ORIGINAL ARTICLE

The Silent Sinus Syndrome: A Clinical Review

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Abstract

Silent sinus syndrome (SSS) is a rare and interesting clinical condition that is associated with spontaneous, painless, unilateral enophthalmos and hypoglobus resulting from downward bowing of the orbital floor, in the absence of any symptomatic sinonasal disease.

It generally affects younger patients between the third and fifth decades of life.

The pathogenesis of silent sinus syndrome is based on chronic maxillary sinus obstruction, related to occlusion of the maxillary infundibulum which results in a hypoventilated sinus and negative pressures. Endoscopic sinus surgery to create a wide maxillary antrostomy with or without orbital floor reconstruction is considered the gold standard treatment of choice.

Keywords: Silent sinus syndrome, imploding antrum syndrome, chronic maxillary atelactasia, enophthalmos, hypoglobus.

INTRODUCTION

Montgomery reported the first case of maxillary sinus opacification and collapse in 1964.[1] However, it was not until thirty years later when Soparker et al coined the term 'silent sinus syndrome' for this phenomenon.[2]

Silent sinus syndrome (SSS) is a rare and interesting clinical condition that is associated with very characteristic clinical and radiological signs. The pathognomonic clinical features are spontaneous, painless, unilateral enophthalmos and hypoglobus resulting form downward bowing of the orbital floor, in the absence of any symptomatic sinonasal disease.

Reports of SSS in the literature are still largely limited to single case reports, with only a small number of larger case series having been described. On closer review, it would seem that of these reported cases, a relatively small number describe true silent sinus syndrome.[3,4] In 2005, Numa et al reported a comprehensive review of 84 previously published cases of silent sinus syndrome.[3] However in 2008, Brandt and Wright published their own systematic review of all reported cases, and they found that only 55 reports contained complete data sets that were suitable for review, and of these just 27 of these cases met their diagnostic criteria for SSS.[4] It would therefore seem that the term silent sinus syndrome is potentially loosely used, and it would be important to differentiate it from other related conditions.

The term chronic maxillary atelactasia (CMA) has often been used interchangeably with silent sinus syndrome, as both have been used to describe the presentation of spontaneous enophthalmos and hypoglobus that occurs in association with a contracted ipsilateral maxillary sinus. Although, these two diagnoses may represent a spectrum of the same clinical entity,[4] there is infact an important differentiation between the two, namely the presence of sinus related symptoms. CMA is classically distinguished by the presence of symptoms analogous to chronic sinusitis.[5,6] However, a small subgroup of patients present in the same way, but do not have any significant history of chronic sinusitis.[4,7] This subgroup have a specific form of chronic maxillary atelactasis referred to as silent sinus syndrome.[2,4,7]

CLINICAL FEATURES

Enophthalmos resulting from spontaneous collapse of the maxillary antrum, which Soparker et al termed SSS,[2] generally affects younger patients between the third and fifth decades of life, with no clear predilection for gender bias.[2,4,8] Although, there have been no clear associations with smoking demonstrated, it is interesting to note that some authors have postulated that there may indeed be a negative association between smoking and the incidence of SSS.[9]

The primary symptom is usually an incidental change in facial appearance, which maybe described as 'sinking down of the eye' or 'drooping of the upper eyelid'. This results from a unilateral enophthalmos and hypoglobus, as a result of expansion of the orbital space due to a downward bowing of the orbital floor,[2] in the absence of any history of ocular disease or orbital trauma. There is usually only a brief

duration of such symptoms, which progresses over a period of months.[2,8] More importantly, there is typically no significant history of sinonasal disease. This characteristic history and the associated clinical signs are highly suggestive of SSS, which can be confirmed by classical radiological findings on a computed tomographic scan (CT).

RADIOLOGICAL FEATURES

The universal radiological feature of SSS is the striking changes that occur in the bony walls of the maxillary antrum. The condition is associated with extensive radiological changes which are characterized by smooth inward bowing of the antral walls on the affected side.[10] The antral roof (orbital floor) is classically depressed, thereby being abnormally concave towards the antrum. There is also frequently concurrent increased concavity of the medial and lateral antral walls of the maxilla.[8] The typical radiological findings results in an overall loss in the volume of the antrum,[2,10] and subsequent increase in the ipsilateral orbital volume. Interestingly, although this condition is primarily a disease of the maxillary sinus, it has also anecdotally been found to be associated with inward collapse of the ipsilateral ethmoid complex in a patient with classical radiological features of SSS.[8]

Furthermore, there is often some notable changes in the texture of the bony walls of the antrum, although these can be quite variable. Soparker et al reported thinning or complete bony resorption of the orbital floor in all patients they reviewed. Similarly, an extensive critical review of all reported cases of silent sinus syndrome also reported evidence of bone demineralization of the roof of the maxillary sinus in some cases.[4] However, Rose et al also reported their own series of 14 patients with SSS. Interestingly, radiological assessment of their patients demonstrated quite varied changes in bone texture of the maxillary walls. They also primarily found the antral roof to be affected, although the other antral walls were also changed. They noted various changes including abnormally thickened, hyperostotic bone, generalized thinning or patchy loss of mineral with focal bone loss.[8] Therefore, although the normal bone mineral content of the antral walls is commonly altered, it would seem that there may not be any consistent changes that are universally identified.

