ORIGINAL RESEARCH

SILENT SINUS SYNDROME
A RETROSPECTIVE REVIEW OF 11 CASES

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ABSTRACT
Objective: The purpose of this study is to describe the clinical and radiological features of Silent Sinus Syndrome (SSS), and to review therapeutic possibilities and their outcomes.

Patients and Methods: Retrospective observational case series in the department of Arthur Vernes Institute between Mars 2007 and November 2012. Clinical records, including ophthalmology and otolaryngology evaluations as well as computed tomography scans and operative reports were carefully examined. A literature review for relevant studies was performed to examine similar cases.

Results: Eleven cases of SSS were identified. Nine men and two women (sex ratio 4.5), aged between 23 and 54 years (mean, 30 years). There was 3 to 4 mm enophthalmos in 10 cases (90.9%), and hypoglobus in all cases, with no effect on visual function. In all 11 cases, the maxillary roof (orbital floor) was drawn downwards, and the one or more walls of the maxilla were concave. In 4 cases septal deviation was present. 8 patients (72.7%) underwent endoscopic sinus surgery, while 3 refused it. Septoplasty was performed in 4 cases (36.3%).

Conclusion: The Silent Sinus Syndrome is a rare entity. It mainly presents as unilateral enophthalmos in younger people and has very characteristic clinical and radiologic signs.
This case series reports our diagnostic and therapeutic experience with this syndrome.

KEYWORDS
Silent-Sinus-Syndrome - Imaging- Endoscopic Surgery

INTRODUCTION
Silent Sinus Syndrome (SSS) consists of an asymptomatic facial asymmetry with enophtalmos due to a chronic atelectasis of one or two maxillary sinuses. Although the clinical diagnosis can be suggested through a morphological examination of the facial symmetry of the patient, radiology remains a key examination to confirm the diagnosis by revealing some characteristics such as a defect of maxillary sinus drainage in the corresponding nasal pathway, opacification of sinus, volume loss caused by retraction of its walls. The syndrome was first described by Montgomery in 1964 referring to chronic atelectasis of the maxillary sinus non-associated with trauma or a surgery [1]. Soparkar and al., in 1994 have noted that this disorder may be the cause of a painless enophtalmos, hence the term "Silent Sinus

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“Silent Sinus Syndrome” [2]. Since then, several cases have been documented and reported in ENT and ophthalmology literature with the publishing of a large series on 22 cases reported by Kass and al in 1997 [3].

In this study, we report 11 cases treated in our tertiary care unit while focusing on clinical manifestations, pathogenic theories and therapeutic means.

MATERIAL ET METHODS

It is a retrospective study searching for patients diagnosed with « Silent Sinus Syndrome », at the ENT service of Arthur Vernes Institute in Paris, for 5 years, from March 2007 to November 2012. Detailed explanations were given to patients in order to get their informed consent in order to include them in the study. All patients accepted to participate in the study or to adhere to different checkups. Clinical inclusion criteria consisted of enophtalmos and/or hypoglobus, facial asymmetry and absence of traumatic and surgical history. Patients with sinus opacification, retraction of ipsilateral maxillary sinus and absence of signs of sinusitis were retained radiologically. Exclusion criteria consisted of a presence of traumatic and surgical history and of clinical signs of rhinosinusitis. Clinical and radiological records of patients were studied retrospectively. A detailed analysis of demographic data, history of functional signs, clinical examination data (exofacial examination and nasal endoscopy), radiological results, surgical care and postoperative clinical results were performed.

The following aspects were detailed radiologically: general appearance and development of maxillary sinus, qualitative volume of sinus, degree of pneumatization, configuration of walls, aspect of infundibulum and uncinate process.

Furthermore, adjacent structures such as the orbit, middle meatus and middle turbinal were also studied. Radiological workup consisted of a CT of the facial bone with coronal and axial slices, osseous and parenchymatous walls. All patients benefited from a CT with thin slices of 3mm on two levels (coronal and axial) with osseous and parenchymatous walls. In 18.1% of cases (n=2), radiological study was completed by a MRI with Gadolinium injection, when suspecting an inflammatory intrasinus phenomena with orbital lesion. Retraction of one or more affected maxillary sinus walls towards sinus lumen was confirmed through scanner in all patients. 100% of patients demonstrated a retraction of orbital floor towards sinus light. All patients (100%) demonstrated a homolateral enlargement of middle meatus.

In 72.7% of cases (n=8), maxillary sinus ostium obstruction by one or many anatomic anomalies, confirmed by endoscopy and/or imaging. Right maxillary sinus was affected in 63.6% of patients (n=7) and the left one in 36.3% of patients (n=4). In 72.2% of cases (n=8), the affected maxillary sinus was completely developed, with extended pneumatization towards a laterally malar eminence and downward alveolar ridge of maxillary. Maxillary infundibulum is obstructed in all patients. In 90.9% of patients (n=10), maxillary sinus was completely opacified.

