ORIGINAL CONTRIBUTION

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The silent sinus syndrome: diagnosis and surgical treatment*

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SUMMARY

The silent sinus syndrome consists of painless facial asymmetry characterized by unilateral enophthalmos. Reabsorbed bone with displacement of the orbital floor is a constant finding. It is secondary to chronic maxillary sinus atelectasis. The onset of symptoms is usually slightly progressive but can be brisk due to sudden collapse of the orbital thin bony floor. The diagnosis is suggested by clinical findings including endonasal endoscopic examination and confirmed on the basis of computed tomography and magnetic resonance imaging. The restitution treatment of the silent sinus syndrome involves functional endoscopic sinus surgery and plastic reconstruction of the floor of the orbit via transconjunctival approach; an additional vestibular incision may be necessary to treat the malar region. Four cases of this rare and therefore relatively unknown disease are fully discussed.

Key words: Silent sinus syndrome, maxillary sinus, enopthalmos, diplopia.

INTRODUCTION

The silent sinus syndrome (SSS) signifies a painless involution of maxillary sinus after occlusion of the infundibulum with associated enophthalmos as opposed to simple chronic maxillary sinus atelectasis (CMA). This condition appears to be always unilateral. The term SSS was coined by Soparkar et al. [(1)] in 1994 to describe painless enophthalmos associated with CMA. Similar clinical features have been described previously by Mongomery in 1964 [(2)]. Since that time no more than 100 cases have been reported in literature [(3)]. Recently there has been an increasing number of cases reported, suggesting that chronic obstruction of the maxillary sinus eventually affects the eye or can cause facial deformities. This condition seems to be more frequent than previously thought or simply it is more likely that it is being recognized and effectively treated as a result of the spread of computed tomography (CT) sinus imaging and endoscopic technology. The presenting symptom is spontaneous one-sided enophthalmos progressing over the course of several months [(4)] but faster onset of the phenomenon has also been reported [(5)]. Most commonly the patient presents to the ophthalmologist complaining about orbital asymmetry. Occasionally the patient complains of nose and sinuses symptoms but mainly he or she is aware of some aesthetic and uncomfortable change in appearance like eyelid retraction, superior orbital sulcus deepening or unpleasant flattening of the malar region. Eye movements and visual function are usually unaffected, although diplopia may occur [(6)]. These findings cause the ENT specialist or ophthalmologist to request a CT scan of the orbit and sinuses that demonstrates unilateral maxillary sinus opacification and collapse, with inferior bowing of the orbital floor. Restitution is provided by grafting of the orbital floor and sinus ostia rehabilitation [(3)]. The aim of this paper is to present a clear definition of the SSS diagnostic criteria; we also discuss the pathophysiology of the condition. Four cases of this uncommon and therefore relatively unknown disease are reported including their clinical features, imaging findings, curative surgical treatments and outcomes.

MATERIALS AND METHODS

From 2002 to 2007 four patients were admitted at our institution, who, after thorough examination of the clinical charts and imaging, fulfilled the diagnostic criteria for SSS. Clinical evaluation of each patient included demographic data, clinical presentation and physical examination with office nasal endoscopy. CT scan and magnetic resonance (MR) imaging of the orbits and sinuses was performed on all the patients. An accurate assessment of the maxillary sinus and the orbit with regard to sinus development, sinus volume, degree of aeration, wall configuration and appearance of the sinus infundibulum were carried out. Assessment of adjacent ethmoidal structures within the middle meatus as well as uncinate process and middle turbinate was also performed. The inclusion criteria were:

- Spontaneous enophthalmos secondary to inferior bowing of osteopenic thin orbital floor in the presence of a fully developed opacified maxillary sinus.
- Absence of major sinus pathology such as nasal polyps, fungus ball or acute rhinosinusitis in the last 6 months.

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- Absence of previous trauma, surgery or other causes of acquired enophthalmos.
- Absence of congenital facial deformity [1,3,4,6-9].

The four patients regarded as having SSS received combined surgical treatments, with functional endoscopic sinus surgery (FESS) to re-establish the physiologic drainage pathway and ventilate the maxillary sinus and concurrent plastic surgery reconstruction of the floor of the orbit and malar region to treat enophthalmos and facial deformity.

CASE REPORTS

Case 1.

A 38-year-old woman with history of mild nasal obstruction due to septal deviation underwent a nasal packing procedure for severe epistaxis at the age of 21. She also experienced several recurrent rhinosinusitis and reported occasional use of medication to relieve nasal congestion. Three weeks after delivering she noticed right eye enophthalmos, though no diplopia occurred. She consulted the ophthalmologist who was concerned about hyperthyroidism, then a primary care physician during physical examination, noticed ocular dystopia. This finding caused the doctor to request CT scan and MR to accurately evaluate the bony and soft tissue structures of the face. Finally he sent the patient to our institution with suspected chronic rhinosinusitis. CT showed a right septal spur with displacement of the medial wall of the right maxillary antrum that was completely opacified. Other findings included inferior bowing of the floor of the orbit into the maxillary sinus, lateral and upward drift of the middle turbinate in close contact with the lateral wall, partial opacification of anterior ethmoid. The uncinate process was hardly visible and seemed to be adherent to the medial wall of the orbit hampering the maxillary sinus outflow (Figure 1 A, C). MR findings confirmed prolapse of the inferior aspect of the orbit, shrinking of maxillary antrum secondary to collapse of medial, superior and posterior walls, thickened mucosal lining with mixed signal secretions content (Figure 1 B, D). Her appearance was of an otherwise healthy young woman apart of eye asymmetry with right enophthalmos and hypoglobus. Office nasal endoscopy showed blockage and retraction of the anterior ethmoid, a wide middle meatus with the uncinate process completely adherent to the lateral wall and no pathologic secretions in the nose. She was scheduled for surgery. The nasal septum was straightened with endoscopic removal of the spur. Patency of maxillary sinus ostium was achieved with an endoscopic approach (FESS). During the same surgical procedure, grafting of the floor of the orbit via a transconjunctival approach (TA) restored normal orbital architecture. Two years after the operation the patient is free from orbital or sinus disturbance. Figure 1 (E, F) shows six months postoperative MR. The volume of the antrum is quite similar now to the healthy left side and normal air content is present through the all sinuses. The patient's nasal symptoms have dramatically improved. Symmetry of the face and orbits returned to normal and the patient is fully satisfied with her physical appearance.

