Maxillary silent sinus syndrome: A retrospective review of 18 cases
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Citation

Abstract
Objective: A retrospective review of 18 cases with MSSS studied of 312 consecutive patients with maxillary sinusitis demonstrated by computed tomography of paranasal sinuses from January 2003 to November 2007. Age, sex, clinical manifestations and radiological findings were studied and compared.

Results: All patients showed a orbital floor retraction toward the lumen of the affected sinus in sinus CT scans. All patients presented a homolateral enlarged middle meatus to the affected maxillary sinus. In 77.8% patients, affected maxillary ostium was obstructed by one or more anatomical causes, which was endoscopically and/or radiologically confirmed. In 55.6% patients, maxillary sinus was totally opacified. In all patients, the orbital content volume was increase in the affected sinus side.

Conclusions: MSSS is a well-known and described entity in otolaryngology and ophthalmology, but nevertheless it is a relative and frequently under-diagnosed disorder and poorly regarded by the radiologists, despite their characteristic CT scan findings.

INTRODUCTION
Maxillary Silent Sinus Syndrome (MSSS) consists of asymptomatic facial asymmetry and enophthalmos caused by chronic atelectasis of one or both maxillary sinuses. Even though its clinical diagnosis through the examination of the morphology and symmetry of a patient’s face may be suspected, radiology is the key for diagnostic confirmation, revealing characteristic findings such as drainage obstruction of the maxillary sinus in the corresponding nasal duct, sinus opacification, and decrease of sinus volume caused by the retraction of its walls. The term “silent sinus syndrome” was first described by Montgomery, in 1964 to refer to the chronic atelectasis of the maxillary sinus. Soparkar et al., in 1994, pointed out that this disorder causes enophthalmos with no pain associated.

Ever since, numerous cases have been documented and reported by otorhinolaryngology and ophthalmology literature with the publication of the greatest number of occurrences, 22 cases, reported by Kass et al., in 1997. According to the bibliographic review performed, this entity has not been described in the radiological medical literature, which evidences the little awareness that radiodiagnosis and imaging specialists have regarding this syndrome and its characteristic radiological findings. Although initial diagnosis of MSSS is clinically performed by means of patient’s examination, confirmation of this disorder is radiological. In this publication, we will refer to 16 cases managed in our unit of tertiary attention and we will review the clinical manifestations, theories proposed on its pathogenesis and its therapeutic manage.

MATERIAL AND METHODS
This is a retrospective review of 18 patients diagnosed with SMSS between 1994 and 2007, in our tertiary referral unit and teaching fellowship program in endoscopic sinus surgery in Venezuela. The patients were given a detailed explanation of this study and obtained their written informed consent to include them in this analysis. No patient refuses to participate and fulfill its controls.

3 patients (16.7%) showed radiological findings suggestive of MSSS, without apparent facial asymmetry. These findings were correlated with the otorhinolaryngological evaluation, emphasizing facial examination (symmetry) and nasal endoscopic evaluation. The clinical records and radiological studies of all patients were retrospectively evaluated. Demographic data of the patients, clinical manifestations, physical examination findings, endoscopic studies, treatment, sinus pathology findings and follow up were reviewed. From the radiological point of view, the following was detailed: (a) general appearance and development of
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maxillary sinuses, (b) qualitative volume of the sinus affected, (c) neumatization degree, (d) wall configuration, and (e) appearance of the sinus infundibulum and the uncinate process.

Likewise, adjacent structures, such as the orbit, middle meatus and middle turbinate, were evaluated. All images were reviewed by the authors. 33.3% of patients (n=6) were imaged with 3mm. biplanar (coronal and axial) computed tomography (CT). The tomographic evaluation for the other 66.7% of patients (n=12) was documented in 3 mm. multiplanar (coronal, axial and sagital). All CT scan were performed with algorithm windows for soft- and bone tissues. Superior wall, roof and medial wall retractions were evaluated by coronal CT scan. Anterior and lateral–posterior walls retractions were evaluated by axial CT scan projection.

The CT scan of 11.1% of the patients (n=2) were complemented with magnetic resonance imaging (MRI) with a contrast medium (Gadolinium), which was requested due to the suspicion of sinus inflammatory disease with intraorbital involvement. Clinical, endoscopic, and radiological findings were tabulate and analyzed.