RESULTS

Of a total of 412 patients followed-up for a maxillary sinusitis proved by CT, 11 cases of SSS were collected. Annual incidence rate in our context was 0.56%. 81.1% of patients (n=9) were men and 18.1% were women (n=2), sex ratio was 4.5. Age varied from 23 to 54, the mean age was 30. 90.9% of patients (n=10) presented with chronic rhinosinusitis symptoms (nasal obstruction, posterior rhinorrhea). 45.5% of cases (n=5) presented with marked ocular asymmetry with an orbital deepening with a progressive evolution. 18.1% of patients (n=2) reported a history of facial or dental pains. No patient had a history of facial trauma. Otorhinolaryngological examination revealed different degrees of enophthalmos in all patients. All patients were directed to an ophthalmological consultation, where examination with Hertel ophthalmometer confirmed enophthalmos of 3 to 4 mm in 90.9% of patients (n=10) and hypoglobus in all patients (downward displacement of globus in the orbit).

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Figure 1a: Silent Sinus Syndrome - CT axial image showing opacification of sinus, retraction of its walls and retraction of uncinate process.

Figure 2a: Silent Sinus Syndrome - CT coronal image showing opacification of sinus, retraction of its walls and retraction of uncinate process.
In 36.3% (n=4), a septal deviation was observed with postero-superior joining to the lateral nasal wall. In 54.4% of patients (n=6); obstruction of the ostium of maxillary was due a lateralization of middle turbinate. In 54.4% of cases (n=5), we observed an enlargement of homolateral middle meatus. In 72.7% of cases (n=8), we noted a lateral retraction of uncinate process with apposition against inferomedial part of orbital wall. In 9.1% of patients (n=1), uncinate process was diminished and slightly developed, causing an occlusion of the infundibulum. In all patients (100%), orbital volume increased due to a downward depression towards the upper side of maxillary sinus. 54.5% (n=6) of all patients demonstrated a retraction of medial sinus wall.45.4% of patients (n=5) had a posterolateral retraction and in 27.2% of patients (n=3), uncinate process was normal.

Figure 2a: Silent Sinus Syndrome - FLAIR MRI sequence showing sinus opacification and orbital floor retraction.

All patients (100%) were treated with local corticotherapy, mucolytic agents and decongestion agents, therefore improving sinus clinical manifestations. With the persistence of symptoms and radiological images after 2 and 3 months of medical treatment; a surgical treatment was proposed which consisted of an endoscopic sinus surgery. This was refused by 27.2% patients (n=8) who never returned for a follow-up. However, 72.7% of patients (n=8) were scheduled to undergo an endoscopic sinus surgery (uncinectomy and middle meatotomy), combined with a septoplasty in 4 patients (36.3%). In 45.5% of patients (n=5), repair of orbital floor by MEDPOR subperiosteal implant (High density porous polyethylene material) was performed by a subciliary route.

In all patients treated surgically, modification of chronic inflammatory phenomena were reported and noted during the post operatory checkup. Only one patient still remains with a facial asymmetry associated with a diplopia and hypoglobus six months after surgery.

DISCUSSION

Silent sinus syndrome is characterized by a painless enophthalmos with involution of maxillary sinus due to an ostial occlusion [4-8]. Silent Sinus syndrome is very similar to type III chronic atelectasis. However, a literature review demonstrated that there is a unanimous accord on clinical criteria of this syndrome: past history of rhinosinusitis, a trauma or tumor exclude the diagnosis, especially if surgery or radiotherapy was performed [2, 3]. These criteria of exclusion are very important especially to differentiate rare SSS from a more frequent chronic atelectasis of maxillary sinus. Real etiology of SSS remains speculative, however, many theories were proposed [9, 10]. The first investigators supposed that chronic inflammatory phenomena in maxillary sinus caused an erosion of orbital floor (lytic osteitis), causing its collapse. However, chronic sinusitis in the absence of osteomyelitis is usually associated with an osteoblastic osteitis, increasing the density and thickness of osseous walls due to hyperemia and inflammation [11, 12]. Since orbital floor becomes thin in silent sinus syndrome, chronic sinusitis associated with inflammatory processes cannot alone explain this pathology.

Another theory proposes that development of sinusites in the congenital sinus hypoplasia causes silent sinus syndrome. On the other hand, there are numerous cases of silent sinus syndrome reported in literature in which imaging was realized before the development of pathology and which reports cases of maxillary sinuses with a normal aspect and size [11, 12]. This proved that congenital nature all alone cannot explain SSS and that another acquired component is mandatory.