Case 2.

A 45-year-old woman complained of right eye dystopia, enophthalmos with deepening of the supratarsal sulcus and right

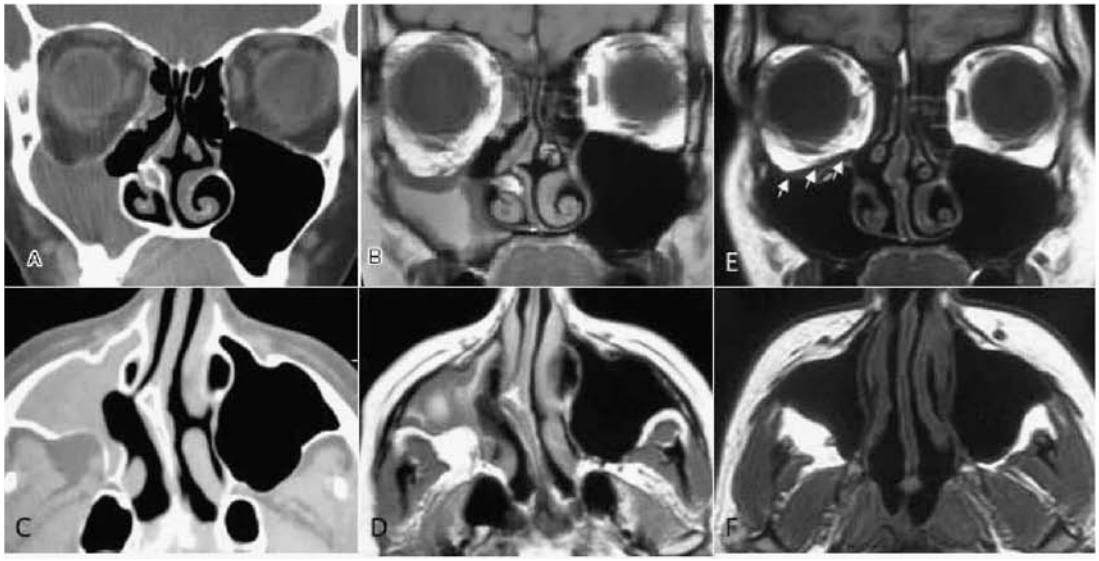

Figure 1. **Preoperative** A) CT in coronal plane shows opacification and atelectasis of the right maxillary sinus with downward displacement of the orbital floor, a septal spur is narrowing the right nasal fossa. The middle turbinate lies on the medial orbital wall. B) T1 weighted MR in coronal plane shows downward displacement of the orbital floor with resulting increase of orbital volume. Maxillary sinus is reduced in volume end completely opacified C) CT in axial plane shows medial and posterior wall inward bowing with enlargement of the retroantral fat pad. D) MR in the axial plane: right maxillary sinus shrinking, retroantral fat enhancement is evident. **Postoperative 6 months follow-up** E) T1 weighted MR in coronal plane shows normal air content of the right maxillary sinus, orbital floor is symmetric with the contra lateral side, septal spur has been removed with normal patency of the nasal fossa and middle meatus. The Medpore graft is visible as void signal intensity replacing the orbital bony floor (white arrows). F) T1 weighted MR in axial plane shows medial and posterior wall recovery, normal air content into the sinus.

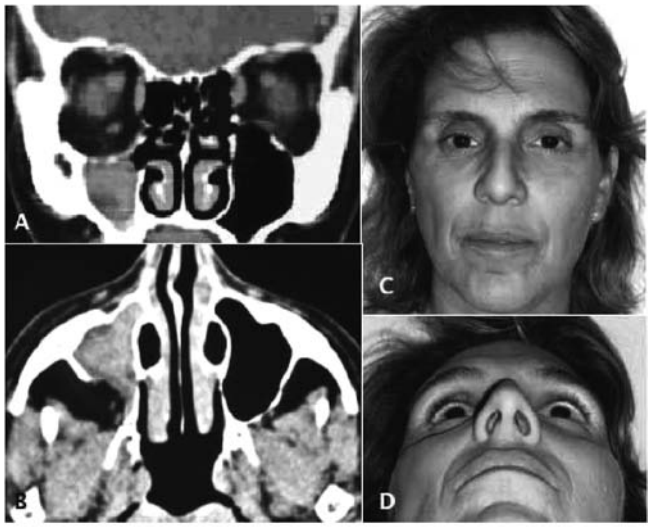

Figure 2a. **Preoperative** A) CT in coronal plane showing opacification of the right maxillary sinus, atelectasis with downward displacement of the orbital floor. B) CT in axial plane shows posterior wall inward bending with enlargement of the retroantral fat pad. C) Frontal view of the patient, notice eyes asymmetry with right enophthalmos. D) Right side downward position of the globe within the orbit "hypoglobus".

