

The Silent Sinus Syndrome*

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Abstract

Patients with silent sinus syndrome typically present for investigation of facial asymmetry. Unilateral, spontaneous enophthalmos and hypoglobus are the prominent findings at examination. Imaging of the orbit and sinuses characteristically show unilateral maxillary sinus opacification and collapse with inferior bowing of the orbital floor. It has been suggested that SSS is due to hypoventilation of the maxillary sinus secondary to ostial obstruction and sinus atelectasis with chronic negative pressure within the sinus. Treatment involves functional endoscopic sinus surgery for reestablishing a functional drainage passage, and a reconstructive procedure of the floor of the orbit for repairing the hypoglobus and cosmetic deformity. Ophthalmologists, otorhinolaryngologists, and radiologists must be familiarized with this relatively newly reported disease.

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Spontaneous enophthalmos and hypoglobus can be ominous signs. These findings, in the absence of related trauma or surgery, may herald orbital floor disruption secondary to malignancy, osteomyelitis, or systemic inflammatory disease. The term “silent sinus syndrome” was coined by Soparkar et al. [1] in 1994 to describe painless enophthalmos associated with chronic maxillary sinus atelectasis, a phenomenon previously described in 1964 by Montgomery [2]. Since that time, about 100 cases have been reported in the ophthalmic, otolaryngologic, and radiologic literature. Recently, in 2003, the term “imploding antrum syndrome” was also proposed to describe SSS [3]. The condition seems to be more frequent than previously thought, and during the last years several large cases series have been documented in the literature [1,3,4].

The presenting symptom of SSS is spontaneous enophthalmos over the course of several weeks to several months. Most commonly, patients present to the ophthalmologist for evaluation of asymmetry of their eyes, which may be interpreted as exophthalmos of the contralateral eye, ptosis of the affected eye, or enophthalmos of unknown etiology. There are no significant sinus symptoms or history of rhinosinusitis. An orbital/sinus computerized tomography scan will demonstrate unilateral maxillary sinus opacification and collapse with inferior bowing of the orbital floor.

This review summarizes the published reports of SSS, with special emphasis on the clinical presentation, imaging findings, pathogenesis theories, differential diagnosis, and treatment.

Epidemiology

The average age at presentation is the fourth decade (range 19–82 years). There is no gender bias, and there is a slight predominance of right-sided disease (57% of cases) [1,3]. Less than half the patients had a history of symptoms of sinus disease in childhood. It is still unclear if this condition is unusual or is exclusive to non-smokers [4]. Patients do not have a history of ocular disease or injury, and are in general good health. The syndrome occurs exclusively in a unilateral fashion.

Clinical features

Patients typically present to an ophthalmologist or otorhinolaryngologist for investigation of a change in facial appearance, either a “sunken eye” appearance or deepening of the upper lid sulcus, or intermittent vague discomfort around the affected orbit [5,6]. As mentioned earlier, patients present with otherwise asymptomatic, unilateral, spontaneous enophthalmos and hypoglobus (downward position of the globe within the orbit). The duration of progressive enophthalmos before presentation (documented by history or by photography) is usually 3–8 months (range 10 days to 2 years). Enophthalmos and hypoglobus average 3 mm (range 2–5 mm) and

Silent sinus syndrome is a rare cause of sub-acute or chronic spontaneous enophthalmos and hypoglobus

3.4 mm (range 2–6 mm), respectively [1,3,4]. Occasionally, patients may describe exophthalmos of the contralateral uninvolved eye. Other ophthalmologic signs include eyelid retraction, lid lag, and lagophthalmos [7]. Some patients may report an audible clicking sound with blinking. Ocular function is normal, with only rare alteration in ocular motility or muscle imbalance; however, gaze restriction can occur [3]. Visual function is unaffected.

Radiographic findings

Although the diagnosis of SSS is initially made clinically, it is confirmed radiologically, usually by CT scan of the orbits and sinuses [8]. Magnetic resonance imaging may be used to better

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SSS = silent sinus syndrome



Figure 1. Unenhanced CT image in coronal plane reveals opacification of right maxillary sinus and severe atelectasia, with downward displacement of the right orbital floor.



Figure 2. T2-weighted MRI image obtained in coronal plane shows complete opacification of the right maxillary sinus with retraction of orbital floor into sinus lumen.

assess orbital and paranasal sinus soft tissue, but it is inferior to CT for bone visualization [7].

The sinus is fully developed and opacified [9–11]. Attenuation or focal absence of the medial sinus wall is observed in more than half the patients; less frequently, attenuation of the posterior, anterior and lateral walls is found [1]. The maxillary ostium is occluded. Occlusion is usually caused by lateral retraction of the uncinate process with apposition of the uncinate process against the inferomedial aspect of the medial wall [12]. The adjacent middle meatus is correspondingly enlarged with varying degrees of lateral retraction of the middle turbinate. The most characteristic imaging feature of the SSS is the inward retraction of the sinus walls into the sinus lumen with associated decrease in sinus volume [1]. Typically, it is the downward retraction of the orbital floor into the maxillary sinus that creates the eye or facial asymmetry identified by the patient [13]. The eyeball is typically displaced inferiorly, resulting in hypoglobus. The extraocular muscles, most often the inferior rectus muscle, may descend along with the globe [3]. Figures 1 and 2 demonstrate a case of SSS.

