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Silent Sinus Syndrome in Children - A Systematic Review

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ABSTRACT

Introduction: Silent sinus syndrome (SSS) is an acquired condition characterized by a decreased maxillary sinus volume along with an inward bowing of one or more antral walls. This review aims to identify and evaluate the management options for SSS in the pediatric population.

Data Sources: The PubMed and Science Direct databases were used to search and review. The studies were screened for inclusion using the following criteria:

1) Cases diagnosed with silent sinus syndrome.
2) Studies based on pediatric patients.
3) Written in English.
4) SSS not caused by head trauma.

Results: Thirteen patients were included in the study. An aesthetic defect in the form of facial asymmetry was the only complaint in 2 patients. Both enophthalmos and hypoglobus were present in 69% of patients. Diplopia was reported by 23% patients. The opacification of the maxillary sinus was present in all the patients.

Conclusion: Restoring sinus aeration allows for resolution of symptoms after sinus surgery. For this reason, simultaneous orbital floor reconstruction is not recommended in children. The management of SSS should be established by a multidisciplinary team of ENT surgeons, ophthalmologists and maxillofacial surgeons. In addition to providing the best possible care, a consistent evidence-based management algorithm for pediatric SSS patients will also help identify the etiology for this rare, and potentially serious, condition.

Keywords: Silent Sinus Syndrome; Maxillary Sinus Atelectasis; Children; Enophthalmos; Hypoglobus; Functional Endoscopic Sinus Surgery; Chronic Sinusitis

Abbreviations: SSS: Silent Sinus Syndrome; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; CRS: Chronic Rhinosinusitis; FESS: Functional Endoscopic Sinus Surgery

Introduction

Silent sinus syndrome (SSS) is an acquired condition characterized by a decreased maxillary sinus volume along with an inward bowing of one or more antral walls [1]. SSS is typically unilateral and occurs without prior history of sinus symptoms, hence the term „silent". The pathophysiology of SSS remains unclear. According to the current hypotheses it is caused by idiopathic occlusion of the osteomeatal complex resulting in long-term hypoventilation and discharge of the sinus. The resorption of secretion creates a negative pressure gradient within the maxillary sinus that leads to demineralization and bony resorption of sinus walls. The process of diminishing the volume of the sinus leads to a retraction of all or most of its walls. Also compensatory augmentation in ipsilateralorbital volume progresses into the lowering of the orbital floor with sinus atelectasis [1,2]. The condition leads to a sinus-related orbitopathy involving progressive enophthalmos, hypoglobus and facial asymmetry. Enophthalmos is a posterior displacement of the globe within the orbit. Clinically, the

eye may appear sunken and have a deeper superior sulcus with either an upper eyelid retraction or upper eyelid ptosis [3]. Hypoglobus refers to the inferior displacement of the globe in orbit. Such anatomic modifications progress over time [1]. The main complaint always regards some aesthetic and uncomfortable changes in the appearance like eyelid retraction, deepening of the superior orbital sulcus, and flattening of the malar region. The condition is painless [4,5].

Additionally, diplopia can occur rare [5-7]. The diagnosis is made on the basis of clinical and radiological findings. Endoscopic examination of a nasal cavity presents: normal or mildly inflamed nasal mucosal lining, enlarged middle meatus on the affected side, and the uncinate process completely adherent to the lateral wall obstructing maxillary natural ostium [5,8]. When suspecting such a diagnosis a CT scan is recommended. Radiographic findings include: unilateral maxillary loss of sinus volume with its opacification, depression of the ipsilateral orbital floor, obstruction of the osteomeatal complex, lateralization of the ipsilateral uncinate process and pterygopalatine fossa enlargement [1,2,8]. Kass et al. reported three stages of chronic maxillary atelectasis varying from the lateralized maxillary fontanel (stage I), inward bowing of one or more osseous walls of the maxillary antrum (stage II) and clinical deformity with enophthalmos, hypoglobus and/or midfacial deformity (stage III) [9]. The difference between the definition of grade III chronic maxillary atelectasis and SSS is the presence or absence of sinus-related symptoms. The treatment is surgical, with functional endoscopic sinus surgery to restore proper ventilation [5,10,11]. If significant facial asymmetry is present a orbital reconstruction is also recommended. Restitution treatment of exophthalmos due to orbital floor displacement involves plastic reconstruction of the floor of the orbit (with implants) via a transconjunctival approach [5,6,12-14]. This condition is more commonly seen in adults but can also occur in children. This review aims to identify and evaluate the management options for SSS in the pediatric population.