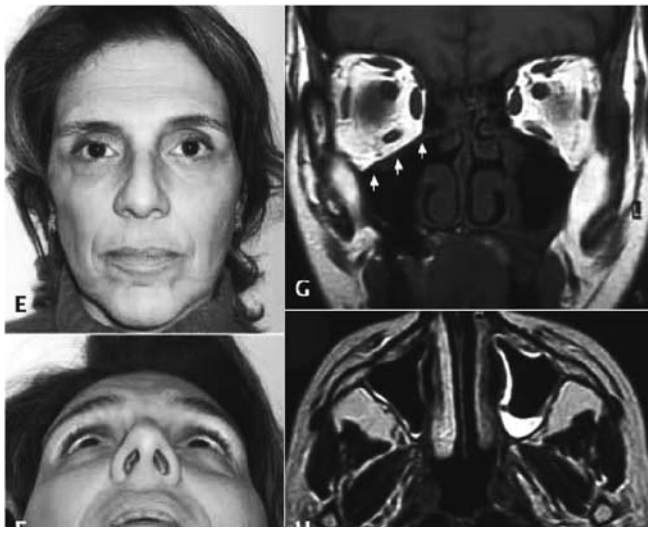

Figure 2b. **Postoperative 6 months follow-up** E) View of the patient showing recovered symmetry with (F) normal projection of the eyes. B) MR in coronal plane showing reconstructed floor of the orbit (white arrows), maxillary sinus with physiologic air content and normal mucosal lining.

upward gaze diplopia. She had experienced acute rhinosinusitis 20 years earlier cured with medications, after which she had no other relevant sino-nasal disturbance. She came to the attention of an ophthalmologist and a dental surgeon, who were concerned about possible sinus pathology, therefore she was sent to CT scan examination. This showed an opacified right maxillary sinus and inferior bowing of the floor of the orbit. Other signs were a notable inward displacement of the posterior-lateral wall, large middle meatus and concavity in the region of the natural ostium with lateral nasal wall deepening into the infundibulum (Figure 2 A, B). Office evaluation showed a certain degree of asymmetry in her face due to right enophthalmos (Figure 2 C) end hypoglobus (Figure 2 D). Nasal endoscopy confirmed the CT findings and showed pale nasal mucosa with slightly fluid secretions. She underwent surgery (FESS plus TA) two months after symptom onset. She fully recovered eye symmetry and sinonasal symptoms went away. Figure 2b E, F shows 6 month follow up control. Notice the restored normal eye projection and filling of the orbital region. MR obtained at the same time (Figure 2b G, H) confirms the clinical findings; see the prosthesis in place (Figure 2b G, white arrows) and normally aerated sinus compartment.

Case 3.

An otherwise healthy 45-year-old man, complaining of chronically blocked nose, became aware of progressive right cheek depression causing visible asymmetry of the face. The physician recommended a CT scan and than sent him to an ENT specialist who, after requesting face and orbit MRI, referred the patient to our institution for further evaluation. The patient's appearance was severely affected with notable right enophthalmos, hypoglobus and sagging of the malar region (Figure 3a). The aesthetic defect was the patient's only complaint; he denied diplopia and any major sinus disturbances or facial pain. CT showed no septal deviation, complete opacification of the right side maxillary sinus with inferior bowing of the floor of the orbit. There was a remarkable reduction to less than 1/5 compared to the contralateral side, in the anterior-posterior diameter of the antrum. This appeared to be mainly a consequence of de-mineralized posterior and anterior walls collapse. Backward displacement of the anterior table of maxillary sinus caused soft tissue retraction in the zygomatic-malar region. The middle turbinate was upright with no other sinus involvement. The uncinate process was not easy to distinguish, being in close contact with the lateral wall (Figure 3b D, E). MR findings showed prolapsed inferior aspect of the orbit, retraction of anterior and posterior walls of the antrum so that the lumen became almost non-existent and filled with secretions. (Figure 3b F, G). Office nasal endoscopy showed no septal deformity, normal mucosal nasal lining, enlarged middle meatus with the uncinate process completely adherent to the lateral wall drawn into the antrum and no secretion out of the ethmoido-maxillary drainage pathway. He was treated with multiple Medpore grafts inserted via transconjunctival and transvestibular incisions. Figure 3c (H, I) shows compete recovery of facial cosmesis after 6 months. On MR control it is possible to appreciate the grafts in place as a void signal (Figure 3c L, M see white arrows).

Case 4.

40-year-old man presents with a one-year history of progressive facial asymmetry and deepening of the right superior orbital sulcus. He also complained of right side nasal obstruction. The

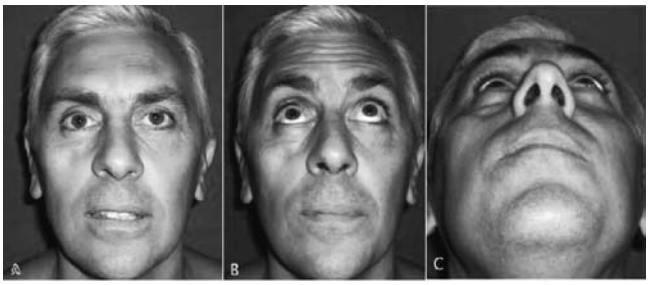

Figure 3a. **Preoperative** A) Frontal view of the patient showing asymmetry of the eyes due to right enophthalmos, (B) evident in the upward gaze C) Right side downward position of the globe within the orbit "hypoglobus" and sagging of the right cheek region.

