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Case Report

Clinical and morphological features of the orbital floor in a patient with silent sinus syndrome: A clinical case report

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Abstract: Silent sinus syndrome (SSS) is a rare condition characterized by spontaneous unilateral displacement of the inferior and posterior globe due to maxillary sinus (MS) atelectasis. The mechanism of MS development is not fully understood, but orbital floor changes due to changes in MS pressure or subclinical inflammation are considered part of the potential pathogenesis. In addition, treatment strategy and optimal timing are not defined. We present a case report of a patient with MS who underwent successful uncomplicated two-stage treatment with good functional and aesthetic results. Orbital floor morphology data obtained for the first time in a patient with SSS may provide additional information about the pathogenesis of this rare disease.

Keywords: silent sinus syndrome, enophthalmos, hypoglobus, orbital floor, bone morphology.

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Introduction

Silent sinus syndrome (SSS) is a rare disorder characterized by spontaneous painless downward displacement of the eye associated with collapse of the ipsilateral maxillary sinus (MS) [1]. Its typical clinical features include enophthalmos, hypoglobus, pseudoptosis or retraction of the upper eyelid, and deepening of the superior sulcus. The disorder occurs equally in men and women. The onset of the disorder is usually between 30 and 40 years of age, and both sides are affected equally. Diagnosis is based on specific clinical features and pathognomonic features detected by multislice computed tomography (CT). Currently, the pathogenesis of SSS remains uncertain, and idiopathic, posttraumatic, and iatrogenic factors are considered as possible etiologies. Moreover, there is no consensus regarding the risk factors, indications for surgical treatment, preferred surgical technique and timing of orbital floor reconstruction, as well as regarding the orbital floor changes characteristic of this disease [2]. The main treatment goals of SSS include restoration of MS aeration and correction of the position of the eyeball [3].

To our knowledge, this is the first case describing morphological changes in the orbital floor. The latter was sampled during a two-stage treatment of a patient with SSS.

Case report

A 42-year-old female patient presented to our Department of Oculoplasty with complaints of left orbital discomfort with eye movements, binocular diplopia, cosmetic defect due to sunken left eye, and palpebral fissure asymmetry. These symptoms had developed three months prior to the visit against a background of complete well-being. Examination of the left eye revealed uncorrected visual acuity of 20/20, enophthalmos of 4 mm, hypoglobus of 0.5 mm, binocular diplopia with upward gaze, marked limitation of ocular motility with upward gaze, and decreased ipsilateral upper eyelid excursion (*Figure* 1A). The patient had no history of recent sinusitis or trauma. On CT, the left MS was completely opacified and reduced in volume with a downward displacement of the orbital floor. The left orbital floor was also thinner than that on the affected side (*Figure* 2A). The first stage of surgical treatment involved functional endoscopic sinus surgery (FESS) performed by an ENT surgeon, which included resection of the lower part of the uncinate process and expansion of the natural ostiomeatal complex. After aspiration of thick mucin from the sinus, significant dislocation of the orbital floor became visible. The postoperative period was uneventful. After FESS, pain and discomfort with eye movement disappeared. CT scan performed six months after FESS showed pneumatization of all paranasal sinuses, an increase in the volume of the right MS by 20%, and an orbital floor elevation of 1 mm (*Figure* 2B). However, since the patient still complained of asymmetry in eye position (3-mm enophthalmos) and residual binocular diplopia in upward gaze, transconjunctival reconstruction of the orbital floor was performed using a molded allogeneic implant. During the orbital surgery, a sample of the orbital floor was taken for subsequent morphological analysis. The orbital stage surgery resulted in normalization of the right eye position, upper eyelid excursion, and binocular status (*Figures* 1B and 2C).

