

## Spontaneous Silent Sinus Syndrome (Imploding Antrum Syndrome): Case Series of 11 Patients

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### Abstract

**Background:** Silent sinus syndrome (SSS) is a spontaneous, asymptomatic collapse of the maxillary sinus and orbital floor associated with negative sinus pressure. It is also known as the imploding antrum syndrome. It typically presents with a 'sunken' eye (enophthalmos), asymmetry of the level of the eyes (hypoglobus) and downward bowing of the orbital floor causing a reduction in the size of the maxillary sinus.

**Methods:** A retrospective analysis of clinical notes and radiological data of 11 patients with spontaneous SSS were reviewed. Their endoscopic and radiological findings were studied.

**Results and conclusion:** Spontaneous SSS is a rare condition characterized by unilateral enophthalmos, hypoglobus with radiological findings of ipsilateral depression of the orbital floor, reduction in volume and opacification of the maxillary sinus. Pathogenesis of this condition remains unclear. The fixed sign is the laterally displaced uncinat process which could be detected in all eleven cases, leading to obstruction of the infundibulum. It may have a direct relation in the development of maxillary sinus atelectasis.

*Key words:* Maxillary sinus, Sinus atelectasis, Silent sinus syndrome, Enophthalmos, Facial asymmetry.

### Introduction

The silent sinus syndrome, which is also known as chronic maxillary sinus atelectasis, consists of painless enophthalmos and inward retraction of the ipsilateral maxillary sinus walls on imaging studies<sup>(1)</sup>. Enophthalmos resulting from maxillary sinus pathology was first reported by Montgomery in 1964<sup>(2)</sup>. The term "silent sinus syndrome" was introduced by Soparkar et al in 1994<sup>(3)</sup>.

Chronic maxillary atelectasis is a descriptive term to describe a long-standing diminution of maxillary sinus volume and radiographic findings of an inward bowing of the walls of the antrum<sup>(4)</sup>. Silent sinus syndrome is a rare disorder, but it is probably underdiagnosed because of a lack of recognition<sup>(1)</sup>. The typical patient with silent sinus syndrome is an adult in the third through fifth decades of life who presents with spontaneous, painless, occasionally progressive enophthalmos and hypoglobus<sup>(1, 3)</sup>.

Physical examination findings may include upper lid retraction, deepened upper lid sulcus, malar depression, facial asymmetry, and diplopia.

The diagnosis of silent sinus syndrome should be differentiated from other causes of spontaneous enophthalmos. Although the diagnosis is usually suspected clinically, it is confirmed radiologically.

The imaging findings are characteristic<sup>(4)</sup>. The primary finding is maxillary sinus volume loss due to inward retraction of the sinus walls, which accounts for the increased orbital volume and enlargement of the middle meatus. Typically, all 4 walls of the sinus are retracted, though one of the medial, anterior, or posterolateral walls may be spared. The orbital floor (maxillary roof) is always retracted and commonly thinned. The other walls may be thinned, normal, or slightly thickened. The maxillary infundibulum is always occluded with opacification of the sinus. The uncinat process is retracted against the inferomedial aspect of the orbital wall.

Events leading to retraction of the walls of the obstructed sinus are not clearly understood<sup>(5)</sup>. Numerous case studies and reviews have shed further light on the pathogenesis and management of this condition<sup>(6)</sup>. Some authors believe that a congenital underdevelopment of the maxillary sinus is responsible for the development of silent sinus syndrome, but the acquired nature of this condition is now more readily apparent<sup>(3, 6)</sup>. Obstruction of the maxillary ostium appears to play a critical role in the development of silent sinus syndrome. Childhood nasal trauma and surgical trauma were reported as factors in the later insidious development of silent sinus syndrome<sup>(7, 8)</sup>. Obstruction of the sinus ostium by the uncinate process is always present, but it is not clear whether this is a cause of the development of intrasinus negative pressure or a result of sinus wall retraction under the negative pressure.

It is a rare condition and to date only 121 cases have been reported in the literature<sup>(6, 9)</sup>. We report the third largest case series of 11 patients with spontaneous SSS and describe their clinical and radiological features.

### Patients and Methods

Eleven cases of silent sinus syndrome were referred to the otolaryngology clinic of our institutions through the last 5 years. There were 7 males (64%) and 4 females (36%), with an age range 22-58 years.