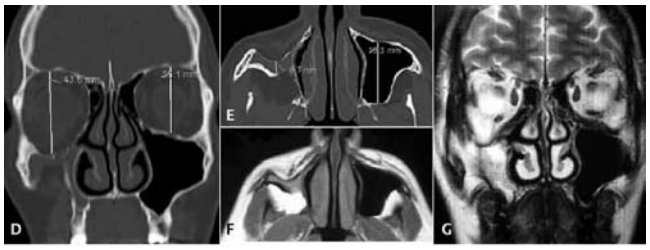

Figure 3b. **Preoperative** D, E) CT in coronal and axial plane show atelectasis of the right maxillary sinus with anterior and posterior walls inward bowing, concavity of the orbital floor with resulting increase of the orbital volume. Measurement of the orbit from frontal bone to infraorbital nerve differs of 8.5 mm from left to right side. The anterior-posterior diameter of the maxillary sinus changes of 28.6 mm F) MR in axial plane shows inward bending of anterior and posterior walls with flattening of the cheek region and enlargement of the retroantral fat pad. Right maxillary sinus collapsed with signal intensity due to thick mucus filling. G) T2 weighted MR in coronal plane showing downward displacement of orbital content.

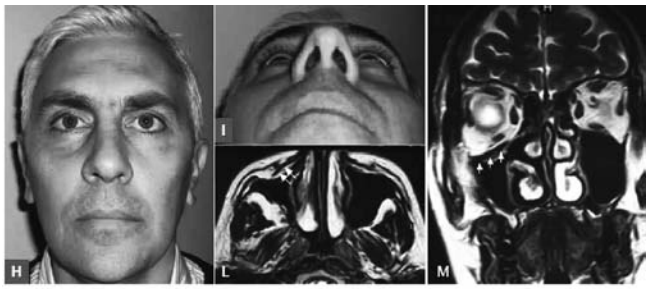

Figure 3c. **Postoperative 6 months follow-up** H) View of the patient showing improved face and eye symmetry. I) Recovered "hypoglobus" with normal eye projection. L) T2 weighted MR in axial plane shows the Medpore graft placed onto the anterior maxillary wall, set in place by microclips to restore cheek convexity (white arrows). M) T2 weighted MR in coronal plane showing reconstructed floor of the orbit; Medpore graft is correct in place and visible as a void signal (white arrows), maxillary sinus with physiologic air content and normal mucosal lining.

ophthalmologist first evaluated the patient suspecting Claude-Bernard-Hornet syndrome, than an ENT specialist requested a CT scan and sent the patient to our institution. CT showed right nasal septal deviation, complete opacification of the right side maxillary sinus that was significantly reduced in volume due to inferior bowing of the floor of the orbit and reduction in the medial-lateral diameter. An increased retroantral fat pad was visible. The right middle meatus was enlarged, the middle turbinate upright with no other sinus involvement, the uncinate process was in close contact with the lateral wall (Figure 4a A, C) MR also showed prolapsed inferior aspect of the orbit, thickened maxillary sinus mucosa and intermediate signal fluid content within the antrum (Figure 4a B, D). The patient's aesthetic appearance was seriously affected with marked right enophthalmos and deepened superior orbital sulcus (Figure 4a E), appearances which caused the patient to experience anxiety and depression. Office nasal endoscopy showed right septal deformity with a spur touching the lateral wall. Normal nasal mucosal lining, enlarged middle meatus with uncinate process completely adherent to the lateral wall, were the relevant features. No pathologic secretions were present. The patient was scheduled for treatment; the septal spur was removed with an endoscopic technique. The infundibulum was freed from the collapsed uncinate process and the maxillary sinus ostium was enlarged. Grafting of the orbital floor via TA and correct alignment of the eyes was accomplished. Figure (4b F) shows stable recovery one year after the operation. MR confirms complete aeration of the sinuses; the Medpore graft is evident maintaining the floor of the right orbit while eye bulbs are symmetric (Figure 4b G, H). The patient has normal nasal breathing and is satisfied about the way he looks with great improvement in his mood.

Preoperative workup included nasal endoscopy, NECT scan done with thin slice, coronal and axial images obtained with bone algorithm and C- MR imaging. Diagnosis has always been based on endoscopic and CT/MR findings, always confirmed at time of surgery. Office nasal endoscopic examination has been thoroughly carried out by means of a 30 degree angled 2.7mm∅ endoscope before and after decongestion of nasal mucosa. This is the best means to clearly identify pathological changes in the nasal lateral wall anatomy and is mandatory for operation planning. The constant findings in all the patients with SSS were:

- Normal or mildly inflamed nasal mucosal lining.
- Enlarged middle meatus.
- Uncinate process completely adherent to the lateral wall obstructing maxillary natural ostium.
- No pathologic secretion in the ethmoido-maxillary drainage pathway.

CT is vital in giving details on bone shape and thickness with clear anatomic picture constantly showing the following features:

- Diminished volume of maxillary antrum with retraction of all or most walls.
- Compensatory augmentation in ipsilateral orbital volume.
- Complete opacification of the affected sinus.
- Lateralized uncinate process and expanded middle meatus with variable retraction of middle turbinate and nasal septal deviation.

