

CASE REPORT

Silent sinus syndrome – a case report and literature review

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ABSTRACT

BACKGROUND. Silent sinus syndrome is a relatively new and rare condition, with up to 160 cases reported in the literature. Silent sinus syndrome is characterized by unilateral enophthalmos and hypoglobus in association with ipsilateral chronic maxillary atelectasis.

MATERIAL AND METHODS. A 54-year-old woman was referred to our clinic with an 8-month history of spontaneous left eye enophthalmos and hypoglobus. A craniofacial CT scan was performed, showing a hypoplastic left maxillary sinus completely opacified. The patient underwent endoscopic medial meatotomy with left uncinectomy. 1 month postoperatively, the patient had still no rhinologic complains and the left eye enophthalmos has improved.

CONCLUSION. Silent sinus syndrome is a rare pathology. Multidisciplinary collaboration is required (ophthalmologists, radiologists and otorhinolaryngologists) to facilitate a prompt diagnostic and surgical treatment and avoid the onset of orbital complication.

KEYWORDS: silent sinus syndrome, enophthalmos, hypoglobus, medial meatotomy, uncinectomy

INTRODUCTION

Silent sinus syndrome (SSS) is a rare condition, which implies the maxillary sinus and the orbit. It is characterized by unilateral enophthalmos and hypoglobus, due to ipsilateral inward retraction of maxillary walls. Association of the syndrome with the term “silent” was made because the whole process is painless, usually without any clinical rhinologic symptom. Montgomery¹ described for the first time in 1964 painless enophthalmos in association with chronic maxillary atelectasis, but the term “silent sinus syndrome” was coined in 1994 by Soparkar et al.².

CASE REPORT

A 54-year-old woman was referred to our clinic with an 8-month history of spontaneous left eye enophthalmos and hypoglobus. She had no complaints regarding visual acuity, diplopia or extraocular movements of the left eye.

Her medical history records consist in septoplasty (24 years ago) and chronic hepatitis B infection. The clinical examination revealed left nasal septum crest and moderate right inferior turbinate hypertrophy. Ophthalmologic evaluation showed a 2 mm enophthalmos and 2 mm hypoglobus, without any impairment of visual acuity or eye movements.

A craniofacial CT scan was performed with slices of 1mm in the coronal, axial and sagittal planes. The following modifications were noted: hypoplastic left maxillary sinus with its complete opacification; depression of the left orbital floor; lateralization of the uncinate process with secondary blocking of the ostiomeatal complex (Figure 1).

Considering the insidious evolution of symptomatology and the clinical and paraclinical findings, the patient was diagnosed with Silent Sinus Syndrome.

Surgical treatment proposed was endoscopic sinus surgery approach – left antrostomy with uncinectomy. We used 0° and 45° endoscopes and a HD camera. Intraoperatively, the following findings were noted: 1. the absence of maxillary ostium; 2. orbital floor decreased; 3.



Figure 1 Left maxillary sinus hypoplasia. Depression of left orbital floor.

mucocele inside maxillary sinus; 4. hypoplasia of maxillary sinus (Figure 2,3,4). After aspiration of mucocele (Figure 3), the nose was packed for 24 hours. The next day, the patient was discharged without any complication. Postoperatively, we performed suction of the nasal crusts and secretions for 3 days in ambulatory settings and we recommended the patient to use saline nasal lavage for 10 days. One month after surgery the patient had no enophthalmos or hypoglobus.

DISCUSSIONS

Silent sinus syndrome is a relatively new and rare condition, with few cases reported in the literature. A review³ of all reported cases between 1964 and 2006 showed only 105 cases. In the last years, it was observed an increased number of reported cases, which pointed that chronic obstruction of the maxillary sinus may affect the eye⁴. Until September 2012, the number of cases raised up to 160 cases⁵. A possible explanation may be the more easy access to CT scans which facilitated a prompt diagnostic and surgical treatment. Patients with SSS are in the third to fifth decade, without a gender predilection⁶ or predisposi-

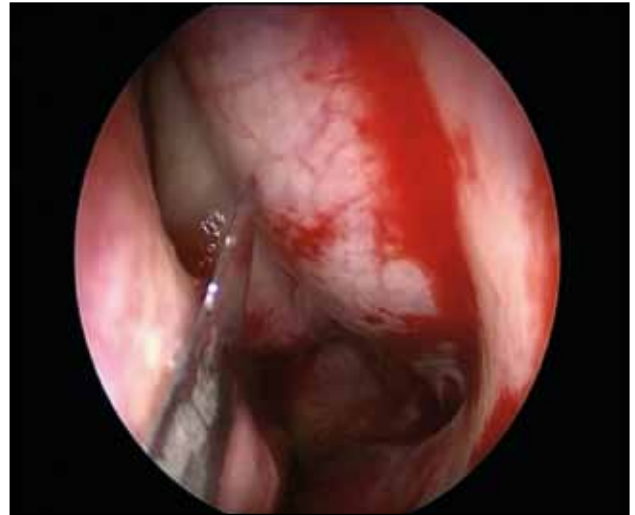


Figure 2 Absence of left maxillary ostium



Figure 3 a. Maxillary mucocele. b. Depression of the orbital floor.

