Case 1969

The silent sinus syndrome
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Patient: 36 years, female

Clinical History:
A patient presented with spontaneous enophthalmos, hypoglobus, and vertical diplopia on extreme upgaze.

Imaging Findings:
A 36-year-old Caucasian female presented with a progressive deepening of the left superior sulcus palpebralis and sinking of the left eye that had taken place over the past three months (Fig. 1). She denied having been subjected to a trauma, and did not have any inflammation or pain and she admitted that she did occasionally have vertical diplopia on extreme upward gaze. She had a history of chronic allergic rhinitis. A clinical examination done revealed a left enophthalmos measuring 2.5 mm and a hypoglobus sized 2 mm, a deep left superior sulcus and vertical diplopia on extreme upgaze. On the CT scan, the left orbital floor was found to be lowered and thinned. The underlying maxillary sinus was fully opacified and appeared smaller compared to the right one, with an inward retraction of the sinus walls. The middle meatus adjacent to the affected sinus was expanded, relative to the non-affected one (Fig. 2). The same findings were detected on MRI (Fig. 3). Under general anesthesia, the left orbital floor was explored via a subciliary incision, revealing a very thin, translucent orbital floor. A Teflon ® orbital implant was inserted to restore the orbital volume. The maxillary sinus was then explored via a Caldwell-Luc approach. The antrum was filled with a non-purulent, stringy mucoid material without the presence of a mucocele or grossly inflamed antral mucosa (Fig. 4). The patient achieved a satisfactory correction of the enophthalmos and hypoglobus and remained symptom-free three years later. Sinus cultures were found to be negative, and the cytology revealed the presence of an acellular material. A histological examination of the antral mucosa showed minimal inflammatory changes. A postoperative Magnetic Resonance Imaging (MRI) showed the orbital implant in situ (Fig. 5).