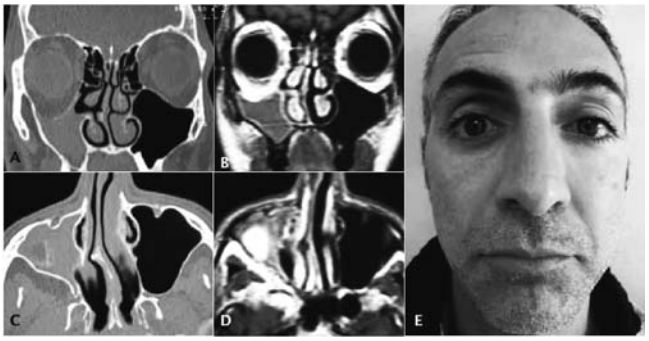

Figure 4a. **Preoperative** A) CT image in coronal plane shows opacity of the right maxillary sinus, downward displacement of the orbital floor with a septal spur narrowing the right nasal fossa. The ethmoid sinus is not involved. B) T1 weighted MR image in coronal plane shows complete opacification of the right maxillary sinus with retraction of the orbital floor into the sinus. C) CT in axial plane shows inward bowing of the posterior wall of the maxillary sinus with enlargement of the retroantral fat pad. D) MR in the axial plane: right maxillary sinus shrinking and retroantral fat enlargement, signal hyperintensity due to orbital fat herniated at the level of maxillary sinus. E) Frontal view of the patient, notice right enophthalmos with deepened superior orbital sulcus.

- Demineralization of sinus walls.
- Expanded retroantral fat pad.

MR, whilst not mandatory, complemented the CT findings showing:

- Opacified sinus with mixed signal contents and diminished volume (T1WI).
- Prominence of orbital fat and widening of the retroantral fat pad (T1 & T2WI).
- Mixed signal central secretions with high signal peripheral thick oedematous mucosal lining within the sinus (T2WI).

All the patients have been treated with FESS to re-establish the physiological drainage pathway and ventilation of maxillary sinus. The usual safe technique to separate the uncinate process from backward to anterior was used. A regular size antrostomy was performed by means of medial displacement and cutting of the edge of uncinate process without traumatising the mucosa. Opening access to the natural maxillary ostium in this way has always offered enough room to aspirate mucous secretions within the atelectasic sinus, and to irrigate with saline sterile solution. Specimens sent for microbiological culture examination have always given negative results suggesting that a non-infective process leads the progression of the disease. Particular attention has been paid preserving the lamina papyracea whilst lifting the posterior boundary of the uncinate process, which was constantly adherent to the lateral nasal wall. This manoeuvre carries a high risk of injury to the orbital content can be safely facilitated by the use of a blunt palpating hook. For all patients reconstructive procedure to the floor of

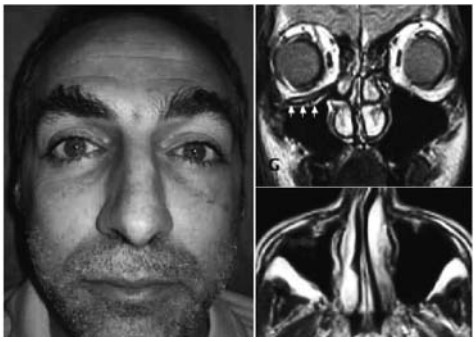

Figure 4b. **Postoperative 1 year follow-up** F) View of the patient with returned eyes symmetry, normal filling of the superior orbital region. G) T1 weighted MR in coronal plane shows the Medpore graft in place (white arrows). H) MR in axial plane shows maxillary sinus with physiologic air content and normal mucosal lining.

the orbit was carried out during the same setting. We used a graft made by a sheet of high-density polyethylene (Medpore®) placed under the periosteal layer onto the floor of the orbit. When the orbital floor has been skeletonized via transconjunctival incision it appeared more or less expanded and eroded. Adherence has been found between sinus mucosa and the periorbita through dehiscent areas of the bone. Sometimes there was fibrous tissue between periorbita and bone. After releasing of the orbital floor has been accomplished from the rim to the inferior orbital fissure, the prosthetic plate is laid in place to restore orbital volume and correct enophthalmos.

The male patient with visible deformity of the malar region (case 3) was additionally treated through intraoral incision at the level of vestibular fornix. The zygoma was approached along the subperiosteal plane with identification and sparing of the infraorbital nerve. Full visualization of the bony defect preceded the positioning of a Medpore® plate stabilized with microscrews to restore symmetry of the face.

RESULTS

All patients were treated successfully with surgery; to date after a follow up range between 12 and 24 months, they are free from ocular disturbances including enophthalmos and satisfied with their aesthetic appearance. Furthermore they have not experienced any nasal discomfort. Thorough examination using nasal endoscopy reveals complete healing with normally patent sinus drainage pathway. Histopathological examination of maxillary sinus mucosa has always shown non-specific chronic inflammatory cell infiltrates; cultured samples of the secretions have never shown viable microorganisms. Follow-up MR obtained six months postoperatively showed normal air content and physiologic mucosal lining with no scar tissue within the maxillary sinus. This confirms that normal shape of the antrum has been reestablished by surgery. No visible signs of remineralization of the dehiscent orbital floor have been observed as yet, therefore long-term follow up is carried on to check for reparative and dimensional changes within the shrunken sinus.

Table 2. Staging of CMA according to Kass et al. [17].

I	Membranous deformity	Lateralized maxillary fontanelle
II	Bony deformity	Inward bowing of one or more osseous walls of maxillary antrum
III	Clinical deformity	Marked deformation of the antral walls, enophthalmos, hypoglobus, midfacial deformity

The synthetic implants have been found to be in place with no signs of inflammatory reaction, scarring or extrusion. Restored stable symmetry of the eyes has been always evident. Table 1 summarizes the demographics, diagnostic and treatment parameters of the four patients of our series.

