process.



**Figure 4.** Intraoperative view (using a 0° rigid endoscope). Right middle meatus is deep and shows a lateralized unciniate process. MT: Middle turbinate; IT: Inferior turbinate; UP: Uncinate process.

scan was arranged, showed the previous finding, and confirmed the diagnosis of SSS.

Once SSS diagnosis is established, it is typically treated by antrostomy. However, Sun et al.<sup>5</sup> reported a patient suffering from bilateral SSS, which was successfully treated using a balloon dilatation of the maxillary sinus ostium in an office-based setting. However, this needs more research to ensure the long-term patency of the sinus. This widens the options of the treating physicians when dealing with SSS. A bilateral maxillary antrostomy was performed on our patient, and he remained asymptomatic, and sinuses patency was ensured for the subsequent follow-up visits.

It is essential to involve ophthalmologists in the care of SSS patients as their role is of importance in the diagnosis as well as the management. In certain severe cases, the orbit floor may need reconstruction using a titanium mesh or autogenous cartilage. Close follow-up is needed to ensure patency of sinus and cessation of progression of the manifestation of SSS.<sup>4</sup>

The fact that this is a single-case report and the rarity of this disease limited our understanding of the development of bilateral SSS.

## Conclusion

Most reported cases of SSS involve one maxillary sinus; however, there are a few reports on bilateral SSS in the literature. Bilateral SSS may be more difficult to detect than unilateral one, given the lack of asymmetry in facial manifestation. This article aims to show the clinical picture of a bilateral SSS. More research is needed to understand the etiology of SSS, recognize the triggers of OMC obstruction, identify patients at risk for bilateral manifestation, and prevent disease progression and facial disfigurement. We believe that a comprehensive understanding of this condition may allow early diagnosis and prevention of potentially permanent facial manifestation or even visual disturbances.

## Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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## Ethics approval

Ethical approval to report this case was obtained from King Saud University institutional review board (22/0593/IRB)\*.

## Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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