The main etiopathogenic theory is based on chronic obstruction of maxillary sinus with hypoventilation of mucosa, and therefore an accumulation of secretions in sinus [2, 12]. This situation can cause, in time, a negative pressure inside the sinus. After the occlusion of the infundibulum, mucosa thickens and secretions start to accumulate in the sinus. Stagnation of the mucosa will eventually lead to an osteolysis of the walls of the sinus. These walls become progressively thinner due to inflammatory reactions and more susceptible to the retraction towards sinus light.

Clinically, it is usually an adult who consults for a painless enophthalmos associated with a facial asymmetry, hypoglobus and diploplia [7, 13, 14]. Average age is 40 with extremes from 19 to 82 years old [3, 7] which is consistent with our work. Unlike our group, there is no gender predominance. The right side is usually affected (65% of cases) [2, 13, 15]. Less than 50% of patients have a history of sinus pathology since childhood. There is no implication related to smoking in this etiopathogenesis of this entity [16]. Visual acuity is not affected [13]. Diagnosis of SSS is essentially clinical but confirmed through orbit and sinus imaging [15]. Generally, radiological evaluation is performed to exclude a subjacent orbital tumor or an orbital floor trauma [17]. It is essential to precise the tridimensional anatomy of the orbitomaxillary complex. It reveals a maxillary opacification (partial or total) with or without liquid levels and mucosal thickening. MRI can be useful in some cases, especially to visualize soft orbital and paranasal tissues; however, it remains confined to osseous evaluation [18]. Biopsies of maxillary sinus tissue show thickening and edema of mucosa with mild, non-specific, chronic, inflammatory cell infiltrates, and reparative changes only in the bone [2, 13, 19].

Differential diagnosis include blowout fractures of the orbital floor, atrophy of soft tissues, primary or secondary
malign tumors, veinal orbital deformities, osseous growth stop due to radiotherapy, congenital etiologies (minor forms of hemifacial microsomia, plagiocephaly, microphthalmia) and pseudo enophthalmos (unilateral blepharoptosis, Claude Bernard syndrome – Horner contralateral exophthalmia, strong contralateral myopia and contralateral palpebral retraction).

SSS can be definitely treated by restoring drainage from the obstructed sinus. The recommended treatment is functional endoscopic sinus surgery with uncinectomy and middle meotomy by widening of the natural maxillary sinus orifice through endoscopic route, secretions aspiration and recovery of physiological functional drainage.

Risk of orbital lesion during an uncinectomy is high due the uncinate process being adherent to orbital lamina. In our case, no post operatory complication was noted. It is essential to perform frontal uncinectomy from behind to front by a retrograde pincer (and not from front to behind) and to look carefully for a security margin of the uncinate process [20, 21]. An exploration with a repair of the orbital floor may be necessary to restore the ocular asymmetry, and may be performed in different ways. In our study, this exploration was considered necessary in almost 50% of patients with implementation of MEDPOR implants. Correction of associated enophthalmos implies the repair of the orbital floor through a transconjunctivitis approach at the same as sinus surgery. However, bibliographic reviews shows that [12, 21, 22], after only sinus surgery, sinus configuration may remain unchanged, or slightly improve. The progression of the disease is stopped without progressive or extensive development of deformity.

After serie of Sivasubramaniam and al [12], all patients with silent sinus syndrome were followed up after sinus surgery and the position of the orbital floor was monitored. In the cases presented, dynamic changes of the orbital floor position were clearly emphasized and were documented in several steps after sinus reventilation. We deduced that elimination of negative pressures within the maxillary sinus after uncinectomy and endoscopic meotomy eliminates the retraction of walls of the sinus and allows a return to a normal state of orbital floor [11].

CONCLUSION

SSS is a rare entity characterized by the retraction of walls of maxillary sinus, causing enophthalmos, sometimes diploplia and depression of the median part of the face. It is also an entity well known within ENT and ophthalmology, but slightly sub diagnosed by radiologists despite its scanographic characteristics. Diagnosis is suspected by the clinic but confirmed by imaging allowing the study of the orbital aspect, maxillary sinus and middle meatus. The most efficient treatment is endonasal surgery with uncinectomy and middle meotomy in order to eliminate obstructive soft tissues and restore normal pressures within the sinus.

Our study endorses data of literature and helps locating clinical signs and especially radiological characteristics not to dismiss in front of any diploptia or enophthalmos without an evident clinical context.

ABBREVIATION

CT
FLAIR MRI
MRJ
SSS

SOURCE OF SUPPORT

Declared none.

COMPETING INTERESTS

The authors declare no competing interests.

AUTHORS’ CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors. Indeed, all the authors have actively participated in the redaction, the revision of the manuscript and provided approval for this final revised version.

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