DISCUSSION

Several studies have suggested that Eustachian tube dysfunction can cause negative air pressure within the middle ear thus leading to tympanic membrane atelectasis; this process has been considered less likely to affect the paranasal sinus. A lot of research has focused on the role of the ostia in equalizing sinus air pressure [10]. On the contrary some authors have described the opposite phenomenon: increased air pressure due to a pneumatic valve mechanism, i.e. air trapping into frontal or maxillary sinus leading to *Pneumosinus dilatans* [11,12,13,14]. The existence of some developmental or acquired anatomical conditions that may provoke a pneumatic valve mechanism within the ostiomeatal complex with sinus one-way air outflow is suggested [15,16]. In all the cases lateralization of the uncinate process was present, suggesting that it plays a key role in the pathogenesis of the disease [15]. This fact could be responsible for the progressive negative pressure within the sinus as well. Resulting inflammation of the mucosa lining the sinus surface with coalescence of mucous secretion could lead to chronic maxillary sinus atelectasis [10,17,18]. This condition is recognized to cause the thinning and demineralization of the bony wall by means of inflammation based immunological bone catabolism [18]. Shrinkage of anterior aspect of the maxilla, which would produce sagging of the cheek soft tissues with visible changing of face profile, is rare. Medial and posterior-lateral walls of the sinus are more likely involved but while inward displacement of these structures is not clinically relevant, a lowering of the orbital floor causes enophthalmos. This condition appears to occur in individuals who make a physical effort that increases intra abdominal pressure such as diaphragm contraction for defecation or delivery. Such motion requires forced brisk inspiration with a closed mouth followed by rapid glottis closure. This is known to blow air out of the sinuses; furthermore arterial blood pressure rises and the central venous pressure increases while compliance decreases. This phenomenon could, at the same time, decrease antral atmospheric pressure and provide a pushing force to the visceral content of the bony orbit. Therefore the hypothesis that, in the presence of CMA with unstable bony boundaries, defecation or delivery effort can cause the maxillary antrum to collapse in an acute way has to be considered [5]. Inferior collapse of the orbital floor is an inconstant feature with CMA but it is of critical relevance. The assumption that anatomically predisposed individuals affected by CMA might develop a collapse within the maxillary sinus with modification of the normal bony boundaries meets with wide acceptance [10,17,19-21] but the exact sequence of events and causal relationship leading to orbital involvement has not been definitively established [8]. We encountered patients with CMA who were apparently not affected by any ocular or aesthetic disorder while imaging showed inward bowing of one or more antral walls, including initial deformation of orbital floor with typical augmentation of intraorbital volume. According to Kass there are three stages of CMA [17]. These patients belong to group II, while onset of ocular disturbances defines group III i.e. SSS.

Do CMA and SSS represent different evolutionary stages of the same disease, or is SSS a different pathological entity from CMA? To answer this question we would need to observe a cohort of patients with CMA without treating them which is not ethically acceptable. Clinical experiences provide enough evidence that the absence of eye and face asymmetry in CMA does not exclude further bone remodeling. On the other hand appropriate surgical treatments have been shown to provide recovery from sinus disease preventing progression of other conditions [22]. The mainstay for the diagnosis of SSS is the ocular deformity and orbital floor displacement; sinus anomalies alone are unlikely to give enough evidence to diagnose the syndrome. CT scan is mandatory to provide reliable findings and MR can be helpful to exclude other pathology [21]. All recent reports show series of adult patients (Table 3). This seems to exclude the appearance of the disease in the pediatric population and reinforce the view that SSS is an acquired condition. The differential diagnosis has to rule out trauma, developmental anomalies and/or systemic disease able to affect the eye unilaterally. Other rare conditions, such as Horner's syndrome, progressive lipodystrophy, and facial hemiatrophy which may also produce enophthalmos has to be taken into account. We did not find endocrinological or immunological problems or other associated disease in our series. Office nasal endoscopic evaluation has a role both in diagnostic assessment and surgical planning. We have been always treated both the sinus and orbital floor during the same surgical setting. However, it should be noted that an implant is not always required at the same time as FESS, as the reinflation of the sinus (as evidenced by the imaging) stops the progression of orbit displacement. Thus, it is possible to adopt a 'wait and see policy' with the possibility of a secondary repair if required, especially in the less severe cases.

We consider that appropriate surgical treatment can arrest the process of bone reabsorbtion by draining the sinus mucous content and establishing physiological ventilation. We are not able in the early postoperative control to check out further re-

Table 1. the demographics, diagnostic and treatment parameters of our series of patients.