The affected maxillary antrum almost always demonstrates soft tissue changes, which can range from total opacification to patchy soft tissue changes.[8,10] Notably, there is rarely any evidence of widespread sinus disease affecting the other paranasal sinuses. The antral mucosal changes both radiologically and histologically are suggestive of mild, chronic inflammatory changes in the sinus,[8] in keeping with the clinically 'silent' sinus disease. Interestingly, Rose et al, did also recognize the presence of consolidation and collapse of the ipsilateral ethmoid sinus in one patient,[8] which suggests that this disease can rarely also affect the neighboring ethmoid complex.[11]

It is interesting to note that there is frequently some evidence of nasal septal deviation towards the affected side,[2,8,12,13] and also some abnormalities of the ipsilateral middle turbinate.[8,10,11,13]

Interestingly, our own experience has identified some patients who maybe presenting with a possible early form of this clinical spectrum of disease. Anecdotally, we have come across a small subgroup of patients with some sinonasal symptoms, who have incidental radiological features of an 'imploding antrum', without radiological evidence of orbital floor collapse (Fig. 1). In this illustrative case, the CT scan of the paranasal sinus shows obvious collapse and inward retraction or 'implosion' of the left antrum (Figs 1 and 2), without the typical features of the syndrome that is synonomous with this (SSS), namely depression of the antral roof. We therefore postulate that there may be some patients who present with early features of this condition. An awareness of this may potentially facilitate early radiological recognition, which may be useful to identify and treat patients before they potentially go on to develop silent sinus syndrome.

PATHOGENESIS

Soparkar et al used the term maxillary hypoplasia, thereby suggesting an underlying congenital abnormality of the maxilla, which may predispose some individuals to

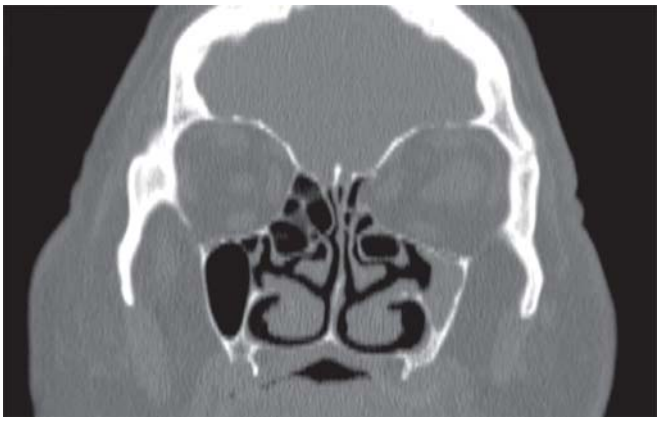

Fig. 1: Coronal section of CT scan paranasal sinuses showing gross inward collapse of the bony walls of the left maxillary antrum. There is marked concavity of the medial wall of the maxillary sinus with evidence of complete bony resorption, with possible hyperostosis of the lateral antral wall. There is a large notable Haller cell on the left, which may contribute to ostial obstruction and subsequent hypoventilation of the maxillary sinus

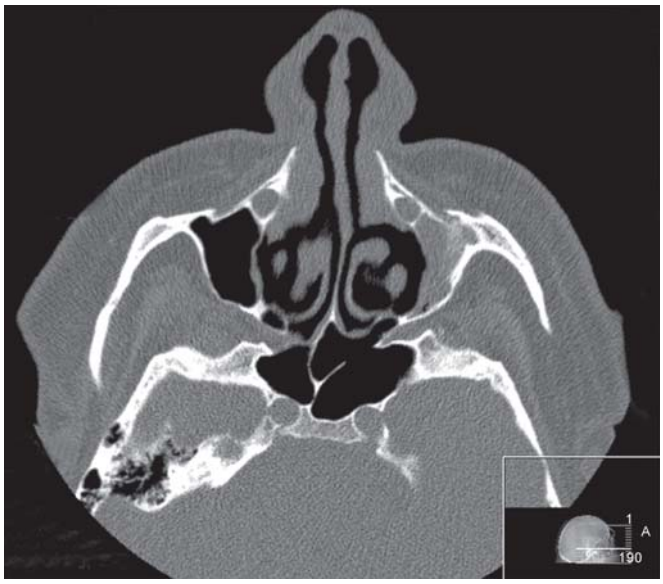

Fig. 2: Axial section of CT paranasal sinuses showing consolidation and contraction left maxillary sinus, and reduced antral volume

developing silent sinus syndrome.[2] However, more recently it has been suggested that the collapse or implosion of the antral walls infact occurs in a previously normal sized and normally developed maxillary sinus.[8,10] The relative sudden onset of the condition is suggestive of an acute event which may precipitate the collapse of the orbital floor, along with widespread implosion of some or all antral walls, thus reflected in the alternative term 'imploding antrum syndrome'.