RESULTS

MSSS incidence was 5.76% (n=18) on a total 312 consecutive patients reviewed with maxillary sinusitis demonstrated by sinus CT scan. 61.1% of patients (n=11) studied were males and 38.9% of patients (n=7) were female. Age ranged between 23 and 54 years old, with a mean age of 38.5 years. All the patients presented chronic rhino-nasal symptoms (nasal obstruction, post nasal drip). 27.8% of cases (n=5) exhibited relevant ocular asymmetry and ocular deepening of several-month development. (Figures 1 and 2) All patients had no ocular pain (100%). Nevertheless, 11.1% of patients (n=2) referred diplopia of less than two years development; 22.2% of patients (n=4) referred history of occasional facial and dental pain of several-month development. No patient had a history of previous facial trauma.

The otolaryngological exam revealed different degrees of enophthalmos in all patients. All patients were referred for an ophthalmologic evaluation. In 11.1% of patients (n=2) In all patients (100%) ophthalmological interconsultation with Hertel ophthalmometer confirmed enophthalmos of 3 and 4 mm. and hypoglobus (downward position of the globe within the orbit). Malar depression, upper lid retraction, and deepened upper lid sulcus were variably present. In all cases, endoscopic findings, lateralization of the middle turbinate against lateral nasal wall were observed (Figure 3)

Inward retraction of one or more maxillary walls of the affected sinus into the lumen was tomographically confirmed in all patients. 100% of patients showed a orbital floor retraction toward the lumen of the affected sinus in sinus CT scans. (Figures 4, 6, 9 and 10) All patients (100%) presented a homolateral enlarged middle meatus to the affected maxillary sinus. (Figures 4, 9 and 10)

16.7% of patients (n=3) showed radiological findings suggestive of MSSS, without apparent facial asymmetry. In 77.8% of patients (n=14), affected maxillary ostium was obstructed by one or more anatomical causes, which was endoscopically and/or radiologically confirmed. The right maxillary sinus was involved in 61.1% of patients (n=11) and the left maxillary sinus in 39% patients (n=7). In 66.7% of patients (n=12), the affected maxillary sinus was fully developed, with sinus neumatization extending laterally into the malar eminence and inferiorly into the maxillary alveolar ridge. The maxillary sinus infundibulum was found obstructed in 100% of patients. In 55.6% of patients (n=10), maxillary sinus was totally opacified. (Figures 4, 5, 6, 7, 8, 9 and 10) In 5.6% patient (n=1), the maxillary sinus was found not totally opacified (around 90%).

In 61.1% of patients (n=11) showed homolateral septal deflection with contact posterior-superiorly to the lateral nasal wall. (Figures 4, 6, and 9) 22.2% of patients (n=4), maxillary ostium obstruction was caused by lateralization of the middle turbinate. 11.1% of cases (n=2) showed enlargement of the homolateral middle meatus into the affected sinus. (Figures 4, 9 and 10); 5.6% of cases (n=1), exhibited an accessory ostium in the affected maxillary sinus, with recirculation of secretion.

16.7% of patients (n=3), showed a lateral retraction of the uncinate process with apposition against the inferomedial portion of the orbital wall (Figure 4). In 5.6% of patients (n=1), the uncinate process was thinned and slightly developed, causing occlusion of the infundibulum. In 5.6% of patients (n=1) an accessory ostium was observed in the affected maxillary sinus, with secretion recirculation.

61.1% of patients (n=11) showed an uncinate process displaced into the maxillary sinus and retraction of the orbital floor. (Figures 4, 9 and 10)

11.1% of patients (n=2) were observed a retraction of all the maxillary walls (orbital roof, medial and lateral–posterior). (Figure 5) In all patients (100%), the orbital content volume
was increase in the affected sinus side as a result of downward retraction of the maxillary sinus roof (orbital floor).

50% of patients (n=9) showed a maxillary medial wall retraction. (Figures 4, 5, 6, 8 and 9); 44.5% of patients (n=8) had a lateral– posterior maxillary wall retraction; and in 38.9% patients (n=7) the uncinate process was normal. (Figures 5, 7 and 8).

All patients were treated with topic steroids, mucolitic agents and decongestants, improving their clinical sinonasal manifestations, after the second to third month of treatment with persistent radiological findings suggestive to MSSS. 55.6% of patients (n=10) refuse surgical treatment option and never returned to medical follow-up. Only 44.4% patients (n=8) were scheduled to endoscopic sinus surgery (uncinectomy and middle meatal antrostomy) (Figures 11 and 12).

In all 8 patients treated by endoscopic sinus surgery, chronic inflammatory changes in maxillary sinus involved were documented and all fulfill post.op follow-up controls. A post-op. CT scan was performed three months after surgery in all these cases. Only one patients managed surgically (12.5% n=8) still have a relevant and persistent facial asymmetry associated with diplopia and hypoglobus up to 6 months post op.

