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Review Article

Silent Sinus Syndrome: Interesting Computed Tomography and Magnetic Resonance Imaging Findings

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ABSTRACT

Silent sinus syndrome (SSS) is the spontaneous unilateral collapse of the maxillary sinus and orbital floor with complete or partial opacification of the collapsed sinus. The key features in a patient who presents with SSS are painless progressive unilateral maxillary sinus disease in the absence of rhinosinusitis, trauma, or surgery. SSS is a rare disorder but could be under-diagnosed because of a lack of recognition. SSS is characterized by spontaneous and progressive enophthalmos ("sunken" eye-eye recession into the globe) and hypoglobus (globe displaced downward; and a drop in the pupillary level), so it is common for these patients to present first to an ophthalmologist. Although mostly observed in adults, there have been reports of SSS in children. SSS in younger individuals has characteristic clinical and radiologic signs with, in many cases, abnormal intranasal anatomic characteristics on the affected side. SSS should be differentiated from other causes of spontaneous enophthalmos, such as Parry-Romberg syndrome and linear scleroderma. The aim of this report was to alert the reader to the imaging findings in patients with SSS. This syndrome is well recognized by rhinologists and to a lesser extent to ophthalmologists but remains relatively unknown to general radiologists.

Keywords: Silent sinus syndrome, Ophthalmology, Rhinology, Magnetic resonance imaging, Computed tomography

INTRODUCTION

The first cases of enophthalmos caused by pauci-asymptomatic sinus disease were reported by Montgomery in 1964,[1-4] However, it was not until 30 years later that Soparkar *et al.* coined the term silent sinus syndrome (SSS, also known as exploding antrum syndrome) to indicate a condition characterized by spontaneous and unilateral enophthalmos and hypoglobus associated with volume reduction and ipsilateral maxillary sinus retraction without any symptoms of sinusitis or any history of trauma or surgical procedure on the nasal and paranasal cavities.[5-7]

SSS is a relatively rare condition. However, its prevalence is probably underestimated because it is largely ignored as a possible cause of diplopia, enophthalmos, and facial asymmetry.[6,8,9] The average age at presentation is in the fourth decade, with a reported age range of 19–82 years. Reports of SSS in children are rare.[1,3] There is a slight predominance of right-sided disease (in 57% of cases).[3] The condition may be exclusive to nonsmokers.[5] SSS most commonly presents unilaterally, although there are reports of it being bilateral. It occurs exclusively in the maxillary sinus. There is one report of SSS in the frontal sinus.[10]

SSS can be primary or secondary. Primary or spontaneous SSS is idiopathic, whereas secondary SSS may arise from mid-face trauma (including surgery), nose surgery, or chronic rhinosinusitis

and has also been reported in up to 1% of patients with Graves ophthalmopathy after orbital decompression.[3] However, SSS is not well known among general radiologists.[11] The aim of this report was to alert the reader to the pathophysiology and imaging findings on SSS.

PATHOPHYSIOLOGY

The pathophysiology of SSS remains unclear.[11] There are several possible explanations for the disease process. The first theory is a progressive development of a small sinus with chronic obstructive sinusitis as an initial cause.[3] Davidson *et al.*[12] and Kass *et al.*[13] have documented negative maxillary sinus pressure in patients with clinical and radiographic characteristics of SSS, supporting the theory of negative maxillary sinus pressure leading to SSS.

In the first theory, the persistence of negative pressure seems to produce progressive slow maxillary sinus atelectasis.[11] Furthermore, progressive osteomalacia[14] and probably reduced osteoblastic activity[15] contribute to the inward bowing of the sinus bone and osteomalacia in the orbital floor.[16] Lateralization of the uncinated process was present in all cases, suggesting that it plays a key role in the pathogenesis of the disease [Figure 1a].[11,17] This condition is recognized to cause thinning and demineralization of the bony wall through inflammation-based bone catabolism.

Several mechanisms have been proposed for the ostial occlusion, namely, occlusion by inspissated mucus, a lateralized or hypermobile medial infundibular wall or lateralized middle turbinate, mucocele, or nasal polyp occluding the atrium or ostium[10] [Figure 1b].