The leading theory regarding the pathogenesis of silent sinus syndrome is based on chronic maxillary sinus obstruction, related to occlusion of the maxillary infundibulum. This results in a hypoventilated sinus and negative pressures.[1,2,5,11-16]

More recently, Rose and Lund have substantiated this theory, based on similarities between patients with the idiopathic condition and those with an iatrogenic form.[17] They identified a subgroup of patients who developed late enophthalmos resulting from maxillary antral consolidation after undergoing bone removing orbital decompression into the paranasal sinus, for thyroid orbitopathy. These patients had clinical and radiological features resembling the idiopathic form of silent sinus syndrome. They identified that this late onset enopthalmos following bone removing orbital decompression seemed to be the result of occlusion of the ethmoidal infundibulum by prolapsed orbital fat, with secondary obstruction of maxillary antral aeration and maxillary antral consolidation. This resulted in subsequent retention of maxillary sinus secretions and a subatmospheric pressure within the sinus. Longstanding negative pressure was thought in turn to lead to implosion of the other sinus walls. Based on the similarities between this iatrogenic form and idiopathic silent sinus syndrome, they supported the widely accepted theories for the underlying mechanism in the primary syndrome. They suggest a particularly severe episode of nasal inflammation may serve as an acute trigger, which results in marked engorgement of the nasal mucosa, often in a limited nasal airspace due to abnormal nasal anatomy, which may cause prolonged impairment of antral drainage. Mucous accumulates and eventually fills the sinus, which maybe associated with subatmospheric pressures.[17] The stagnant mucous within the sinus is thought to induce a low grade inflammatory response within the sinus and cause osteolysis of the sinus walls, resulting in implosion of the antrum.[6,10]

The suggestion of maxillary sinus obstruction as being the underlying precipitant is also supported by another case of secondary, iatrogenic form of the disease that was described in a child who underwent endoscopic sinus surgery.[18] In this case, damage to the osteomeatal complex was postulated as the causative factor.[18] There are however, a number of different possible causative factors that may lead to ostial obstruction including occlusion by mucous, mucocele or nasal polyp, an abnormally lateralized middle turbinate, lateral retraction of the uncinate process or presence of Haller cells, thus narrowing the maxillary ositum.[7]

The theory of negative sinus pressure is thought to occur in a similar fashion to the mechanisms suggested in glue ear.[19-21] Indeed, this phenomenon of negative sinus pressure has indeed been demonstrated in one patient with silent sinus syndrome.[16] It is therefore now widely accepted that the theory of obstruction of mucous drainage from the sinus, leading to a chronic hypoventilated state as being central to the pathogenesis of silent sinus syndrome.

MANAGEMENT

The treatment of SSS is aimed at correcting the problems that lead to its development as well as re-establishing normal orbital anatomy in some cases. Definitive treatment for silent sinus syndrome is therefore by creating an outlet for mucous drainage from the obstructed sinus.

Historically, surgical treatment was accomplished by performing a Caldwell - Luc procedure and transconjunctival repair of the orbital floor. However, advances in endoscopic surgery have now allowed for endoscopic treatment. In 1993, Blackwell et al described their success using endoscopic maxillary antrostomy in conjunction with a transconjunctival orbital floor repair in three patients.[22] Endoscopic sinus surgery to create a wide maxillary antrostomy is now considered the surgical gold standard treatment of choice.

However, in doing so, the surgeon must remain mindful of the abnormally low position of the globe, in order to avoid orbital complications. Following surgery, the configuration of the sinus may remain unchanged, improve to some degree or be restored towards normal over time.[11-13] Regardless of the final appearance of the sinus, disease progression is arrested without development of further deformity.[2,11-14]

Reconstruction of the orbital defect has been described using a variety of materials, including alloplastic materials such as nylon sheets, titanium plates, silicon blocks and porous polyethylene implants (Medpore), or alternatively using autologous bone.[3,4] There is however, considerable contention as to whether orbital floor augmentation should be considered concurrently with sinus surgery or as a delayed second stage procedure, or not at all. There is some evidence to suggest that a two stage procedure maybe warranted, in light of some reported cases of spontaneous resolution of the enophthalmos following sinus surgery to restore maxillary sinus aeration.[23]

SUMMARY

In summary, SSS is an interesting clinical entity that has very striking clinical and radiological features. Notable features of idiopathic silent sinus syndrome are the absence of any significant sinus symptoms, a brief duration of symptoms which usually progresses over a period of months, and a high incidence of abnormal sinonasal anatomy. Our own impression is that there maybe infact be some patients who present early, with implosion of the antrum alone, and this may represent a subgroup of patients who may have gone on to develop SSS, with orbital floor wall involvement, if left untreated.

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