Nine patients presented to ENT clinic by facial asymmetry and nasal complaints. Last two cases were referred from ophthalmology clinic with enophthalmos and diplopia (Figure 1).

All the patients were subjected to full history, clinical examination with endoscopic examination of the nasal cavity. CT scan was done to the patients to confirm the diagnosis of silent sinus syndrome. The patients were imaged with unenhanced CT in bone algorithm with 3-mm-thick slices in both the coronal and axial planes. Study of the nasal cavity anatomical abnormalities through the endoscopic and radiological findings was done in all cases. The anatomical study included the following:

No.	Age year	Sex	Duration of symptoms	Presentation
1	58	M	18 months	Nasal obstruction & Facial asymmetry
2	30	M	24 months	Nasal obstruction & Facial asymmetry
3	26	F	10 months	Nasal obstruction & Facial asymmetry
4	28	M	12 months	Diplopia & enophthalmos
5	35	F	6 months	Diplopia
6	57	M	12 months	Nasal obstruction & Facial asymmetry
7	22	M	10 months	Nasal obstruction & Facial asymmetry
8	46	F	12 months	Nasal obstruction & Facial asymmetry
9	31	M	6 months	Nasal obstruction & Facial asymmetry
10	43	F	8 months	Nasal obstruction & Facial asymmetry
11	50	M	24 months	Nasal obstruction & Facial asymmetry

Table (1):The collective data of the patients regarding the age, sex, duration of symptoms and presentation. [M = Male; F = Female]

the size of the maxillary sinus, retraction of the sinus walls, aeration of the sinus, the uncinate process displacement, middle turbinate abnormalities and nasal septum deviation.

### Results

Table (1) shows the collective data of the eleven patients regarding the age, sex, duration of symptoms and presentation.

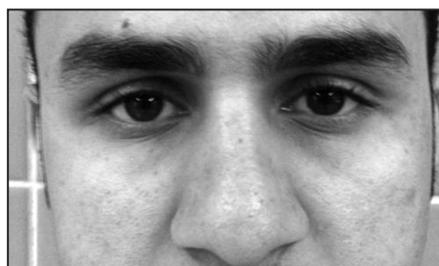


Figure (1): A case of Left maxillary sinus atelectasis (Patient n.9). Note the left hypoglobus, enophthalmos and facial asymmetry.

Analysis of the radiological findings of the affected sinus showed that all the cases have the radiological characters of atelectatic sinus in the form of reduced sinus size, with retraction of the sinus walls and opacity of the sinus.

Study of the anatomical findings of the nasal cavity endoscopically and radiologically of all patients revealed that one case has nasal septal deviation towards the affected side and two cases have septal deviations towards the other side (Table 2). One case has bilateral concha bullosa with unilateral atelectatic sinus. One case has paradoxical middle turbinate on the atelectatic side, concha bullosa on the non-atelectatic side and both sides are filled with mucus. Six cases have within normal septum and turbinates with unilateral atelectatic sinus. The uncinate process was laterally displaced with obstruction of the infundibulum in all cases.



Figure (2): CT scan coronal cut (Patient n.1) shows septal deviation to the right side with right opaque atelectatic maxillary sinus.

No.	Sinus retraction	Sinus opacity	Infundibulum	Uncinate process	Septal deviation	Concha bullosa	Paradoxical turbinate
1	Right	Right	Right closed	Displaced	To the right	Absent	Absent
2	Right	Right	Right closed	Displaced	Absent	Absent	Absent
3	Left	Left	Left closed	Displaced	Absent	Absent	
4	Right	Right	Right closed	Displaced	Absent	Absent	Absent
5	Right	Right	Right closed	Displaced	Absent	Absent	Absent
6	Left	Left	Left closed	Displaced	Absent	Absent	
7	Right	Right total Left partial	Right closed	Displaced	Absent	Bilateral	Absent
8	Left	Bilateral	Left closed	Displaced	Absent	Right	Left
9	Left	Left	Left closed	Displaced	Absent	Absent	Absent
10	Right	Right	Right closed	Displaced	Absent	Bilateral	Absent
11	Right	Right	Right closed	Displaced	To the right	Absent	Absent

Table (2): The endoscopic and radiological finding of the patients.

Case (1): Male patient 58 years old presented to the ENT clinic with nasal obstruction and facial asymmetry for 18 months. CT scan shows nasal septal deviation to the right side with right opaque atelectatic maxillary sinus (Figure 2). Lateral displacement of the uncinate process obstructing the infundibulum was also detected.

