CASE REPORT

Journal of Research in Dental and Maxillofacial Sciences

DOI: 10.61186/jrdms.9.4.68

Clinical and Radiographic Evaluation of the Silent Sinus Syndrome: The Importance of Simultaneous Assessment

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Article History
Received: 11 April 2024
Accepted: 16 July 2024

Abstract

Background and Aim: The silent sinus syndrome (SSS) is indeed a rare clinical condition. The exact cause of SSS is still unknown, but it is believed to result from chronic negative pressure within the maxillary sinus, leading to progressive inward retraction of the sinus walls and subsequent orbital floor remodeling.

Case Presentation: This case report describes a 61-year-old male with slight facial asymmetry and nasal septum deviation with a normal range of vision. According to cone-beam computed tomography (CBCT) findings, the right maxillary sinus appeared hypoplastic and opaque and the right orbit had 4.58 mm displacement compared to the left orbit.

Conclusion: Accurate diagnosis of the SSS requires assessing clinical and radiographic findings. Rhinologists and, to a lesser extent, ophthalmologists and dentomaxillofacial radiologists are skilled at diagnosing SSS through CBCT imaging of the paranasal sinuses. However, general radiologists are not often well aware of this syndrome. Increasing awareness among general radiologists is vital for prompt identification and appropriate care.

Keywords: Enophthalmos; Maxillary Sinus; Syndrome

Cite this article as: Zare Bidoki F, Heidarkhani Tehrani S, Kiyamehr Z. Clinical and Radiographic Evaluation of the Silent Sinus Syndrome: The Importance of Simultaneous Assessment. **J Res Dent Maxillofac Sci. 2025; 10(1):68-72.**

Introduction

The silent sinus syndrome (SSS) is a rare clinical disorder characterized by the maxillary sinus atelectasis resulting in painless enophthalmos, hypoglobus, and facial asymmetry [1,2]. It usually occurs unilaterally and in the third to fifth decades of life, without any gender predilection. Children are rarely affected by the SSS [3]. Most patients with this syndrome experience progressive enophthalmos without any sinus symptoms, and go through a painless phase of the disease, and since it progresses slowly, it has been dubbed as "silent" [4]. The SSS can be classified as primary or secondary. The primary type is spontaneous and idiopathic, while the secondary type is caused by trauma, facial injuries, rhinoplasty, or chronic sinusitis. The secondary type has also been observed in 1% of patients with the Graves' disease [1].

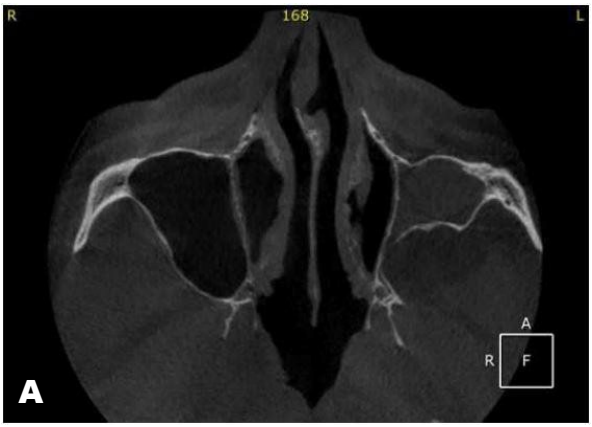
Typically, the SSS presents with facial asymmetry, enophthalmos of 2-5 mm, and long- standing hypoglobus (lasting months to years). The unclear pathophysiology of the SSS has led to several possible theories regarding its development. One of the most plausible theories suggests that chronic obstruction leads to gradual air resorption from the maxillary sinus ostium/ostia resulting in negative sinus pressure. As a consequence, all four walls of the maxillary sinus including the roof (orbital floor), medial, posterolateral, and anterior walls gradually collapse inwards. A progressive increase in orbital volume occurs with enophthalmos and variable malar eminence flattening [5].

However, based on the unclear pathophysiology, slow progression, and limited mention in the radiology literature, many radiologists are unfamiliar with this syndrome and its associated radiographic findings [6]. Delayed diagnosis of SSS can result in various complications, including orbital changes, visual impairment, facial deformities, sinus obliteration, enophthalmos, and hypoglobus. On the other hand, early diagnosis of the SSS can prevent the development of these problems and the need for invasive surgery [7, 8]. Therefore, when the SSS is suspected based on clinical features, radiographic confirmation becomes essential [4]. It is crucial for accurate diagnosis, treatment planning, and prognosis to have sufficient knowledge about the radiographic and clinical features of the SSS. This case report describes a unique case of the SSS.

Case Presentation

A 61-year-old male patient underwent cone-beam computed tomography (CBCT) to assess the suitability of an implant insertion site. Clinical examinations only revealed a slight asymmetry of the face. According to the patient's medical history, he was asymptomatic and had a normal range of vision. Furthermore, no other symptoms such as diplopia, abnormal eye movements, or rhinitis were observed. The patient mentioned a history of recurrent sinusitis and respiratory problems over the past 5 years. There was no evidence of facial trauma, systemic infection, or cranial malformation. According to an ENT specialist, a deviation of the nasal septum and concha bullosa were the main reasons for the patient's respiratory problems. The axial and coronal sections of the CBCT revealed hypoplastic and opaque maxillary sinus ossification on the left side and a slight deviation of the nasal septum to the right, as well as thinned and depressed orbital floors in the posterior sinus wall (Figure 1A).

Moreover, thickening of the sinus floor mucosa was observed in the right maxillary sinus. On the right side, there was a downward displacement of the sinus floor accompanied by an increase in the orbital content. The right orbit showed a displacement of 4.58 mm compared to the left orbit. The lateral wall of the maxillary sinus had also shifted. Another incidental finding in the coronal section was the presence of concha bullosa in the middle turbinate on both sides (Figures 1B and 1C). The deviated shape of the nose and its lateral adhesion to the lamina papyracea had obstructed the drainage pathway of the ostiomeatal complex. Based on the clinical and radiographic examinations mentioned above, a diagnosis of the SSS was made.



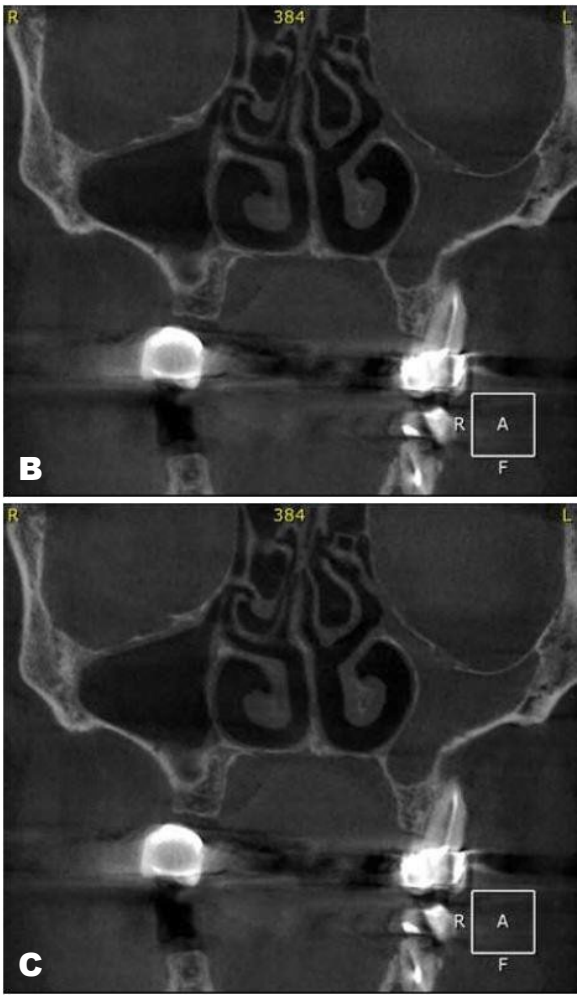


Figure 1. A (upper left): Axial view of the maxillary sinus showing the inward bowing of the posterior wall in the left side maxillary sinus and opacification and hypoplasia of the sinus. B (upper right) and C (down): Coronal view of the maxillary sinus and 4.58 mm inferior displacement of the left orbital floor and obstruction of the complex and inward bowing of the lateral wall.