Histopathology

Exploration of the maxillary sinus with tissue biopsy shows thickening and edema of the maxillary sinus mucosa with mild, non-specific, chronic, inflammatory cell infiltrates, and reparative changes only in the bone [1,14].

Pathogenesis

The etiology of SSS is still unclear. Several theories have been proposed to explain the cascade of events before the classical radiologic appearance of this condition occurs. The most popular theory, suggested by several authors [1,7–11,15,16], is of hypoventilation of the maxillary sinus secondary to ostial obstruction and sinus atelectasis with chronic negative pressure within the sinus. Several mechanisms have been proposed for the ostial occlusion – namely, occlusion by inspissated mucous, lateralized or hypermobile medial infundibular wall or lateralized middle turbinate [9],

mucocoele or nasal polyp occluding the antrum or ostium [1], inflamed mucosa in the presence or absence of rhinosinusitis, hypoplastic maxillary sinus and antrum leading to decreased ostial diameter resulting in more frequent occlusion [17], and finally, previous nasal intubation resulting in maxillary sinus outflow obstruction and subsequent hypoventilation [18].

Such intra-sinus negative pressure has been recorded in occluded rabbit maxillary sinuses [19] and in humans [20,21]. Negative sinus pressure may cause thinning and inward bowing of the sinus walls, including the orbital floor, resulting in hypoglobus. If the maxillary os reopens, the sinus fluid, which initially may have provided some support for the thin orbital floor, may drain, allowing further depression of the orbital floor and globe. This may account for the rapid presentation in some patients, the variable patency of the os, and the variability of sinus fluid.

Patients usually present in their fourth decade of life

The acquired nature of the SSS has been demonstrated in cases with normal pre-disease imaging of sinuses and orbit [22]. Recently, a new term – “implosion antrum” – has been proposed to describe the pathogenesis of SSS [3,23]. Rose and Lund [23] described a “secondary” enophthalmos after bone-removing orbital decompression for thyroid orbitopathy. The authors suggested that a particularly severe nasal inflammation with marked engorgement of a misdirected turbinate may cause prolonged impairment of antral drainage. Retained secretions may be associated with sub-atmospheric pressures, causing secondary collapse of the antral walls (implosion) until the sinus re-creates as the acute intranasal inflammation settles.

Differential diagnosis

The clinical presentation of spontaneous and otherwise asymptomatic inward and downward sinking of an affected eye is not

specific to SSS, and other causes of enophthalmos must be excluded. Although all patients with SSS have characteristically abnormal orbital imaging with ipsilateral maxillary sinus atelectasis or hypoplasia, an incorrect diagnosis is not infrequent and has been reported in up to 25% of cases [14]. Correct diagnosis of SSS is essential, since the treatment and prognosis differ considerably among the different conditions causing spontaneous enophthalmos. The most frequent causes of erroneous SSS diagnosis are tumor, trauma, congenital facial asymmetry, diffuse facial lipodystrophy, Parry-Romberg syndrome, and linear scleroderma. In these two last entities there is no marked hypoglobus, and periorbital soft tissue or facial contour changes are more frequent than in SSS [14]. Patients may complain of exophthalmos of the contralateral uninvolved eye and only CT of the orbit and paranasal sinuses will demonstrate the typical radiographic findings in the affected side.

Orbital/sinus imaging typically demonstrates unilateral maxillary sinus opacification and collapse with inferior bowing of the orbital floor.

Treatment

Treatment of SSS involves two steps: a) clearing the sinus and reestablishing a functional drainage passage, and b) restoring the normal orbital architecture. Most authors advocate a staged procedure with initial treatment consisting of either a Caldwell-Luc operation or functional endoscopic sinus surgery, followed by a reconstructive procedure with either a trans-conjunctival or sub-ciliary approach and placement of titanium mesh, autogenous nasal septal cartilage, split-thickness bone or auricular concha cartilage onto the floor of the orbit [1,2,9-12,24,25].

The functional endoscopic sinus surgery alone can diminish the enophthalmos in some patients [26]. The orbital floor repair is usually performed in patients with diplopia or severe cosmetic deformity, and in patients with little improvement after functional endoscopic sinus surgery. Some authors have tried simultaneous reconstruction of the orbital floor with aeration of the sinus, without complications such as infection. After surgery, the configuration of the sinus may remain unchanged, improve slightly, or be restored to a near-normal configuration over time [1,2,9-12]. Regardless of the final appearance of the sinus after functional endoscopic sinus surgery, disease progression is arrested without development of further deformity. The reported postoperative complications are very rare and not serious. The enophthalmos typically resolves after surgery. Conservative treatment of vertical diplopia with prisms alone may, in mild selected cases, be an effective alternative to reconstructive surgery [27].

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