Case (2): Male patient 30 years old presented to the ENT clinic with nasal obstruction and mild facial asymmetry for 2 years. The radiological findings were in the form of right atelectatic opacified maxillary sinus, left maxillary sinus opacity but not atelectasis and midline nasal septum (Figure 3).

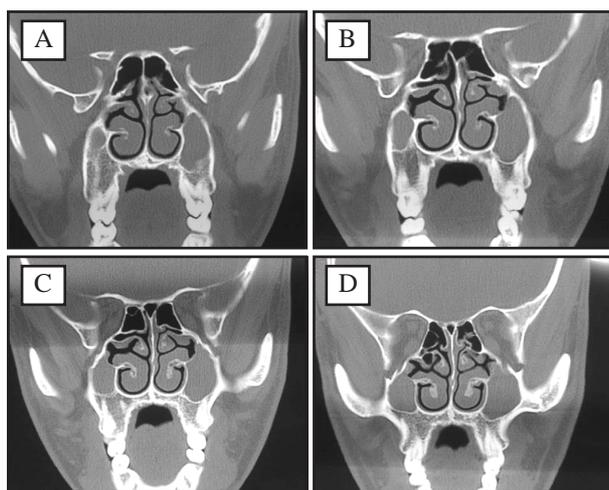


Figure (3): A, B, C and D: CT scan coronal (Patient n.2) shows opaque right maxillary sinus with reduced volume.

Case (3): Female patient 26 years old presented to the ENT clinic with nasal obstruction and facial asymmetry for 10 months. The radiological findings were in the form

of left atelectatic, completely opacified maxillary sinus, midline nasal septum and normal turbinates.

Case (4): Male patient 28 years old presented to the ophthalmology clinic with enophthalmos and was referred to the ENT clinic after diagnosis of silent sinus syndrome. CT scan shows opaque right maxillary sinus with reduced volume, laterally displaced uncinate process obstructing the infundibulum and abnormally wide middle meatus. The nasal septum, middle turbinate, middle meatus uncinate process and maxillary sinus on the left side were within normal.

Case (5): Female patient 35 years old presented to ophthalmology clinic with diplopia for 6 months. The radiological findings were in the form of opaque right atelectatic maxillary sinus with downward displacement of the right globe (hypoglobus). The nasal septum and turbinates were normal.

Case (6): Male patient 57 years old presented to the ENT clinic with nasal obstruction and facial asymmetry for 1 year. The radiological findings showed left atelectatic, totally opacified maxillary sinus, closed infundibulum, normal nasal septum and turbinates.

Case (7): Male patient 22 years old presented to the ENT clinic with history of nasal obstruction and facial asymmetry for 10 months. The radiological findings showed bilateral concha bullosa with right totally opaque atelectatic maxillary sinus and partially opaque left maxillary sinus with normal size and uncinate process. The uncinate process on the right side was laterally displaced and obstructing the right infundibulum.

Case (8): Female patient 46 years old presented to the ENT clinic with history of nasal obstruction plus facial asymmetry for 12 months. CT scan shows right concha bullosa and left paradoxical middle turbinate (Figure 4). The left maxillary sinus was opaque and atelectatic, while the right maxillary sinus was also opaque but not atelectatic.



Figure (4): CT scan coronal cut (Patient n.8) shows right concha bullosa and left paradoxical middle turbinate, the left maxillary sinus is opaque and atelectatic while the right maxillary sinus is also opaque but not atelectatic.

Case (9): Male patient 31 years old presented to the ENT clinic with nasal obstruction and mild facial asymmetry for 6 months. The radiological findings were in the form of left atelectatic opacified maxillary sinus and midline nasal septum.

Case (10): Female patient 43 years old presented to ENT clinic with nasal obstruction and mild facial asymmetry for 8 months. The radiological findings were in the form of right atelectatic opacified maxillary sinus, midline nasal septum and bilateral concha bullosa.

Case (11): Male patient 50 years old presented to the ENT clinic with history of nasal obstruction, headache and facial asymmetry for 24 months. CT scan shows mild septal deviation to the right, opaque and atelectatic right maxillary sinus while the left maxillary sinus was normal (Figure 5).