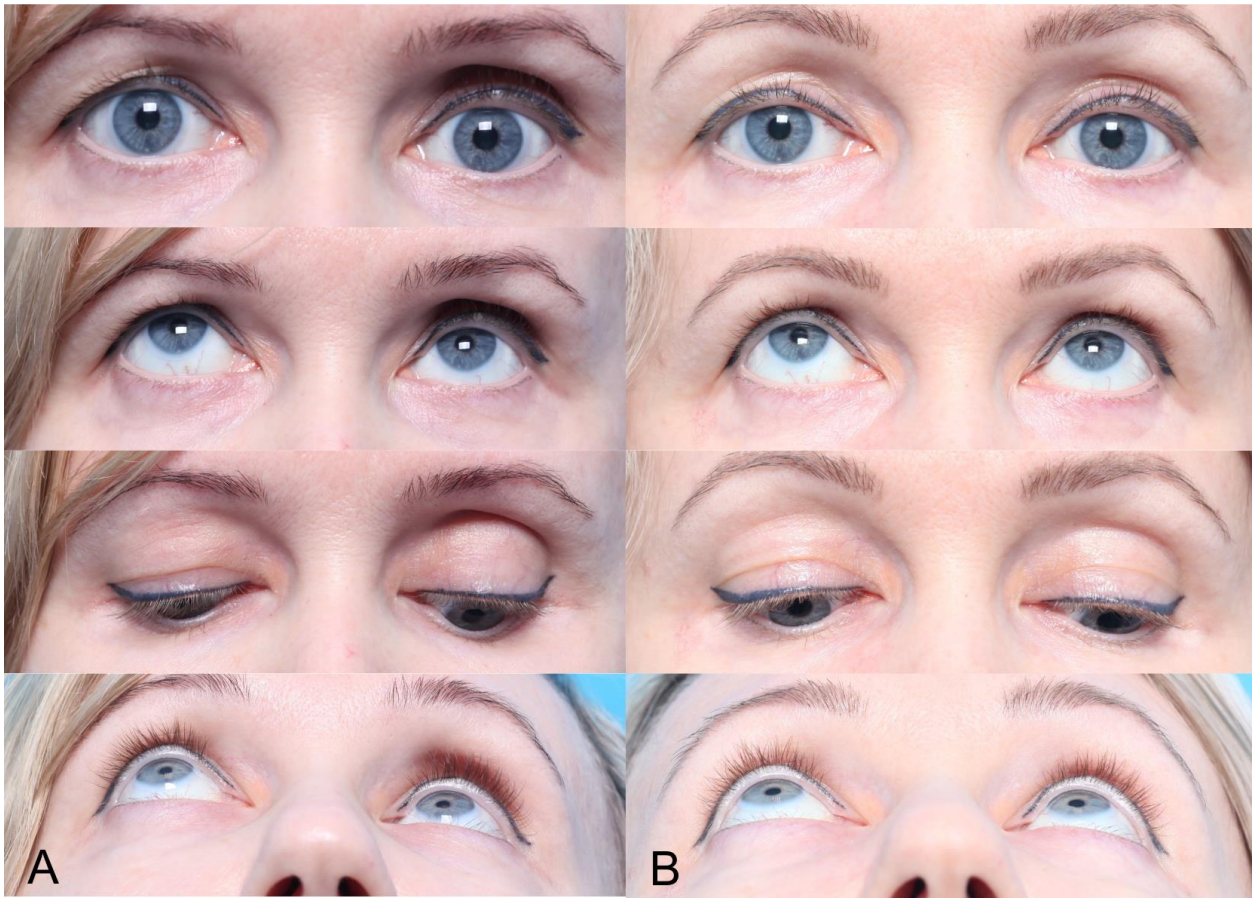

Figure 1. Clinical photographs of a patient with silent sinus syndrome in the primary position (frontal view), upward gaze, downward gaze, and vermis view (A); one month after orbital floor surgery (B).

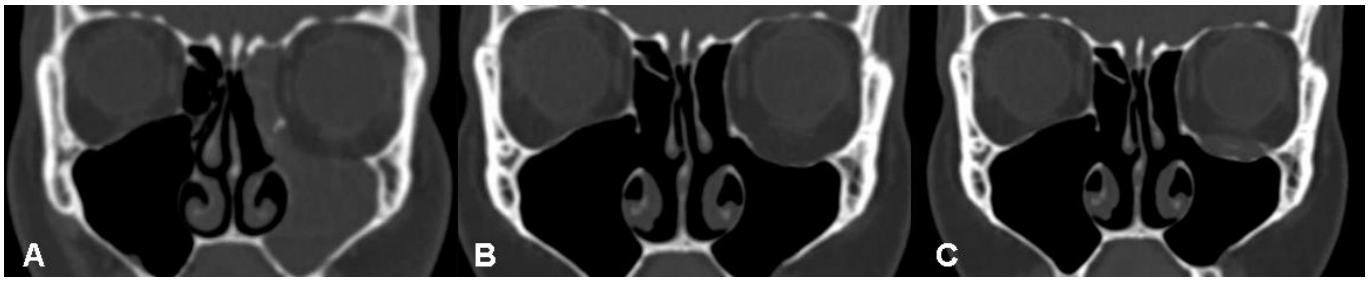

Figure 2. Multislice computed tomography scan of the paranasal sinuses of a patient with silent sinus syndrome at admission (A), 6 months after functional endoscopic sinus surgery (B), and one month after orbital floor surgery (C).