Discussion:
It is known that enophthalmos and hypoglobus usually result from a trauma to the orbital area and especially from blow-out fractures of the orbital floor. Spontaneous enophthalmos without a prior history of trauma is rare and was originally reported by Montgomery in 1964 (1). He presented two patients in whom the development of enophthalmos was associated with the presence of mucoceles in the maxillary sinus. This opposed to the fact that mucoceles are frequently present with exophthalmos, as expanding, bone-destructive lesions in the paranasal sinuses. The suspected cause of the enophthalmos was thinning of the orbital floor. Wilkins and Kulwin in1981 reported the effect of chronic maxillary sinusitis in the development of spontaneous enophthalmos. They studied five patients in whom asymptomatic enophthalmos appeared to develop after a long period of chronic nasal sinusitis with outflow obstruction. The development of asymptomatic spontaneous enophthalmos associated with either the presence of maxillary mucoceles or chronic sinusitis was reported infrequently by several authors (2). Although until 1990 the syndrome was practically unknown with only a few cases reported in the literature, an increased number of
cases have recently been published. Soparkar et al. (3), in an effort to clarify the etiology of this peculiar syndrome, presented the cases of 19 patients with spontaneous enophthalmos and associated asymptomatic maxillary sinus disease and reviewed the literature. They proposed the term “Silent Sinus Syndrome”, emphasizing the asymptomatic pre-existing maxillary condition as the causative agent for the enophthalmos. Patients with the “silent sinus syndrome” seem to develop enophthalmos differently from those with mucoceles or active sinus infections. In a few rare cases of acute or chronic rhinitis, usually in the presence of a deviated nasal septum, the inflammation of the nasal mucosa can obstruct the nasal ostium. Such an obstruction of the nasal ostium disrupts the communication of air between the maxillary sinus and the nasal cavity. The confinement of air inside the sinus does not necessarily produce infection of the mucosal lining, but results in the development of a negative pressure inside the cavity. The negative pressure leads to a hinning and inward bowing of all sinus walls. The thinned orbital floor cannot support the pressure from the overlying orbital content, which gradually expands in the sinus. This explains how enophthalmos is caused and the appearance of the sinus being “smaller” than the opposite non-affected one. If this negative intra-maxillary pressure remains for long, the mucosal secretory epithelium of the sinus produces mucous secretions that gradually fill the maxillary cavity and equalize the negative pressure. This explains the presence of a sterile acellular thick mucosal fluid in the sinuses, as in our patient. The causative agent for the development of enophthalmos in cases of maxillary mucoceles or chronic sinus infection seems to be the dissolution of the orbital floor due to a positive pressure inside the sinus cavity (mucocele) or to bone resorption induced by chronic inflammation (sinusitis). This, along with a temporary obstruction of the sinus ostium, increases the pressure inside the sinus cavity, which eventually results in osseous resorption, especially in areas of relative bone weakness as in the case of the orbital floor, which is the weakest bony wall of the maxillary sinus. When the obstruction of the sinus ostium resolves, the orbital content can expand into the sinus through the thinned orbital floor. The process does not occur immediately, as in a blowout fracture, but may take several weeks or months to develop. This explains the occurrence of spontaneous enophthalmos without associated symptomatology from either the infraorbital nerve or the extraocular muscles. In these cases, the thickness of the remaining sinus walls may be increased (as in chronic maxillary sinusitis), and the sinus volume may be decreased (as in sinusitis) or expanded (as in mucoceles). These conditions are also associated with a long history of symptoms of congestion, facial pain, headache, etc. It is therefore safe to speculate that not all patients with spontaneous enophthalmos and maxillary chronic sinusitis or mucoceles represent the spectrum of the same disease. Patients suffering from the “silent sinus syndrome” have a distinct and characteristic clinical and radiological picture, and an entirely different pathogenetic mechanism of development of enophthalmos. The existence of the “sunken-in” appearance of the sinus and the presence of a thick acellular mucoid fluid should be added to the description of the characteristics of the syndrome. A similar imaging appearance may be presented in the case of opacified hypoplastic maxillary sinus, but in this case, the sinus walls' thickness will be normal, and the presence of enophthalmos will not be spontaneous (4). Soparkar and Patrinely (4) further described the exact criteria that characterize the “silent sinus syndrome” as not every patient with enophthalmos and underlying maxillary sinusitis or mucoceles should be considered as having the syndrome. In order to classify a patient with spontaneous enophthalmos as having the “silent sinus syndrome”, the following criteria should exist: (1) the patient usually is in the third or fourth decade of life, (2) the patient has a spontaneous enophthalmos and hypoglobus in the absence of other symptoms, and (3) this condition should be associated with a homolateral “small”, rather than hypoplastic, maxillary sinus with focal areas of bone thinning and loss. The radiological findings should be lower and thinner orbital floor with full opacification of the underlying maxillary sinus which is smaller in size, with an inward retraction of the sinus walls. The middle meatus adjacent to the affected sinus usually is expanded relative to the non-affected one. This is due to the lateral retraction of the uncinate process that causes the obstruction of the maxillary sinus infundibulum (5). The duration of symptoms does not exceed six months and on average the enophthalmos amounts to 3 mm and the hypoglobus to 2 mm.

**Differential Diagnosis List:** Silent sinus syndrome.

**Final Diagnosis:** Silent sinus syndrome.
References:


Figure 1

Description: The left enophthalmos and hypoglobus with a deep left superior sulcus palpebralis.

Origin:
Description: Coronal view: the left orbital floor is lower in position and is thinner. The underlying maxillary sinus is fully occupied by a soft-tissue mass and appears smaller than the right one. The middle meatus adjacent to the affected sinus is expanded, relative to the non-affected one. Origin:
**Description:** Coronal view, bone window. **Origin:**

**Description:** Axial view: the left maxillary sinus is fully occupied by a low density soft-tissue mass, with an inward retraction of the sinus walls. **Origin:**
Figure 3

Description: Coronal view: the left maxillary sinus is smaller in size than that on the right and is fully occupied by a soft-tissue mass. The left orbital floor is lower in position. Origin:

Description: Axial view: the fully occupied left maxillary sinus is smaller in size than that on the right, with an inward retraction of the sinus walls. Origin:
Description: A sagittal view showing the fully occupied left maxillary sinus. Origin:
Description: The antrum was filled with a non-purulent, stringy mucoid material. Origin:
Figure 5

Description: A coronal SE T1-weighted image showing the orbital implant in situ. Origin:

Description: A sagittal SE T1-weighted image showing the orbital implant. Origin: