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Not so silent sinus syndrome: A case report

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ABSTRACT

INTRODUCTION: Silent sinus syndrome (SSS) is a rare disorder with protean manifestations. An absence of familiarity with ambiguous and atypical presentations may complicate diagnosis and delay management.
CASE PRESENTATION: A 28 year old female patient presented with a chronic history of headache, post-nasal discharge and recurrent facial pain refractory to analgesics. Enophthalmos and hypoglobus progressed over a period of 2 months, and a diagnosis of SSS was confirmed via imaging. Definitive treatment was withheld given the patient's postpartum state and improvement of symptoms.
DISCUSSION: SSS typically manifests with painless and progressive, unilateral, enophthalmos and hypoglobus. Since presentation is dominated by ophthalmologic complaints, the ordinary route by which SSS is diagnosed is through ophthalmology review. The predominant complaint in our patient was chronic headaches with facial pain, and mild enophthalmos and hypoglobus were only noted 2 months later at follow-up. This represents an atypical presentation of SSS, and exemplifies the subtle and often ambiguous presenting features of this disorder.
CONCLUSION: The protean manifestations of SSS mean that patients may initially present to specialities other than ophthalmology. To ensure rapid diagnosis and appropriate management, it is important that clinicians, particularly in ophthalmology, maxillofacial surgery, and ears, nose and throat (ENT), are familiar with this obscure condition.

1. Introduction

The first case of maxillary sinus opacification and atelectasis was reported by Montgomery in 1964 [1]. It was not until 30 years later, however, that the term 'silent sinus syndrome' was attributed to this phenomenon [2]. Patients typically present with unilateral painless enophthalmos and hypoglobus progressing over a period of several months [3]. Until recently, the definition of SSS applied only to cases of idiopathic origin. A greater understanding of the underlying pathogenesis, however, has broadened the spectrum of included aetiologies; idiopathic, post-traumatic and iatrogenic causes are now generally accepted [4].

With exception to enophthalmos, which is identified in almost all cases, the presenting features are diverse and often non-specific. Symptoms include upper-lid retraction, lid lag and lagophthalmos, restriction of gaze, deepening of the superior sulcus and malar depression [4,5]. Visual acuity is usually unaffected, though impaired ocular motility may manifest as diplopia [6]. Silent sinus syndrome has no gender predilection, and the condition presents in the third to fifth decades of life [4]. Obstruction of the maxillary ostium is invariably seen in all cases, and is thought to produce a negative pressure that precipitates retraction of the sinus walls [7].

We present an atypical case of silent sinus syndrome in a 28-year-old female patient. The case highlights the need for a low index of suspicion given the diverse manifestations of this rare condition.

2. Case presentation

A 28-year-old female was referred to the ears, nose and throat (ENT) department with a chronic history of headaches, post-nasal discharge and recurrent facial pain unresponsive to conventional analgesics. No enophthalmos and hypoglobus were noted, and ophthalmology review confirmed no visual impairment. The patient reported no previous history of facial trauma, sinus surgery or malignancy.

Nasoendoscopic examination of the nose revealed deviation of the nasal septum towards the right side with inflammation of the nasal mucosa. Initial computed tomography (CT) revealed an incompletely opacified hypoplastic left maxillary sinus with mucosal thickening at the left ostiomeatal unit (Fig. 1). The frontal, ethmoidal and sphenoidal sinuses were normal. Maxillofacial opinion was requested following suspicion of a left orbital floor fracture, which was excluded upon review of the CT.

Clinical examination two months later identified a very mild degree of enophthalmos, though the face remained symmetrical (Fig. 2). Repeat CT showed pathognomonic signs of silent sinus

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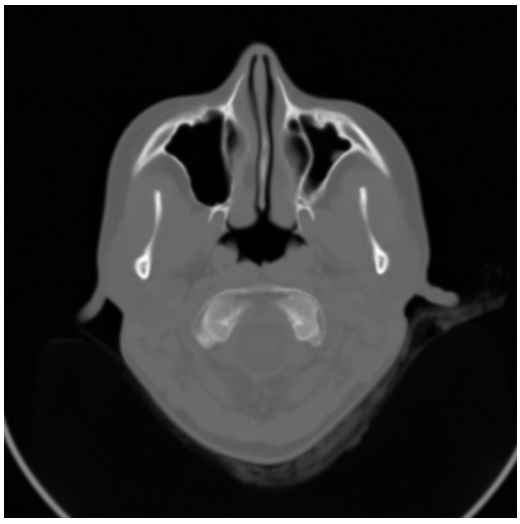

Fig. 1. CT showing incomplete opacification of a hypoplastic left maxillary sinus.

syndrome, with implosion of the left maxillary antrum, downward traction on the orbital floor and inward traction of both the lateral antral and nasal walls (Fig. 3). The patient reported improvement in symptoms by the time of outpatient presentation and, in light of recent pregnancy, it was agreed to review the situation when definitive management was more appropriate.

3. Discussion

Despite the first description of the disease process in 1964, the term 'silent sinus syndrome' was coined much later in 1994 to describe certain cases of enophthalmos and hypoglobus [1,2]. SSS is often used interchangeably with 'chronic maxillary atelectasis' (CMA) to describe the concurrent presentation of enophthalmos and hypoglobus. Nevertheless, it has been argued that SSS should be considered a subtype of CMA, whereby SSS presents in the absence of symptoms of chronic sinusitis [4,5].