Rose *et al.*[5] proposed that obstruction of aeration in the maxillary antrum may cause secondary fluid retention due to sub-atmospheric pressure in the sinus, similar to the mechanism suggested for "glue ear." This sub-atmospheric pressure would be responsible for an indrawing of the sinus walls, resulting in depression of the orbital floor [Figure 1c and d]. However, the phenomenon leading to this negative pressure is unclear. The lack of a previous medical history of chronic sinusitis in the patients in the studies supports this concern; closure of the antrum is a common consequence of chronic sinusitis, and re-opening it is a classical treatment.[12,18] Given the rarity of SSS and the very high prevalence of obstruction of the maxillary sinus ostium, this theory has come under question.[3]

Several authors have demonstrated absolute normality of the maxillary sinus walls before clinical and radiological manifestations of SSS, which confirms that it is an acquired condition.[1,6,12] In this enclosed cavity, subsequent gas resorption leads to sub-atmospheric pressure that creates a vacuum, thus creating a suction effect of negative pressure within the maxillary sinus; this vacuum may induce osteopenia, bone remodeling, and retraction of the sinus walls, similar to retraction in the middle ear caused by Eustachian tube dysfunction.[19] Sánchez *et al.* described an image of a "pseudo-pneumo-orbit" that can also be seen due to air trapped under the upper eyelid.[20]

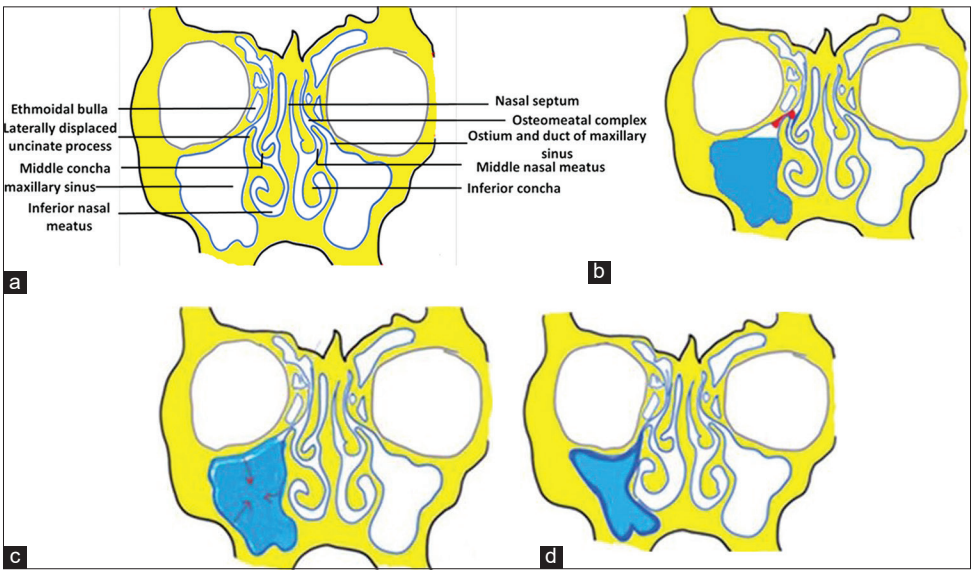

Figure 1: Three stages of development of silent sinus syndrome. (a) The right uncinate process is superiorly and laterally displaced in direct contact with the inferomedial wall of the right orbit. (b) Occlusion of the right maxillary ostium by inspissated mucus or a lateralized hypermobile medial infundibular wall such that air will be reabsorbed from the sinus. (c) Sub-atmospheric pressure would be responsible for an indrawing of the right maxillary sinus walls. (d) Reduction of the size of the opacified maxillary sinus results in enlargement of the right orbit. Case courtesy of Associate Professor Frank Gaillard, Radiopaedia.org, rID: 9447.

The second theory of SSS pathogenesis is inflammatory erosion. Chronic inflammation could induce erosion of the orbital floor. Inflammatory cells can produce cytokines that inhibit the replication of osteoblasts and collagen synthesis, thus favoring the osteopenia process, but the negative pressure in the maxillary sinus must also take part in the mechanism.[3] However, the acquired nature of this condition is now well recognized. Obstruction of the sinus ostium is always present, but it is not clear whether or not this is the cause or result of sinus wall retraction. Complete obstruction of the mucous membrane-lined sinus results in gas resorption. This theory also fails to explain the almost dominant involvement of the maxillary sinus.[1,3]