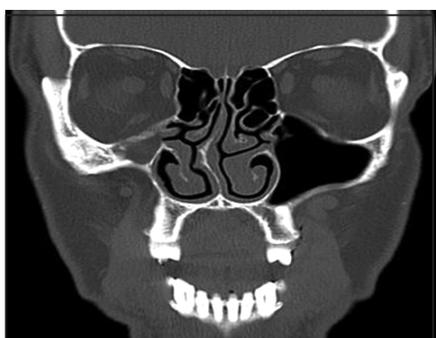


Figure (5) CT coronal cut (Patient n.11), shows opaque and atelectatic right maxillary sinus with normal left sinus.

## Discussion

The term Silent Sinus Syndrome was first used in 1994 by Soparker et al. <sup>(1)</sup> to describe a subgroup of patients with spontaneous unilateral enophthalmos and hypoglobus associated with a small ipsilateral maxillary sinus, but no other symptoms.

It is an uncommon condition probably more familiar to ophthalmologists and otolaryngologists than radiologists. Several anatomical variations can be found associated with this entity. Two cases of this study have marked nasal septal deviation and one case has mild septal deviation, but not every case with nasal septal deviation can develop silent sinus syndrome. Two cases have bilateral concha bullosa but only one side is atelectatic. One case has paradoxical middle turbinate on the atelectatic side, concha bullosa on the non-atelectatic side, and both sides are filled with mucus with obstructed drainage and lack of aeration on both sides. However, only one side was atelectatic. Six cases have within normal septum and turbinates. In all cases the uncinate process was found laterally displaced obstructing the infundibulum. This was the consistent finding that could be detected in all eleven cases. It can explain the maxillary sinus obstruction with no relation to other anatomical variations of the nasal septum and middle turbinates.

Many authors tried to find an explanation to the development and the cause of the negative pressure inside the atelectatic maxillary sinus. Soparkar et al stated that the pathophysiology of silent sinus syndrome remains unclear <sup>(1)</sup>. Initially, it was suggested that a developmentally small sinus with chronic obstructive sinusitis was the cause. The acquired nature of this condition, however, is now well recognized. Negative intrasinus pressure has been demonstrated in patients with silent sinus syndrome <sup>(3)</sup>. Obstruction of the sinus ostium is always present, but it is not clear whether this is the cause or the result of sinus wall retraction. Complete obstruction of the mucous membrane-lined sinus resulting in gas resorption and negative pressure formation, in a similar manner to middle ear atelectasis due to Eustachian tube dysfunction, is the most plausible explanation <sup>(10)</sup>.

Given the rarity of silent sinus syndrome and the very high prevalence of maxillary sinus obstruction, however, one has to question this explanation. This theory also fails to offer an explanation for the exclusive involvement of the maxillary sinus. Perhaps, other compounding factors such as trauma or anatomic predisposition play a role.

Roula et al, 2005 stated that childhood nasal trauma and subsequent surgical trauma were likely factors in the later insidious development of silent sinus syndrome.

Subsequent scar contracture or maxillary sinus hypoventilation might then have resulted in the sinus atelectasis, hypoglobus, and enophthalmos typical of silent sinus syndrome<sup>(7)</sup>. Levine and Mitra, 2000, also reported the case of a child who had undergone endoscopic sinus surgery. In this patient, damage to the osteomeatal complex was postulated as the causative factor<sup>(8)</sup>.

The leading theory regarding the pathogenesis of the silent sinus syndrome is based on chronic maxillary sinus obstruction with hypoventilation, a state that eventually causes negative pressure to develop within the sinus. Mucus begins to accumulate and eventually fills the sinus after occlusion of the maxillary infundibulum. The stagnant mucus incites a low-grade inflammatory response within the sinus and causes osteolysis of the sinus walls. The sinus walls, thinned by inflammation, are pulled into the sinus by the negative sinus pressure<sup>(1, 2, 3, 4, 11, 12, 13, 14, 15)</sup>.

It is probable that, regardless of the inciting event, the final common pathway leading to the silent sinus syndrome is obstruction of mucus drainage from the sinus, which leads to a chronic hypoventilated state. Although the exact sequence of events and causal relationship have not been definitively established, negative sinus pressure, osteolysis of the orbital floor, and chronic inflammatory changes have been documented in the literature, both in experimental models and in patients with the silent sinus syndrome<sup>(3, 4, 16, 17, 18, 19)</sup>.