Data Sources

The systematic review was performed according to the PRISMA 2020 checklist [15]. The PubMed and Science Direct databases were used to search and review the most significant papers on the silent sinus syndrome. The following terms were used for the database search: silent sinus syndrome and pediatric and children. Only studies published in English were considered for inclusion. The studies were screened for inclusion using the following criteria:

1) Cases diagnosed with silent sinus syndrome.
2) Studies based on pediatric patients.
3) Written in English.
4) SSS not caused by head trauma.

The criteria for exclusion included:

1) No access to the full-text paper.
2) Reviews based on the adult population.
3) Not written in English.
4) SSS caused by head trauma or congenital defects.

Results

A total of 289 studies were retrieved through the systematic search of bibliographic databases. After removing 3 duplicates and 1 paper with no open access, 285 papers were eligible for the title and abstract screening. After the title and abstract screening, the authors excluded 253 articles and read the full text of 32 papers. 12 papers were rejected due to the wrong patient population, 8 studies were based only on radiological data and 2 papers were not available in English. In 2 studies, the described cases were proven to be related to a head injury. One patient was diagnosed with Williams syndrome. This patient was included in this study based on absent abnormalities in sinus imaging up to 5 years of age. The patient had neither a history of sinonasal complaints nor recurrence of upper respiratory tract infections. At 7 years of age the examination showed facial asymmetry with right hypoglobus and enophthalmos that had never been reported during the first years of life. This ultimately led to the inclusion of 8 publications [16-23] containing case reports of 13 children with silent sinus syndrome (Figure 1). All 8 studies were written in English and published from 2014 to 2025. The results from this retrospective review are shown in Table 1. The clinical data, including sex, age at diagnosis, symptoms, computer tomography findings and treatment were analyzed. Out of the 13 patients, 6 were male and 7 were female. The mean age was 8,9 years (with the range between 3-14 years). We have also described two cases diagnosed and treated in our department.