The third theory involves maxillary sinus hypoplasia. Surprisingly, SSS has been reported to be the result of an infection in a congenitally hypoplastic maxillary sinus; however, one-third of patients has no history of sinus disease in childhood, and SSS can occur in a normal and well-developed maxillary sinus.[3]

Considering the meaning of the thinned or thickened sinus walls in SSS, a report by Hourany *et al*. supports SSS being an acquired condition, given that thickening of the sinus walls is probably related to chronic inflammation and not to underlying developmental hypoplasia.[1]

To the best of our knowledge, there is only one report in the literature of progressive orbital fat herniation into the pterygopalatine fossa.[18] Progressive enophthalmos was likely secondary to entrapment of orbital fat. The hypothesis was that the negative pressure generated by masticatory muscle contraction led to "aspiration" of orbital fat through the defect in the orbital floor and herniation into the temporalis fossa.[18]

CLINICAL FEATURES

The presenting symptom of SSS is spontaneous gradually progressive enophthalmos and hypoglobus over several months. There are no significant sinus symptoms or a history of rhinosinusitis.[10]

Occasionally, the patient complains of symptoms involving the nose and sinuses but is mainly aware of an esthetic and uncomfortable change, such as retraction of the eyelid, deepening of the superior orbital sulcus, or unpleasant flattening of the malar region. Eye movement and visual function are usually unaffected, although diplopia may occur.[9] The average interval between onset of progressive, characteristic orbitopathies, and presentation is 3 months (range 10 days to 2 years). The average amount of enophthalmos at presentation is 2.96 mm (± 0.16 mm) and that of hypoglobus is 2.78 (±0.25 mm).[3] Patients typically present to an ophthalmologist or otorhinolaryngologist. As mentioned earlier, patients may be asymptomatic, are more likely to be female, and may present with the eye retracted into the orbit (with one globe prominent) and displaced downwards.[10,21]

Patients may also present with shrinkage of the anterior aspect of the maxilla, which produces sagging of the soft tissues of the cheek, with a visible change in the profile of the face. The medial and posterolateral walls of the sinus are more likely involved; while the inward displacement of these structures may not be clinically relevant, a lowering of the orbital floor causes enophthalmos.[17]

Findings on nasal endoscopy are either completely normal or of a widened middle meatus on the affected side with inward retraction of the uncinate process.

Histopathological examination of the mucosa of the maxillary sinus consistently shows non-specific chronic inflammatory cell infiltrates; cultured samples of the secretions have never shown viable microorganisms.[17]

Clinically, hypoglobus and spontaneous enophthalmos are non-specific symptoms that may suggest another disease, so additional criteria for SSS include lack of orbital trauma, absence of congenital facial deformity, no history of chronic rhinosinusitis, and no episodes of acute rhinosinusitis in the preceding 6 months.[6,22] The differential diagnosis for SSS includes trauma to the orbit (especially blowout fracture), previous orbital decompression for Graves' orbitopathy, Wegener's granulomatosis, orbital metastasis, human immunodeficiency virus lipodystrophy, and prior orbital radiation therapy. There are also some rare conditions to consider, including orbital fat atrophy, von Recklinghausen's disease (absence of the sphenoid wing), Parry-Romberg syndrome (progressive hemifacial atrophy), linear scleroderma, and pseudoenophthalmos.[3]

IMAGING FINDINGS

Radiologically, chronic sinusitis, and maxillary sinus hypoplasia are the conditions that most frequently enter the differential diagnosis with SSS, whereas trauma and cancer are rare.[16] Radiologists should be able to recognize signs on CT and magnetic resonance imaging (MRI) that is typical of this condition because they may be required to formulate a diagnosis of SSS solely on the basis of these findings and without clear clinical suspicion.[6]

The standard and consistent findings in all patients with SSS are as follows: A normal or mildly inflamed nasal mucosal lining; an enlarged middle meatus (common); and lateral deviation of the uncinate process, which is adherent to the lateral wall with concomitant obstruction of the natural maxillary ostium.

Coronal CT scans provide more anatomical detail concerning bone shape and thickness and a clear morphological picture

that includes the following: Reduction of the volume of the maxillary antrum with retraction of all or most maxillary walls; concomitant compensatory augmentation in ipsilateral orbital volume; complete opacification of the affected sinus; a lateralized uncinate process and expanded middle meatus with variable retraction of the middle turbinate and deviation of the nasal septum; demineralization of the sinus walls; and an expanded retroantral fat pad[1,6,9,10,16] [Figure 2].