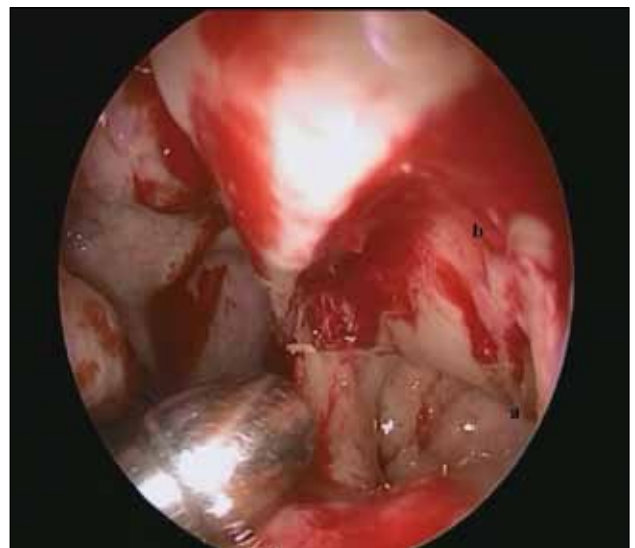


Figure 4 a. Maxillary sinus hypoplasia b. Orbital floor depression

tion for right or left side. It is unknown if this syndrome is unusual or is exclusive to non-smokers⁷.

Usually, patients address to an ENT specialist or an ophthalmologist for some modification in the facial appearance. Their complaints may be a “sunken eye” appearance, deepening of the upper lid sulcus or some discomfort in the affected orbit^{8,9}. Some may describe exophthalmos of the contralateral eye. Visual function and eyeball movement are not altered. Usually, the patient presents with 2-6 mm of hypoglobus and 2-5 mm of enophthalmos¹⁰.

Even if the disease is characterized by unilateral affection of the maxillary sinus, one author reported a bilateral sinus syndrome¹¹.

The main imagistic exploration in SSS is craniofacial CT scan, which confirms the clinical diagnosis¹². If some involvement of the soft tissue is suspected, magnetic resonance may be useful, but this investigation is inferior to CT for bone visualization¹³.

The most characteristic aspect on CT scan is the inward retraction of the maxillary sinus walls into the sinus lumen and decreased sinus volume². It may appear fully or partially opacified, with or without air-fluid level and mucosal thickening. Depression of the orbital floor is the cause that produces the enophthalmos¹⁴, engaging the eyeball, which is displaced inferiorly. The occlusion of the maxillary ostium is observed. This is as a result of the lateral retraction of the uncinat process¹⁵.

Analysis of the tissue sample in the histopathology department shows modifications of the maxillary mucosa: thickening, edema and inflammatory cell infiltrates.

Usually, bacterial cultures are negative.

Until now, the etiology of silent sinus syndrome is unclear. There were a lot of theories like the thickening of mucus, lateralization of the middle turbinate, lateralized or hypermobile medial infundibular wall¹⁶. Anyway, the most frequent theory is of hypoventilation of the maxillary sinus, as a result of ostium blockage and sinus atelectasis with chronic negative pressure inside the sinus. Lateralization of the uncinat process plays a key role in the pathogenesis of the disease⁴.

Clinical symptoms are not pathognomonic of SSS. Other diseases that we should consider first are tumors, trauma, congenital facial asymmetry, diffuse facial lipodystrophy, Parry-Romberg syndrome, linear scleroderma, congenital maxillary sinus hypoplasia and, sometimes, subjective contralateral exophthalmos.

SSS has a slow development and is a rare pathology, but it may alter patients' quality of life in terms of onset of diplopia or aspect of their face. After the diagnosis of SSS is made, surgical treatment is required, which consist in two stages: 1. clearing the

mucocoele from the sinus and reestablishing normal ventilation; 2. restoring the orbital floor. For the first stage, it is recommended the functional endoscopic sinus surgery or Caldwell-Luc operation (in our opinion, this operation is outdated and also dangerous for this disease because of the presence of intrasinus orbital content). If ocular symptoms are mild, functional endoscopic sinus surgery may be enough¹⁷. Orbital floor repairing is required in cases with severe cosmetic deformity, patients with diplopia, or if there was no result or slight improvement after the endoscopic approach. Otherwise, a delay of 18 months after sinus surgery is required¹⁰. Some authors have tried simultaneous reconstruction of the orbital floor with functional endoscopic sinus surgery, with no following complication¹⁸.

CONCLUSIONS

Multidisciplinary collaboration is required because, at first, the patient usually seeks an ophthalmology evaluation for spontaneous onset after several weeks or months of asymmetry of the eyes, with apparent exophthalmos of the contralateral eye or enophthalmos with unknown etiology. After computer tomographic evaluation, the ENT surgeon is the one to restore sinus ventilation, which is the cause of sinus atelectasis with downward bowing of the orbital floor. Ophthalmologists, radiologists and otorhinolaryngologists should be familiar with this condition, in order to facilitate prompt diagnostic and treatment¹⁹. We report a clinical case of SSS with mild enophthalmos and hypoglobus that underwent endoscopic sinus surgery and was free of symptoms one month postoperatively.

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