Subject ID	**Case 1**	**Case 2**	**Case 3**	**Case 4**
Age	38	45	45	40
Sex	F	F	M	M
Side	R	R	R	R
Presenting symptoms	Enophthalmos, orbital asymmetry.	Enophthalmos, deepening of the supratarsal sulcus, diplopia.	Enophthalmos, face asymmetry with sagging of the malar region.	Enophthalmos, deepening of the supratarsal sulcus with orbital asymmetry.
Presenting service	Ophthalmology	Ophthalmology	ENT	Ophthalmology
Differential diagnosis	Chronic sinusitis, hyperthyroidism	Basedow	Chronic sinusitis	Claude-Bernrd-Horner Sd
Rhinosinusitis	Recurrent episodes	Distant history	None	None
Nasal complaint	Respiratory rigth side nasal obstruction	Nasal constipation, frontal headache	Nasal constipation	Respiratory rigth side nasal obstruction
Facial pain	None	None	None	None
CT/MR	Right septal spur, lateral displacement of the medial wall, thickened mucosal lining with mixed signal secretions content into maxillary antrum, bowing of the floor of the orbit, enlarged retroantral fat pad	Opacification of the maxillary sinus, bowing of the floor of the orbit, deepened supratarsal sulcus, shrinkage of the lateral-medial diameter of the maxilla, enlarged fat pad	Opacification, reduction in the anterior-posterior diameter of the maxillary sinus, bowing of the floor of the orbit, retraction of anterior and posterior walls of the antrum, enlarged retroantral fat pad	Opacification of the maxillary sinus with bowing of the floor of the orbit, deepened supratarsal sulcus, right septal spur, enlarged retroantral fat pad reduction in the a-p diameter of the maxilla
Hertel enophthalmos	4 mm	2 mm	5 mm	6 mm
Endoscopic sinus surgery	Endo Septoplasty. Bilateral etmoidectomy and middle antrostomy	Bilateral antrostomy and etmoidectomy, clearance of the frontal recess	Right uncinectomy and middle antrostomy	Endo Septoplasty. Right uncinectomy and middle antrostomy
Plastic surgery	Transconjunctival approach	Transconjunctival approach	Transconjunctival + vestibular approach	Transconjunctival approach
Implant material	Medpore	Medpore	Medpore	Medpore
Postop Hertel	0	0	1	0
Follow up	24 months	18 months	18 months	12 months

Table 3. Literature review and terminology.

Autor	Terminology	Age	Sex	Side	Signs & symptoms	Treatment			Follow-up	Associated diseases
						Endoscopy	Caldwell	Plast. Surgi		
Facon F, 2006 [23]	SSS	56	F	Right	Toothache, right cheek pain	X			2 months	–
Hens G, 2005 [25]	SSS	25	M	Right	Enophtalmos	X				Chronic sinusitis
	SSS	33	M	Left	Enophtalmos	X				
Hourany R, 2005 [21]	SSS	30	M	Right	Eyes asymmetry, enophthalmos		None		–	Tonsils hypertrophy
Hobbs CGL, 2004 [6]	SSS	41	F	Right	Diplopia, enophthalmos	X			–	Crohn's disease
Castelein S, 2002 [5]	CMA	30	M	Left	Diplopia, enophthalmos	X			–	–
Ong LY, 2003 [19]	SSS	46	F	Left	Diplopia, enophthalmos, depressed left malar region	X		X	–	Multiple sclerosis
Van der Meer JB, 2001 [4]	SSS	38	M	Right	Emianopsia, enophthalmos	X		X	10 months	–
	SSS	38	F	Left	Enophthalmos	X		X	2 months	–
	SSS	45	M	Left	Eyes asymmetry, enophthalmos	X		X	1 year	–
	SSS	47	M	Right	Eyes asymmetry, enophthalmos	X		X	3 years	
Hazan A, 1998 [20]	SSS	34	F	Left	Enophtalmos	X		X	1 year	–
	SSS	32	F	Right	Headache, stuffy nose	X				
Boyd JH, 1998 [10]	CMA	35	F	Right	Entropion, enophthalmos	X		X	3 years	–
	CMA	46	M	Left	Diplopia, enophthalmos	X		X	3 years	Chronic sinusitis, septal deviation
Kass ES, 1997 [17]	CMA	38	F	Right	Facial pain	X			–	Septal deviation
	CMA	38	F	Right	Headache, stuffy nose	X			1 year	Allergy, septal deviation
	CMA	45	M	Right	Diplopia, enophthalmos		X	X	1 year	Obesity
Soparkar C, 1994 [1]	CMA	37			Enophthalmos		X	X	–	–
	CMA	50	F		Malar region sagging	X			–	–
Blackwell KE, 1993 [26]	CMA	41	F		Diplopia	X			–	Allergy, chronic sinusitis
	CMA	42	M		Enophthalmos	X			–	–
Antonelli PJ, 1992 [24]	CMA	32	M	Right	Malar region sagging		X		6 months	Tubercolosis, asthma
	CMA	65	M	Left	Malar region sagging, nasal obstruction		X		6 months	Type II DM, coronary artery disease, prostate cancer

modeling of maxillary sinus boundary apart from which obtained at the time of surgery. We are looking forward to follow reparative phenomena in the long term. We recommend the functional endoscopic approach performing uncinectomy and enlargement of the maxillary ostium, trying not to leave bare bone after mucosal stripping or cause injury to the very thin medial orbital wall. Carefully use of blunt hooks and micro-cutting instruments will allow this practice. Transconjunctival incision has proven to be safe to reach the floor of the orbit even in the presence of thin or dehiscent bone. The operation is well tolerated by the patients and we encountered a high degree of compliance thanks to the avoidance of scar, lack of postoperative pain or bleeding.

CONCLUSION

Some anatomical conditions, such as a large floppy uncinate process closely adjacent to the lateral nasal wall, possibly associated with septum deviation, may provoke a one-way air outflow pneumatic valve mechanism within the ostiomeatal complex leading to atelectasis of the maxillary sinus. SSS seems to be part to the natural history of CMA, with the orbital floor implosion representing a pathological continuum due to demineralization of the bony antral walls. This phenomenon could be elicited by some physiological respiratory actions which produce negative pressure in the sinuses. Absence of ocular symptoms in CMA is not a negative predicting factor for the development of facial or eye deformity. Further studies are necessary in order to understand why some patients develop enophthalmos and/or hypoglobus, as opposed to isolated CMA. The differential diagnosis should include other causes of enophthalmos such as congenital facial asymmetry and trauma. FESS is the gold standard treatment to arrest the progression of the disease, while reconstructive treatments are usually needed to reestablish facial and eyes symmetry. Change of the shape of the maxillary sinus obtained with surgery are stable, as well as aesthetic results; long term thorough observation is expected to show some degree of orbital repair due to newly deposited bone eventually following sinus reinflation.