Discussion

The SSS is characterized by spontaneous unilateral maxillary sinus atelectasis and ossification. As a result of the low prevalence of this disease and lack of sufficient knowledge about it, a precise estimate of its prevalence cannot be made [9]. Nevertheless, based on the available literature, approximately one hundred cases have been reported so far [10]. The SSS typically affects individuals between the ages of 30 and 60 years, who present with symptoms such as progressive enophthalmos and painless, spontaneous hypoglobus [11]. There is no significant evidence pointing to a correlation between the SSS and factors such as gender, occupation, smoking, alcohol consumption, or genetics. The development of the SSS can be explained by two theories: (I) maxillary sinus atelectasis, which may be idiopathic or recurrent after surgery or trauma, and (II) maxillary sinus hypoplasia, which is usually asymptomatic and is often incidentally detected during imaging.

Rose et al. [11] conducted a study involving 14 patients with the SSS, and found that all of them exhibited enophthalmos ranging from 1 to 4 mm, and 4 mm hypoglobus. Perera et al. [12] and Sánchez-Dalmau et al. [13] examined patients with hypoglobus and enophthalmos. All patients were in the age range of 24-74 years, with a mean age of 40.3 years. There was no significant difference between the right and left sides of their sinuses. They did not find any association between gender, occupation, smoking status, alcohol consumption, or genetics with the SSS. Furthermore, there was no evidence of a correlation between the SSS symptoms and chronic sinusitis.

Both the clinical examination and imaging findings play important roles in the diagnosis of the SSS. During physical examination, certain signs can be observed, such as the upper eyelid contraction, upper eyelid depth, malar depression, facial asymmetry, and diplopia. Imaging findings provide additional information and help in the diagnosis. The initial imaging symptom is the loss of the maxillary sinus volume due to retraction of the sinus walls, which is the main reason for the increase in orbital volume and enlargement of the middle meatus. It is common for all four sinus walls to shrink, but one or more of the inner, anterior, or posterior walls may be completely removed. As a result, orbital floor shrinkage always occurs and, in some cases, orbital floor thinning may also be present. The other sinus walls may

appear normal or slightly thin or thick [14]. In a study by Tousidonis et al. [4] five patients with the SSS were examined; it was found that in all cases, the affected side showed increased orbital volume due to the retraction of either the sinus roof or the orbital floor into the maxillary sinus.

The exact pathophysiological cause of the SSS remains unknown, despite several theories being proposed. The most plausible explanation involves complete obstruction of the ostium, leading to hypoventilation and accumulation of secretions in the maxillary sinus.

It is important to accurately diagnose the SSS because different conditions that cause enophthalmos require different treatments and have varying prognoses. The SSS can be misdiagnosed as other conditions, including tumors, trauma, congenital facial asymmetry, diffuse facial lipodystrophy, Parry Romberg syndrome, and linear scleroderma [6].

Following appropriate procedures, both the normal position of the eyeball and proper ventilation of the maxillary sinus can be restored. In the past, a method known as the Caldwelle Luc was commonly used; but in modern times, endoscopic sinus surgery with uncinectomy and antrostomy has become the gold standard for treatment and restoring the sinus function while preserving the structures of the maxillary sinus [15]. The reconstruction of the orbital floor remains a topic of debate. Recent reports suggest that this procedure is commonly performed about 2 months after the sinus surgery, serving as a follow-up to address issues such as enophthalmos and hypoventilation, and to restore normal sinus pressure. In case of severe enophthalmos, orbital floor reconstruction may be necessary. By undergoing this single procedure, patients often experience significant improvement and a reduction in hospital stay. Additionally, since most cases show negative sinus culture after the initial surgery, the risk of infection following this procedure is minimal [16].

Conclusion

The SSS is indeed a rare condition, and its exact cause remains unknown. The diagnosis of SSS is typically made when ocular complaints, such as enophthalmos, or hypoglobus are observed without any prior signs of sinus disease. Since the etiology of the SSS is not well understood and there is limited knowledge about this syndrome, it is important to increase awareness and knowledge about the SSS among healthcare professionals, particularly radiologists and dentists. This is because the SSS can be an incidental radiographic finding during imaging studies. By enhancing the knowledge of these professionals, they can recognize the characteristic imaging findings associated with the SSS and consider it as a potential differential diagnosis when evaluating a patient. This can lead to earlier identification and appropriate management of the SSS cases.

Conflict of Interest

The authors declare no conflict of interests.

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