Although a diagnosis of SSS may be made clinically, it is best confirmed radiologically. The imaging findings of SSS are pathognomonic. Invariably there is occlusion of the maxillary infundibulum, ordinarily caused by lateral retraction of the uncinate process with apposition against the inferiomedial aspect of the orbital wall [8]. The orbital floor is inferiorly displaced, and there may be increased concavity of the medial and lateral maxillary walls towards the antrum, the ultimate consequence of which is an overall loss in antral volume [2,9]. The nasal septum may deviate towards or away from the affected sinus. Further, changes in bone structure of the maxillary sinus have been extensively reported, though no changes seem to be uniformly consistent [5,9]. Soft tissue changes, including increased opacification of the maxillary sinus and mild-chronic inflammatory changes of the mucosa, are found in almost all patients [8,9].

Two main theories have been proposed for the pathogenesis of SSS. In the 'obstruction of outflow' theory, an acquired obstruction of the maxillary infundibulum causes hypoventilation of the sinus and consequential accumulation of secretions. Reabsorption of secretions leads to formation of a negative pressure in the maxillary antrum, which causes the characteristic changes in the maxillary walls observed in SSS. A number of possible causative factors may lead to infundibular obstruction and the resulting accumulation of secretions, the most important being underlying aberrant nasal anatomy. 62% of patients in a case series of 16 SSS patients exhibited lateral deviation of the middle turbinate [10]. Mucous plugging, mucoceles, and nasal polyps may also contribute to occlusion [11]. Reports of iatrogenic cases of SSS, whereby surgery was thought to have damaged the ostiomeatal complex, supports obstruction of maxillary sinus outflow as a viable common mechanism of pathogenesis [4,12]. The finding of negative

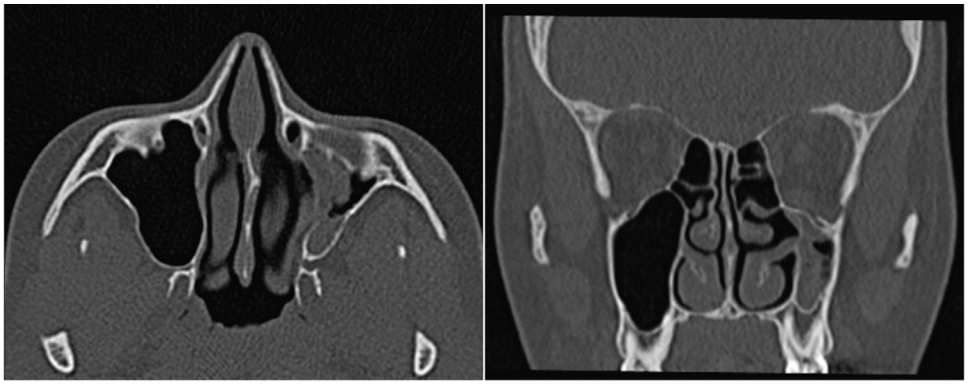

Fig. 2. Sagittal and coronal CT images showing complete opacification and implosion of the left maxillary antrum with downward traction on the orbital floor.

Fig. 3. Portrait and superior perspective of enophthalmos of the left eye. Line added to demonstrate the subtlety of hypoglobus.

pressure has not been substantiated however. Manometric studies of patients with complete infundibular occlusion found an isobaric pressure in the maxillary antrum comparable to controls [13]. The second theory, known as the 'mechanical' theory, posits that contraction and relaxation of masticatory muscles may cause aspiration of a closed maxillary antrum, leading to collapse of the sinus walls [14].

The natural history of SSS is variable. Whilst Rose et al. suggest that SSS is a non-progressive disorder, a retrospective study of 64 patients found that most patients report progressive changes over a period of weeks to months [15,9]. Thus, management is ordinarily commenced soon after presentation. Treatment is usually surgical, and aims to both restore normal ventilation of the maxillary sinus and to reverse the residual skeletal abnormalities. Conventionally, blockage of the ostiomeatal complex is removed through endoscopic uncinectomy and antrostomy, and reversal of enophthalmos and hypoglobus is achieved through orbital floor or medial wall implants. More recently however, a report documented the use of balloon sinuplasty to successfully treat SSS. The Seldinger technique was used to advance a balloon into the infundibulum before inflation, causing sustained restoration of maxillary sinus ventilation and reversal of symptoms [16]. However, displacement of the orbital floor in this patient was minimal, and it is inconceivable that sustained remodelling of the uncinate process is achievable with all cases of SSS, given different aetiologies, severity of disease, and anatomical variation.

Enophthalmos is the most common presenting feature of silent sinus syndrome, reported in 96% of cases [5]. Since presentation is dominated by ophthalmologic complaints, the most common route by which silent sinus syndrome is discovered is through ophthalmology review [11]. Our patient presented with a chronic history of headaches and left facial pain, without associated ophthalmologic complaints. Enophthalmos developed over a 2 month period following ENT review, coinciding with progression of radiological changes in the left maxillary sinus. The degree of maxillary implosion however, as demonstrated by CT, was more marked than suggested through the clinical presentation. These findings demonstrate the ambiguous manifestations of SSS.

4. Conclusion

As this case demonstrates, the ambiguous and often subtle clinical features of silent sinus syndrome means that patients may initially present to specialities other than ophthalmology. To ensure timely management, it is important that clinicians, particularly in ENT and maxillofacial surgery, are familiar with this obscure condition.

Conflicts of interest

The authors declare no conflicts of interest.

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Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

The idea of the project was conceived by Vijay Santhanam and Shadi Basyuni. Ashley Ferro and Shadi Basyuni carried out the literature review. The paper was written and reviewed by Ashley Ferro. All authors contributed to the refinement of the case report and approved the final manuscript. Vijay Santhanam was the senior consultant in charge of this case.

Guarantor

Ashley Ferro, Shadi Basyuni.

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