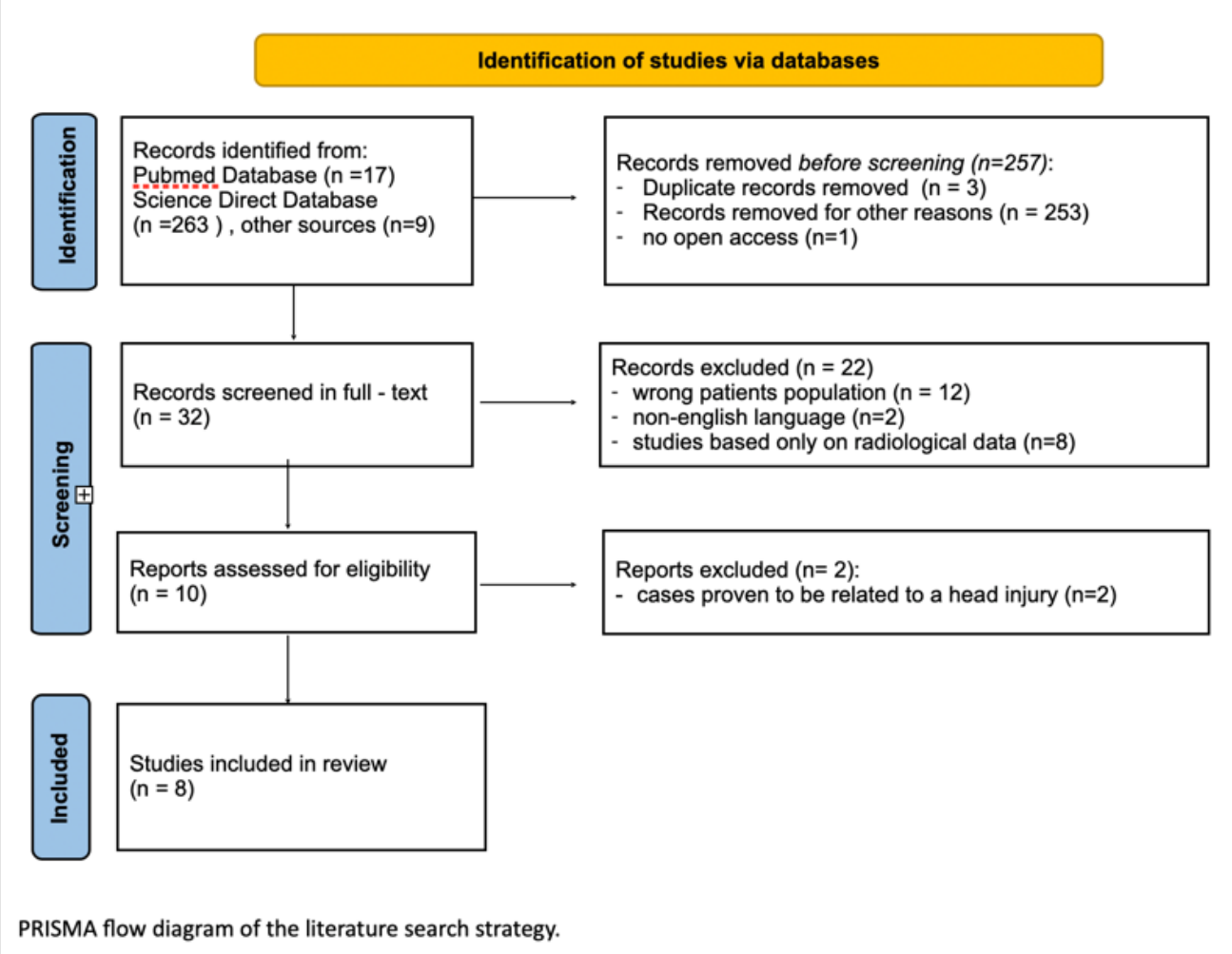


Figure 1: Literature search strategy.

Table 1: Clinical features of patients with SSS.

Papers	Sex	Age	Symptoms	Radiographic Findings in CT scan	Affected Side	Treatment
Farneti, et al. [16]	M	7	Enophthalmos, hypoglobus, headache	lateralization of the uncinate process, opacification of the maxillary sinus, depression of the orbital floor	L	FESS + A
	M	8	Headache, hypoglobus, nasal congestion	lateralization of the uncinate process, opacification of the maxillary sinus, depression of the orbital floor	R	FESS + AT
	M	9	enophthalmos hypoglobus, nasal congestion	lateralization of the uncinate process, opacification of the maxillary sinus, depression of the orbital floor	L	FESS + A

Farneti, et al. [16]	M	10	enophthalmos, hypoglobus, headache, diplopia	lateralization of the uncinate process opacification of the maxillary sinus, depression of the orbital floor	L	FESS + A
	M	11	enophthalmos, hypoglobus, headache, diplopia	lateralization of the uncinate process opacification of the maxillary sinus, depression of the orbital floor	R	FESS
	F	14	Enophthalmos, sinusitis	lateralization of the uncinate process, opacification of the maxillary sinus,	R	FESS
Ihnat, et al. [17]	F	4	Symptoms of astma and allergy rhinitis at the age of 13 for repeat evaluation. Her mother reported that she had not had any sinonasal intervention, however she did start omalizumab after which there was marked improvement in both her asthma and allergic rhinitis.	CT at 4 year-old lateralization of the uncinate process opacification of the maxillary sinus, depression of the orbital floor CT at 13 year-old a right maxillary sinus smaller than the left, but well-aerated, without opacification. slight depression of the orbital floor	R	omalizumab
Chang, et al. [18]	F	7	headache, facial asymmetry, hypoglobus, enophthalmos	diminished sinus volume opacification of the maxillary sinus,	R	FESS + AT
González, et al. [19]	M	3	No symptoms SSS-related	At age 3 bilateral maxillary atelectasis At age 6 spontaneous resolution of the right maxillary atelectasis and persistence of the left maxillary atelectasis	R L	No treatment
Leidens, et al. [20]	F	12	facial asymmetry, hypoglobus, enophthalmos	bowing of the medial wall of the maxillary sinus. opacification of the maxillary sinus,	L	FESS
Petraroli, et al. [21]	F	7	facial asymmetry, hypoglobus, enophthalmos	retraction of the maxillary sinus walls diminished sinus volume lateralization of the uncinate process opacification of the maxillary sinus, retraction of the middle turbinate demineralization of the sinus walls	R	No treatment No progression of disease
Dorlodot, et al. [22]	F	12	Nasal discharge Postnasal drip Facial pressure/pain	Inward bowing of the maxillary walls, lateralization of the uncinate process opacification of the maxillary sinus, depression of the orbital floor	R	FESS
Mohindra, et al. [23]	F	12	diplopia, eyes asymmetry, enophthalmos, hypoglobus, restriction in elevation of the eyelid	retraction of all the walls of maxillary sinus, diminished sinus volume, lateralization of the uncinate process, opacification of the maxillary sinus, depression of the orbital floor,	R	FESS Orbital floor reconstruction with nasal septal cartilage

Note: A- adenoidectomy; AT - adenotonsillectomy; L - left; R- right.