MRI, while not mandatory, complements the findings on CT, showing an opacified sinus with mixed-signal contents and diminished volume, prominence of orbital fat, and widening of the retroantral fat pad [Figure 3].

MRI may also be used for diagnosis and follow-up. Therefore, the correct diagnosis of SSS can be obtained using a single method*, and the combination of MR and CT should be restricted to exclude other differential diagnoses before surgical planning.*[6] The eyeball is typically displaced inferiorly, resulting in hypoglobus. The extraocular muscle, most often the inferior rectus muscle, may descend along with the globe.[1]

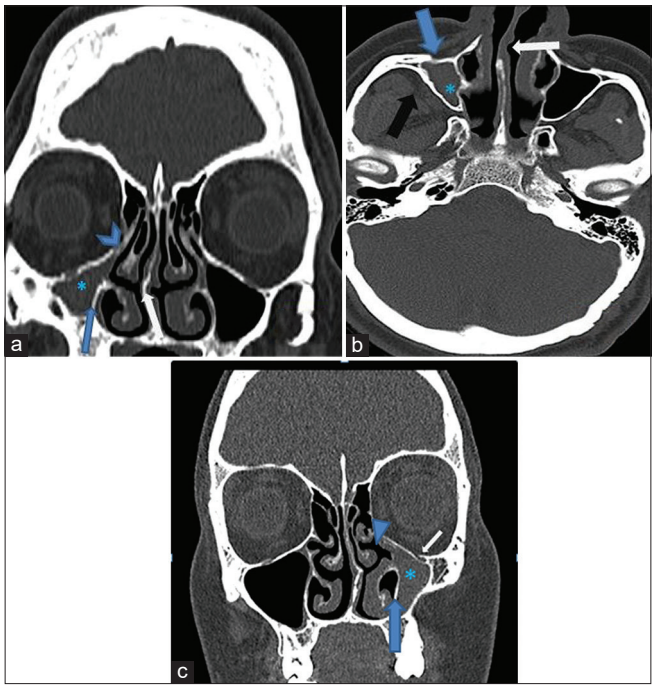

Figure 2: (a) Selected cuts on a coronal computed tomography (CT) scan of the paranasal sinus showing a lateralized right uncinate process (arrowhead), retraction of the medial maxillary sinus wall (blue arrow), retraction of the middle turbinate, and deviation of the nasal septum (white arrow). The right maxillary sinus is completely opacified (*) with a reduction in maxillary sinus volume. (b) Selected cuts on an axial CT scan showing inward retraction of the walls of the right maxillary sinus (blue arrows), complete opacification of the right maxillary sinus (*), with a decreased volume and an expanded right retroantral fat pad (black arrow), and deviation of the nasal septum (white arrow). (c) Selected cuts on a coronal computed tomography (CT) scan of the paranasal sinus showing retraction of the medial maxillary sinus wall (blue arrow) and downwards bowing of the ipsilateral orbit (white arrow). The right maxillary sinus is completely opacified (*) with enlargement of the left medial meatus (arrowhead).

Attenuation or focal absence of the medial sinus wall is observed in more than half of patients; less frequently, there is an attenuation of the posterior, anterior, and lateral walls.[1] CT is essential to obtain details on bone shape and thickness and to confirm that there is no pathological secretion in the ethmoid-maxillary drainage pathway.[17] When SSS is suspected, the maxillary sinus can be normally developed or hypoplastic but is opacified.[6] Axial CT scans show maxillary sinus opacity with inward bowing ("implosion") of the maxillary sinus walls and associated lateral displacement of the middle turbinate [Figure 2]. Maxillary sinus "implosion" indicates retraction of the anterior, posterior, and medial walls into the lumen of the sinus.