**DISCUSSION**

MSSS is characterized by painless enophthalmos with involution of the maxillary sinus after occlusion of its ostium. It is a well-known problem in the field of otolaryngology, but it is rarely reported as such by radiologists in spite of its characteristic radiological findings. As a result of the 6 patients discussed and the different cases documented in the medical literature, the typical patient with silent maxillary sinus syndrome is an adult who consults the otolaryngologist or ophthalmologist with clinical manifestations where the enophthalmos, without associated pain, facial asymmetry, diplopia, and rhinonasal manifestations are present.

Although cases of MSSS without sinusitis symptoms have been described, the presence of drainage alterations of the affected sinus involves variable degrees of inflammatory disease of the sinus and, therefore, variable degrees of sinusopathy. As a result, it is important that the patients with MSSS should be evaluated from otorhinolaringological point of view. Enophthalmos is usually seen in physical examination in variables degrees. Hypoglobus, upper lid retraction, deepened upper-lid sulcus and malar depression are also variably present. Generally, radiological evaluation is performed to exclude underlying orbital tumor or evidence of orbital floor trauma.

The main theory regarding the pathogenesis of SMSS is based on chronic obstruction of the maxillary sinus drainage with hypoventilation of its mucous membrane and inner accumulation of secretions. This situation may cause through time negative pressure inside the sinus. The stagnant mucous is an important stimulus for a low-grade inflammatory response inside the sinus and progressively results in osteolysis of the sinus walls. Sinus walls progressively become thinner due to inflammatory reaction and become susceptible to retraction into the sinus lumen with presence of negative pressure within the sinus.

Scharf et al. pointed out in an experimental study performed with rabbits that when the maxillary sinus ostium was occluded, negative pressure into the sinus was incited to sinus atelectasis due to this pressure, causing bone thinness and osseous changes of the sinonasal walls and their bowing into the sinus. The presence of negative pressures inside atelectatic maxillary sinuses with total obstruction of the maxillary sinus ostium was manometrically registered and reported in a study performed by Kass et al. Some authors have compared and established similarities of the process of sinus wall retraction into the sinus lumen in MSSS cases to the tympanic membrane retraction caused by chronic Eustachian tube obstruction.

Although most patients with MSSS show an idiopathic origin, Levine et al. reported the case of a male child who had a previous maxillary sinus surgery. In this patient, the aggressive surgical trauma on the sinus or the damage to the osteomeatal complex was probably the eventual factor of this disorder. It is likely that, without taking into account the causal event, the common and definite cause leading to MSSS is obstruction of mucous drainage of the sinus, which leads to a chronic hypoventilation and inflammatory state. Although the precise evolution and the causal relationship of MSSS have not been definitely established, a negative pressure occurs within the sinus. Osteolysis of the orbital floor and chronic inflammatory changes of the sinus mucous.
have been documented in the literature, experimental models, and in patients with MSSS.

A similar process may be responsible for the appearance of hypoplastic opacification of the maxillary sinus, an entity with findings similar to those of MSSS and often compared and mistaken in the literature. Hypoplastic opacification of the maxillary sinus may represent a less severe form of MSSS, with a smaller sinus volume and a shorter sinus, with thicker and broader sinus walls as a response of the sinus walls against osteolysis and the effects of inner negative pressure.

This condition must also be suspected in the pediatric population in cases of maxillary sinusitis refractory to treatment. It develops from months to years and it is not associated to previous trauma background. In the presentation, maxillary sinuses may be obvious or occluded, and the sinus may be partially or fully opacified.

Radiological findings of SMSS are characteristic. Radiological findings involving the maxillary sinus, medial meatus, and orbit confirm the diagnosis of this disorder. In general, the sinus is fully developed and opacified. The maxillary infundibulum is occluded. This is usually caused by lateral retraction of the uncinate process with apposition of the uncinate process against the inferomedial aspect of the orbital wall. The adjacent middle meatus correspondingly enlarges with variable degrees of lateral retraction of the middle turbinate. The radiological finding that characterizes SMSS is the retraction of the sinus walls into the sinus lumen with sinus volume loss and increase of orbital volume. In our report, the medial, posterolateral and roof of the sinus or orbital floor are involved in most of the patients. Downward retraction of the orbital floor into the maxillary sinus is responsible for the facial asymmetry and enophthalmos present in these patients.

SMSS may be effectively and definitely treated by restoring mucous drainage from the obstructed sinus. The most effective treatment of MSSS is functional endoscopic sinus surgery with uncinectomy and endoscopic maxillary antrostomy to remove the obstructing soft tissue and to reestablish an isobaric sinus.