Symptoms

An aesthetic defect in the form of facial asymmetry (with enophthalmos and hypoglobus) was the only complaint in 2 out of 13 patients. Both enophthalmos and hypoglobus were present in 9 out of 13 patients (69%). Diplopia was reported by 3 patients (23%.). Complaints of headache were present in 4 children (30%). Three other children had additional symptoms of chronic sinusitis and another child had chronic sinusitis with asthma and allergic rhinitis.

CT Scan for Abnormalities

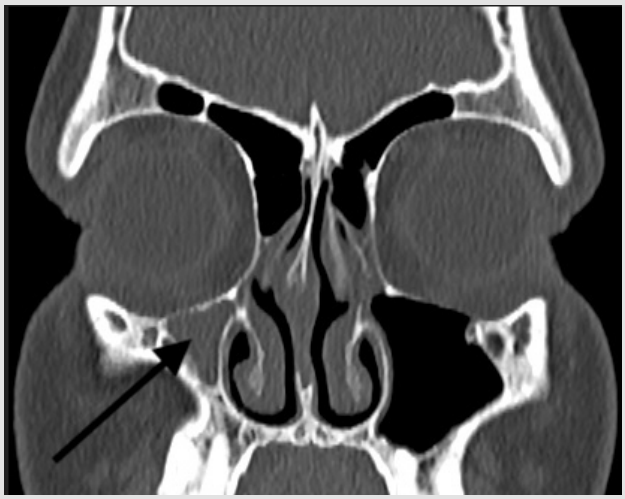
The opacification of the maxillary sinus was present in all the patients (100%), lateralization of the uncinate process in 10 out of 13 patients (77%) and depression of the orbital floor in 9 children (69%).

Treatment

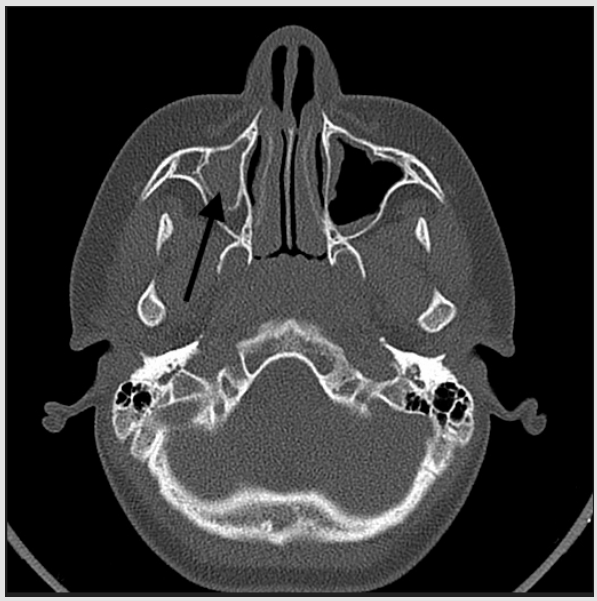
10 patients (77%) underwent surgical treatment: 4 out of 13 had FESS alone, 3 children underwent FESS with adenoidectomy and 2 children had FESS with adenotonsillectomy. The remaining one patient underwent FESS and orbital floor reconstruction with nasal septal cartilage via a subciliary approach. The child with asthma and allergy rhinitis was treated with omalizumab. 2 patients chose not to undergo treatment due to no progression of disease.

Case 1

A 17- year-old boy was admitted to our department for sinus surgery. He reports a history of recurrent nasal congestion on the right side, postnasal drip and weakened sense of smell for 3 years. There were no complaints of pain. CT imaging showed a diminished right maxillary sinus, with loss of continuity of its medial wall and a thickening of its mucosa (Figures 2 & 3). The osteomeatal complex on the right side was narrowed, on the left side partially blocked. There was a slightly thickened mucosa of the right ethmoidal cells. A right-sided antrotomy was performed under general anesthesia. There was also the lateralization of the uncinate process. After uncinectomy the ostium of the right maxillary sinus was opened and widened. A thick, mucous discharge was removed from the interior of the sinus. The sinus was rinsed. No complications were observed in the perioperative period. No facial deformities were observed despite typical abnormalities for SSS in CT images. The child's follow-up shows no progression of changes in the craniofacial bones.