The orbital floor is retracted and often thinned, while the other walls may be thinned, normal, or slightly thickened. Orbital floor thickening is also a possibility. There is some debate about whether or not the diagnosis is SSS when CT scans show that the uncinate process is lateralized and orbital volumes are increased without clinical enophthalmos or hypoglobus. Such findings potentially represent early SSS before the development of clinical orbital findings and should be treated as early SSS.[16] Although uncommon, an aerated maxillary sinus in a patient with unexplained enophthalmos, hypoglobus, and maxillary atelectasis should not exclude a diagnosis of SSS.[23]

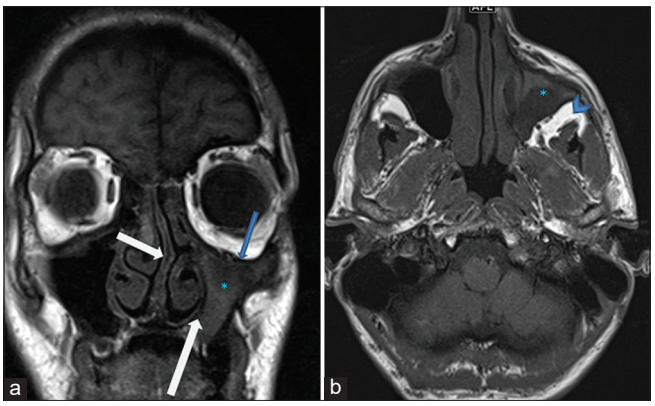

Figure 3: Coronal and axial T1-weighted magnetic resonance images. (a) Total opacification of the left maxillary sinus (*). There is concomitant compensatory augmentation in ipsilateral orbital volume, downwards bowing of the ipsilateral orbital floor (blue arrow), retraction of the medial maxillary sinus wall, and deviation of the nasal septum (white arrow). (b) Reduction of the volume of the left maxillary antrum with retraction of maxillary walls and total opacification of the left maxillary sinus (*) with an expanded left retroantral fat pad (arrowhead).

The distinction between the imaging appearance of a hypoplastic maxillary sinus and SSS is not well understood. The changed architecture of the maxillary sinus seen in SSS should be differentiated from maxillary sinus hypoplasia and chronic maxillary atelectasis.

TREATMENT

The initial management of SSS should be conservative, and not all patients diagnosed on imaging required surgery unless the patient symptomatology progress.[1] The objectives of treatment for SSS are to restore the eye position and orbital floor height, prevent the progression of enophthalmos, restore sinus ventilation and drainage, and avoid infection. These objectives are achieved by single-stage or two-stage surgery.[3,24,25]

A single-stage operation is indicated when severe enophthalmos, diplopia, and/or cosmetic disfigurement are present, as a reconstruction of the orbital floor is mandatory and performing floor reconstruction with sinus surgery reduces morbidity, costs, and hospitalizations.[26] Surgical treatment of SSS is performed by a rhinologist and typically consists of functional endoscopic sinus surgery (FESS) to remove the obstruction and restore positive pressure. If the regression of clinical symptoms is not achieved, the orbit is reconstructed with the aid of implants. The orbital floor may be lifted and reconstructed during the same session or at a later date.[6] Babar-Craig *et al*. advised waiting for at least 6 months after antrostomy, given that only two of 16 patients in their series required reconstruction of the orbit.[8,25]

FESS alone can minimize enophthalmos in some patients.[8] However, most authors recommend FESS as an initial treatment followed by reconstruction with placement of titanium mesh, autogenous nasal septal cartilage, spilled-thickness bone, or auricular concha cartilage onto the floor of the orbit.[7]

Endoscopic antrostomy in combination with orbital floor repair is a single-stage procedure that is preferred in patients with severe enophthalmos.[27,28] A single-stage procedure has the advantages of decreased morbidity, less patient discomfort, and shorter hospitalization time.[27] Post-operative complications, such as implant-related infection, post-operative diplopia, loss of vision, and hematoma, are less common after a single-stage procedure.[27,28]

CONCLUSION

SSS is a rare condition that is often misdiagnosed and has controversial pathology. The clinical diagnosis is based on the gradual onset of enophthalmos and/or hypoglobus in the absence of orbital trauma (including surgery) or the previous symptoms of sinus disease. SSS presents with a constellation of ocular and sinonasal manifestations. Therefore, radiologists, otorhinolaryngologists, and ophthalmologists should be familiar with this entity. The diagnosis is made after complete imaging studies that show a reduction in the volume of the maxillary antrum and retraction of all or most of the maxillary walls. Treatment of the disease consists of restoration of aeration of the maxillary sinus and reconstruction of the orbital floor if needed.

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Declaration of patient consent

Patient's consent not obtained as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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