Bibliographic reports indicate that, after surgery, the configuration of the sinus may remain unaltered, improve slightly, or restore to a normal configuration through time. In spite of the final appearance of the sinus after functional endoscopic surgery, the progression of the disease stops with no development of progressive or extensive deformity. Some recommendations given by several authors regarding endoscopic surgery on the lateral nasal wall in this setting of altered uncinate process anatomy. Penetration of the lamina papyracea might occur if a vigilant search for the free margin of the lateralized uncinate is not undertaken. Exploration with reconstruction of the orbital floor might be necessary to re-establish ocular symmetry, and it can be performed in a variety of ways. We did not find orbital implantation necessary in this case because the enophthalmos resolved by simply alleviating the inciting vacuum process.

The orbital floor may be restored via a second endonasal endoscopic surgery in the case of patients showing diplopia or persistent, severe cosmetic deformity or in patients exhibiting little improvement after primary functional endoscopic surgery.

CONCLUSIONS

MSSS is a well-known and described entity in otolaryngology and ophthalmology, but nevertheless it is a relative and frequently under-diagnosed disorder and poorly regarded by the radiologists, despite their characteristic signs on CT scan images.

This condition is characterized by painless enophthalmos and facial asymmetry caused by chronic atelectasis of the maxillary sinus.

The most effective treatment of MSSS is functional endoscopic sinus surgery with uncinectomy and endoscopic maxillary antrostomy to remove the obstructing soft tissue and to reestablish a normal pressure inside the sinus.
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Figure 1
Figure 1: Patient female, 50 years old, with a history (3 months) of diplopia, right enophtalmos and facial asymmetry, without facial pain. Dotted-line show the evident facial asymmetry with a right orbital descent.

Figure 2
Figure 2: Frontal view with head in a backward position taken in same patient Dotted-line reveals clearly a right enophtalmos and an important sinking of the right cheek.

Figure 3
Figure 3: Endoscopic view (30º of right nasal cavity of male patient, 41 years old, with facial asymmetry and right enophtalmos with middle turbinate lateralization into the lateral nasal wall.

Figure 4
Figure 4: Coronal CT scan of patient anterior case. The uncinate process is apposed to the inferomedial aspect of the orbital floor, occluding the maxillary sinus ostium (lower arrow). Lateralization of the middle turbinate into the lateral nasal wall with enlargement of the middle meatus wall (asterisk) may also be seen. The orbital floor is retracted into the sinus lumen (upper arrow), showing a downward position of the orbital floor and an increase of the orbital content compared to the contralateral side.
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Figure 5
Figure 5: Axial CT scan of the same case. This study reveals a complete opacification of the right maxillary sinus, with a retraction of anterior wall (upper arrow) and postero-lateral wall (right arrows) and less marked in the maxillary medial wall (left arrow).

Figure 6
Figure 6: Coronal MRI (T2) of the anterior case reveals hypointense image signal in maxillary sinus with retraction of the lateral walls and orbital floor of the right sinus lumen.

Figure 7
Figure 7: Coronal CT scan of patient male, 38 years old, with history of facial asymmetry and right enophtalmos without facial pain. The posterior aspect of the maxillary sinus reveals retraction of the lateral-posterior wall into the sinus lumen (ar)

Figure 8
Figure 8: Coronal MRI (T2) of anterior case reveals hypointense image signal in maxillary sinus with retraction of the postero-lateral (black arrow) and inferior-medial (white arrow) walls into the right maxillary sinus lumen.
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Figure 9
Figure 9: Coronal CT scan of patient with history of facial asymmetry (4 months), enophthalmos and diplopia that reveals a total opacified and well-developed right maxillary sinus with retraction of the orbital floor (upper arrows). The lateral wall is thicker and broader (black star). And the enlargement of the medial wall and middle meatus (asterisk) is evident.

Figure 10
Figure 10: Coronal MRI (T2) of previous case with total opacified right maxillary sinus, which is fully developed. An image corresponding to a possible retention cyst is observed inside the sinus. The white arrow points to a retraction of the orbital floor into the sinus lumen. The morphology of the left uncinate process (U) is normal; however, the right uncinate process is apposed to the inferomedial aspect of the orbital floor, involving the maxillary sinus ostium (white arrow). Medial wall and middle meatus lateralize, causing enlargement of the middle meatus wall (black arrow).

Figure 11
Figure 11: Intra-operative endoscopic view (30°) which demonstrate aspiration of mucus secretion (thick glue) in right maxillary silent sinus during middle meatal antrostomy.

Figure 12
Figure 12: Intra-operative endoscopic view (30°) of contracted right maxillary silent sinus which demonstrates a retraction into the right maxillary sinus lumen of posterolateral lateral maxillary wall (superior and inferior arrows) and orbital floor (dotted arrows).

References
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