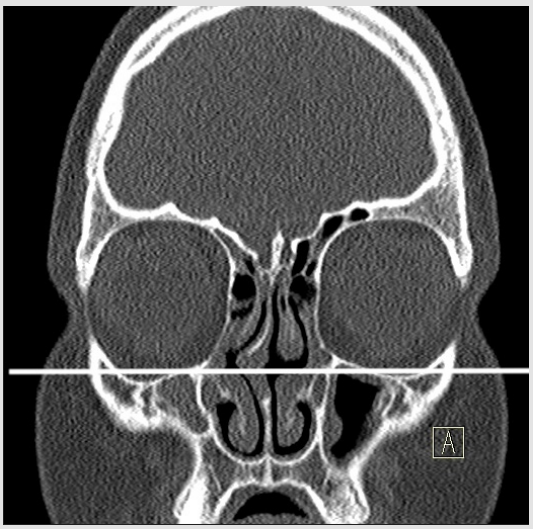
Note: Black arrow indicates opacification of diminished right maxillary sinus and downward bowing of the orbital floor.
Figure 2: CT (coronal view).



Note: Black arrow indicates thinning of sinus walls with opacification of diminished right maxillary sinus.
Figure 3: CT (axial view).

Case 2

A 14 - year-old boy with chronic nasal congestion presented at the hospital because of diplopia and impaired visual acuity in the right eye. A CT scan of the sinuses showed asymmetry of the maxillary sinuses with a significant reduction in the volume of the right maxillary sinus. A total opacification of the right maxillary sinus with inward bowing of its medial wall was present (Figures 4 & 5). There was also lowering of the right orbital floor shown (Figure 4). The child was qualified for an endoscopic sinus surgery. The endoscopy showed a displaced lateral wall of the nose to the right. The lateralization of the uncinate process with its adhesion to the lateral wall was observed (Figure 5). The gentle luxation of the uncinate with a ball probe and its removal was performed. The sinus opening was located and widened. Thick discharge was found inside the maxillary sinus. Multiple rinsings were performed. The mucosa of the right maxillary sinus was slightly swollen. No complications were observed in the perioperative period.



Note: White line indicates horizontal plane.
Figure 4: CT scan. Lowering of the right orbital floor.

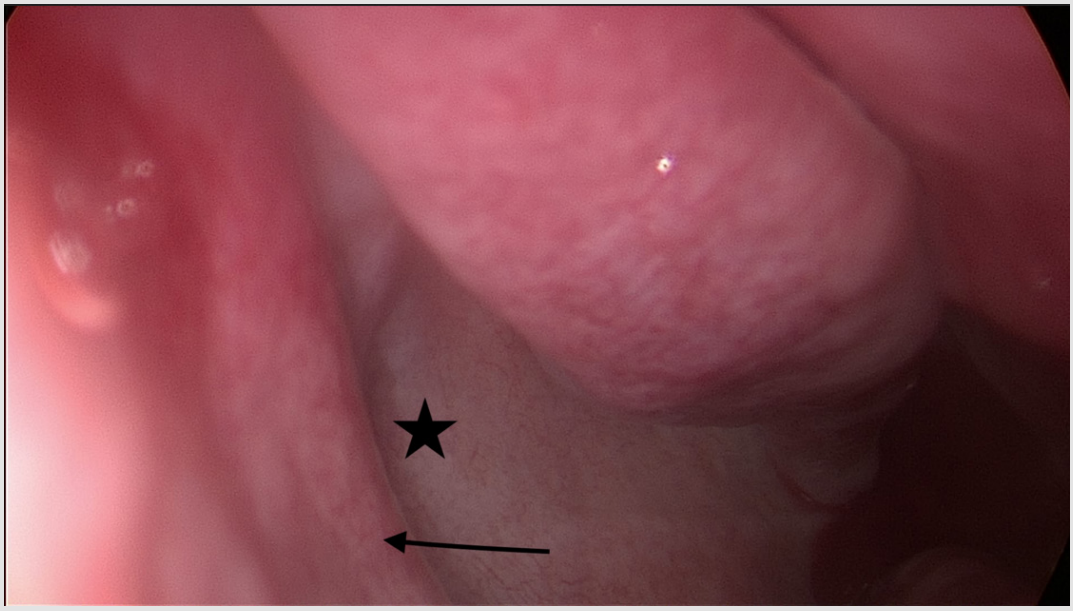


Figure 5: The lateralization of the right uncinate process (black arrow) and inward bowing of medial maxillary sinus wall (black star) in endoscopic view.