Morphological analysis of the orbital floor (*Figure 3*) showed that the correct orientation of the bone trabeculae of the lamellar structure was preserved; however, the thickness of the bone trabeculae was slightly reduced. The Haversian canals contained osteocyte nuclei. The intertrabecular spaces were filled with mature adipose tissue without atypia. There were no signs of inflammation or osteoclastic activity.

Discussion

The pathogenesis of SSS is still poorly understood. Characteristic CT features include ipsilateral collapse and opacification of the MS with downward displacement of the orbital floor. However, radiographic bone findings remain controversial. While most authors describe focal demineralization and reduced density, others report bone thickening [4, 5].

Animal models suggest that complete closure of the native ostiomeatal complex causes a significant decrease in air pressure. This leads to collapse of the MS, and the remaining space is filled with acellular transudate. The accumulation of mucous secretions may cause inflammation and subsequent erosion of the sinus walls in the nonventilated sinus [6]. Rose et al. noted that this mechanism may be similar to that occurring in the middle ear with Eustachian tube obstruction. The authors found that a pressure decrease of only 2 mmHg significantly increased osteoclast activity. As a result, resorption of the middle ear walls occurred two weeks after tube obstruction [4].

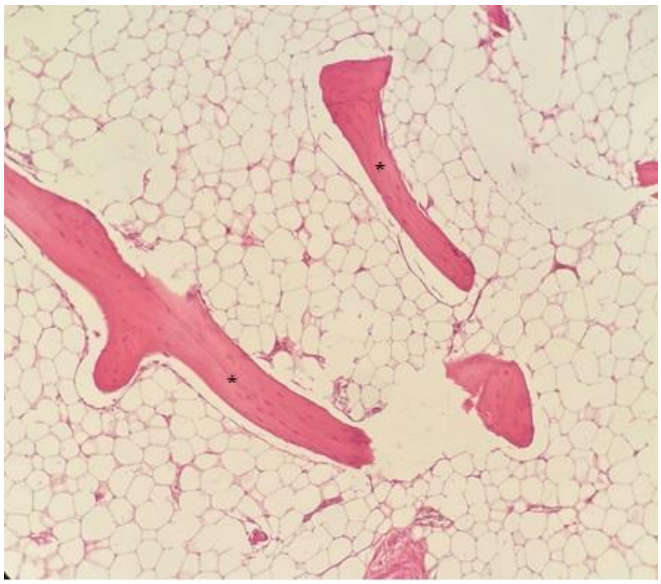

Figure 3. Orbital floor bone specimen of a patient with silent sinus syndrome (*bone trabeculae, description in text). Hematoxylin and eosin. Magnification ×100.

Previously described specific morphological changes in the mucosa of the posterior wall of the MS include compression atrophy and lymphoplasmacytic infiltration of the sinus mucosa, as well as thickening of the basement membrane [7].

The gold standard of surgical treatment is a two-stage approach, but some authors give priority to simultaneous MS surgery and orbital surgery. In the first stage, FESS is performed, which helps restore MS aeration. The second stage is aimed at eliminating the orbital volume deficit. Indications for orbital surgery and its timing remain controversial, mainly due to the assertion that restoration of MS aeration leads to normalization of the orbital floor position and regression of enophthalmos. Recently, Amin et al. followed the change in MS volume and orbital volume after FESS using CT, confirming a significant change in the eye position after FESS [8]. Thus, the need for orbital floor reconstruction depends on the severity of diplopia and cosmetic defect due to enophthalmos, as well as on the postoperative changes in the sinus. Arguments in favor of delaying the intervention for 3-6 months after FESS include the benefits of delaying implant placement in preventing hypoglobus overcorrection or inflammatory complications [9]. The use of nonsurgical injection techniques for orbital volume correction in patients with SSS has also been described [10].

Conclusion

We describe the case of a patient with SSS who underwent successful two-stage treatment (FESS with delayed orbital implant placement) with good functional and cosmetic results. For the first time ever, an orbital floor was sampled for morphological examination, which showed no signs of osteolysis in this patient. It is worth noting that although osteoclastic activity was absent in the tissue sample, osteoclasts may participate in bone remodeling in the early stages of the disease.

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