Thomas et al studied the volumetric analysis of the left maxillary sinus in five patients with silent sinus syndrome<sup>(20)</sup>. Endoscopic antrostomy revealed a preoperative volume of 16.85 +/- 0.06 cm<sup>3</sup> and a postoperative volume of 19.56 +/- 0.07 cm<sup>3</sup>. This represented a 16% increase in maxillary sinus volume postoperatively indicating that ostium obstruction is the key of the etiology of atelectatic maxillary sinus.

In conclusion, spontaneous SSS is a rare condition characterized by unilateral enophthalmus and hypoglobus with radiological findings of ipsilateral depression of the orbital floor, reduction in volume and opacification of the maxillary sinus. Pathogenesis of this condition remains unclear. The fixed sign is the laterally displaced uncinat process which could be detected in all our cases. The obstruction of the infundibulum may thus have a direct relation in the development of maxillary sinus atelectasis.

## References

1. Soparkar CN, Patrinely JR, Davidson JK: Silent sinus syndrome: new perspectives? *Ophthalmology* 2004; 111:414-415.
2. Montgomery WW. Mucocele of the maxillary sinus causing enophthalmos. *Eye Ear Nose Throat Monthly*, May 1964; 43:41.
3. Soparkar CN, Patrinely JR, Cuaycong MJ, et al. The silent sinus syndrome: a cause of spontaneous enophthalmos. *Ophthalmology* 1994; 101:772-778.
4. Rose GE, Sandy C, Hallberg L, Moseley I. Clinical and radiologic characteristics of the imploding antrum, or "silent sinus," syndrome. *Ophthalmology* 2003; 110: 1475.
5. Illner A, Davidson HC, Harnsberger HR, et al. The silent sinus syndrome: clinical and radiographic findings. *AJR Am J Roentgenol* 2002; 178:503-506.
6. Brandt M.G, Wright E.D. The silent sinus syndrome is a form of chronic maxillary atelectasis: A systematic review of all reported cases. *Am J of Rhinol*. 2008; 22: 68-73.
7. Roula H, Nafi A, Charly C et al. Silent Sinus Syndrome: An Acquired Condition. *Am J Neuroradiol* 2005; 26: 2390-2392.
8. Levine SB, and Mitra S. Maxillary sinus involution after endoscopic sinus surgery in a child: a case report. *Am J Rhinol* 2000; 14: 7 - 11.
9. Kayhanian H, Babar-Craig RH, Lund VJ. Spontaneous silent sinus syndrome (Imploding Antrum Syndrome). *British rhinologic Society.org.uk/2009*.
10. Gillman GS, Schaitkin BM, May M. Asymptomatic enophthalmos: The silent sinus syndrome. *Am J Rhinol* 1999; 13: 459 - 62.
11. Roach HD, Shuttleworth GN, Powell N. A case of unilateral enophthalmos. *British J Radiol*. 2003; 76, 577-578.
12. Kass ES, Salman S, Montgomery WW. Manometric study of complete ostial occlusion in chronic maxillary atelectasis. *Laryngoscope* 1996; 106: 1255-1258.
13. Hobbs CGL, Saunders MW, Potts MJ. Spontaneous enophthalmos: silent sinus syndrome. *J Laryngol Otol* 2004; 118: 310-312.
14. Boyd JH, Yaffee K, Holds J. Maxillary sinus atelectasis with enophthalmos. *Ann Otol Rhinol Laryngol* 1998; 107:34 - 39.
15. Hunt SM, Tami TA. Sinusitis-induced enophthalmos: The silent sinus syndrome. *Ear Nose Throat J* 2000; 79: 576-581.
16. Wan MK, Francis IC, Carter PR, et al. The spectrum of presentation of silent sinus syndrome. *J Neuroophthalmol* 2000; 20:207-212.
17. Davidson JK, Soparkar CNS, Williams JB, et al. Negative sinus pressure and normal imaging in the silent sinus syndrome. *Arch Ophthalmol* 1999; 117: 1653 -1654.
18. Boyd JH, Yaffee K, Holds J. Maxillary sinus atelectasis with enophthalmos. *Ann Otol Rhinol Laryngol* 1998; 107:34 - 39.
19. Scharf KE, Lawson W, Shapiro JM, et al. Pressure measurements in the normal and occluded rabbit maxillary sinus. *Laryngoscope* 1995; 105: 570-574.
20. Thomas RD, Graham SM, Carter KD, et al.: Management of the orbital floor in silent sinus syndrome. *Am J Rhinol* 2003; 17 (7): 97-100.