Discussion

The term "hypoplastic sinus" should not be used as a substitute for silent sinus. A hypoplastic sinus is referred to a congenital unilateral or bilateral underdevelopment. A CT image in sinus hypoplasia is stable, whereas in SSS, it changes. The term ''silent sinus syndrome'' is defined as a unilateral, spontaneous, progressive and painless enophthalmos associated with atelectasis of a fully developed maxillary sinus [24,25]. The key factor in distinguishing between SSS and chronic maxillary atelectasis with enophthalmos is the presence or absence of sinusitis symptoms. The absence of sinusitis symptoms should be one of the diagnostic criteria for SSS [9,22]. In this study 4 out of 13 children had sinusitis symptoms. The recurrent upper respiratory tract infection linked to adenoid hypertrophy makes this definition much more challenging in the pediatric population [16]. Despite the fact that SSS mainly affects the maxillary sinus, other sinuses can also be affected. The frontal SSS leads to ipsilateral hyperglobus and enophthalmos. Developing this pathology in ethmoid sinuses causes prominent left superior sulcus of the eye and a medially and inferiorly deviated left orbit [26,27]. The opposite condition to silent sinus syndrome is pneumosinus dilatans, in which overpneumatisation of sinus is leading to exploding of the sinus. This pathology can affect any paranasal sinus [24,28]. The gas-distended frontal sinus is pushing down the eyeball [24].

Despite being idiopathic, SSS abnormalities on CT scans can be also caused by mid-face trauma, surgery on the nose and sinuses, chronic rhinosinusitis and Graves ophthalmopathy following orbital decompression [24,27,29-32]. There is a description of a case of ethmoidal SSS development which happened after an injury to the nasal cavity during swab collection [31]. SSS should be differentiated from other causes of spontaneous enophthalmos, such as Parry-Romberg syndrome, IgG4 disease, post radiotherapy atrophy and linear scleroderma [1,33,34]. Nasal endoscopy, CT scan and clinical evaluation are essential for the diagnosis. An endoscopic uncinectomy and maxillary antrostomy is the treatment of choice to restore sinus aeration and pressure. This procedure provides to stop the natural evolution of the disease. In the majority of cases, it leads to resolution of symptoms [22,35]. In a minority of patients, orbital floor reconstruction is necessary to correct persistent enophthalmos [6,13,36]. Some authors prefer to perform sinus surgery concurrently with orbital floor reconstruction. Others recommend a wait-and-see strategy-performing sinus surgery with postponement of orbital floor reconstruction. The spontaneous re-expansion of sinus and resolution of orbital symptoms was reported after FESS [36]. Although the restoring aeration and drainage of the silent sinus is recommended, there are reported cases of spontaneous recovery without treatment [19]. In our patients endoscopic sinus surgery was sufficient treatment and did not require orbital wall reconstruction.

In addition, the bacterial biofilm covering the adenoid contributes to recurrent or chronic sinusitis. In the pediatric population up to 12 years of age, adenoidectomy is recommended concurrently with sinus surgery [16,18]. Our patients were at 14 and 17 years of age and with no adenoid required removal. Most of the studies of this disease and therapeutic recommendations available in the literature are based on the adult population. It should be remembered that in the pediatric

patient group, the sinuses are in the developmental stage and the course of the disease and also the results of surgical treatment may differ from adults. Early recognition of SSS and early intervention may be favorable to prevent long-term orbital changes and complications. If sinus-related orbitopathy develops, the endoscopic sinus surgery is indicated to restore proper sinus ventilation. In pediatric patients the orbital reconstruction should not be perform concomitantly with FESS. There is a greater probability that the symptoms associated with the lowering of orbital floor will resolve and that craniofacial development will return to normal after sinus surgery. Orbital reconstruction should be postponed and considered only if facial asymmetry and diplopia persist [10,16-18,36]. Silent sinus syndrome is relatively uncommon. Although the syndrome is described in literature since 1964, the pathophysiology is still not completely understood. The disease is likely to progress without surgery and using a multidisciplinary approach should deliver the highest quality healthcare [37].

Conclusion

Restoring sinus aeration allows for resolution of symptoms after sinus surgery. For this reason, simultaneous orbital floor reconstruction is not recommended in children. The management of SSS should be established by a multidisciplinary team of ENT surgeons, ophthalmologists and maxillofacial surgeons. In addition to providing the best possible care, a consistent evidence-based management algorithm for pediatric SSS patients will also help identify the etiology for this rare, and potentially serious, condition.

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