REFERENCES

1. Soparkar CNS, Patrinely JR, Cuaycong MJ, et al. The silent sinus syndrome: a rare cause of spontaneous enophthalmos. Ophthalmol 1994; 101: 772-778.
2. Montgomery WW: Mucocele of the maxillary sinus causing enophthalmos. Eye Ear Nose Throat Mon 1964; 43: 41-44.
3. Monos T, Levy J, Lifshitz T, Puterman M. The silent sinus syndrome. IMAJ 2005; 7: 333-335.
4. Vander Meer JB, Harris G, Toohill RJ, Smith TL. The silent sinus syndrome. A case series and literature review. Laryngoscope. 2001; 111: 975-978.
5. Castelein S, Cohen M, Ayache D, Kalp P. Atelectasis of the maxillary sinus: report of a case of acute onset. Rev Laryngol Otol Rhinol 2002; 123: 99-102.
6. Hobbs CGL, Saunders W, Potts MJ. Spontaneous enophthalmos: silent sinus syndrome. J Laryngol Otol 2004; 118: 310-312.
7. Buono LM. The silent sinus syndrome: maxillary sinus atelectasis with enophthalmos and hypoglobus. Curr Opin Ophthalmol 2004; 15: 486-489.
8. Illner A, Davidson HC, Harnsberger HR, Hoffman J. The silent sinus syndrome: Clinical and radiographic findings. Am J Roentgenol 2002; 178: 503-506.
9. Rose GE, Sandy C, halberg L, et al. Clinical and radiologic characteristics of the imploding antrum, or "silent sinus" syndrome. Ophthalmol 2003; 110: 811-818.
10. Boyd JH, Yaffee K, Holds J. Maxillary sinus atelectasis with enophthalmos. Ann Otol Rhinol Laryngol 1998; 107: 34-39.
11. Urken ML, Som PM, Lawson W, Edelstein D, Weber AL, Biller HF. Abnormally large frontal sinus. Nomenclature, pathology and symptoms. Laryngoscope 1997; 1987: 606-611.
12. Draf W, Costantinidis J, Weber R, Haque R. Pneumosinus dilatans frontalis. Aetiologie, symptomatik und operationstechnik. Laryngo-Rhino-Otol 1996; 75: 660-664.
13. Kalavagunta S, Reddy KTV. Extensive maxillary sinus pneumatization. Rhinology 2003; 41: 113-117.
14. Trimarchi M, Lombardi D, Tomenzoli D, Farina D, Nicolai P. Pneumosinus dilatans of the maxillary sinus: a case report and review of the literature. Eur Arch Otorhinolaryngol. 2003; 260: 386-389.
15. Bolger WE, Woodruff WW Jr, Moreshead J, Parsons D. Maxillary sinus hypoplasia: classification and description of associated uncinate process hypoplasia. Otolaryngol Head Neck Surg 1990; 103: 759-765.
16. Davidson JK, Soparkar CNS, Williams JB, Patrinely JR. Negative sinus pressure and normal predisease imaging in silent sinus syndrome. Arch Ophthalmol 1999; 117: 1653-1654.
17. Kass ES, Salman S, Montgomery WW. Manometric study of complete ostial occlusion in chronic maxillary atelectasis. Laryngoscope 1996; 106: 1255-1258.
18. Scharf KE, Lawson W, Shapiro JM, Gannon PJ. Pressure measurements in the normal and occluded rabbit maxillary sinus. Laryngoscope 1995; 105: 570-574.
19. Ong LY, McNab AA. The silent sinus syndrome: a case with normal predisease imaging. Orbit 2003; 22: 161-164.
20. Hazan A, LeRoy A, Chevalier E, Benzaken J, Waisberg A, Cymbalista M, Adotti E, Peytral C. Processus atélectasique du sinus maxillaire analyse des stades évolutifs à propos de 4 cas. Ann Otolaryngol Chir Cervicofac 1998; 115: 367-372.
21. Hourany R, Aygun N, Della Santina C, Zinreich SJ. Silent Sinus Syndrome: An acquired condition. Am J Neuroradiol 2005; 26: 2390-2392.
22. Sciarretta V, Pasquini E, Tesei F, Modugno GC, Farneti G. Endoscopic sinus surgery for the treatment of maxillary sinus atelectasis and silent sinus syndrome. J Otolaryngol 2006; 35: 60-63.

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23. Facon F, Eloy P, Brasseur P, Collet S. The Silent Sinus Syndrome. Eur Arch Othorhinolaryngol 2006; 263: 567-571.
24. Antonelli PJ, Duvall AJ III, Teitelbaum SL. Maxillary sinus atelectasis. Ann Otol Rhinol Laryngol 1992; 101: 997-981.
25. Hens G, Hermans R, Jorissen M. Chronic maxillary atelectasis. B-ENT 2005; 1: 25-29.
26. Blackwell KE, Goldberg RA, Calcaterra TC. Atelectasis of the maxillary sinus with enophthalmos and midface depression. Ann Otol Rhinol Laryngol 1993; 102: 429-432.

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