Etiology, Early Diagnosis and Proper Treatment of Silent Sinus Syndrome Based on Review of the Literature and Own Experience

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Purpose: The authors' aim was to review the literature in terms of the etiology of the syndrome, the frequency of Silent Sinus Syndrome (SSS) and surgical procedure, as well as to present their own experience.

Methods: The authors used PubMed, Medline, and Science Direct websites to find and review the most significant papers related to SSS. The case reports of SSS published between 2010 and 2020 were reviewed. A retrospective case review of 8 patients with SSS treated at the authors' departments was done.

Results: The silent sinus syndrome has been reported in both children and adults. It is relatively rare and should be differentiated from congenital sinus hypoplasia or atelectasis. It most often affects the maxillary sinus. SSS is usually diagnosed when facial asymmetry or vision problems occur. Late diagnosis requires endoscopic sinus surgery, involving orbital wall reconstruction. The etiology of the syndrome, including the role of bacterial flora found in the sinuses, is unclear.

Conclusion: Early diagnosis of SSS enables avoiding orbital complications and limits surgical intervention to endoscopic surgery. Further research into bacteriology may help to understand the pathophysiology of the silent sinus syndrome.

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The silent sinus syndrome was first described by Montgomery[1] in 1964. Thirty years later, Soparkar[2] proposed the term „Silent Sinus Syndrome" (SSS) and systematized the diagnosis as spontaneous enophthalmos and hypoglobus on the ipsilateral hypoplastic maxilla, secondarily causing orbital floor resorption. The silent sinus is sometimes described as hypoplastic. However, this term should be reserved for congenital unilateral or bilateral underdevelopment. Hypoplasia has 3 stages depending on the sinus size and uncinate process hypoplasia. It is possible to differentiate between SSS and hypoplasia on the basis of computed tomography before any symptoms occur. A computed tomography (CT) image in sinus hypoplasia is stable, whereas in SSS, changes in CT scans are progressing.[3]

Another disease entity often compared to SSS is chronic maxillary atelectasis (CMA). In contrast to SSS, CMA is defined as a persistent and progressive decrease in the sinus volume of the maxilla due to inward bowing of 1 or more antral walls. CMA is classified into 3 stages according to the degree of wall collapse.[4] The fundamental difference between CMA and SSS is the absence of sinusitis symptoms and facial pain (Fig. 1,2). Based on a literature review, Brandt[5] suggests that stage III of CMA and SSS is a spectrum of the same condition.

The silent sinus syndrome is a rare condition and is usually diagnosed when the orbital floor is lowered and the patient complains about vision disturbances. Cases without enophthalmos, but with negative pressure in the maxillary sinus, causing lateralization of the uncinate process and collapse of the maxillary sinus walls visible in CT scans, are assumed to be an early stage of SSS.[6] SSS can be diagnosed among both children[7,8] and adults, with a slight predominance between the 3rd and 5th decade.[9] Some authors suggest that early diagnosis of SSS could possibly diminish the number of complications and deescalate sinus surgery traumaticity. The authors did a literature review in terms of clinical and radiological manifestations, possible pathology and treatment methods of SSS. Additionally, they present their own experience.

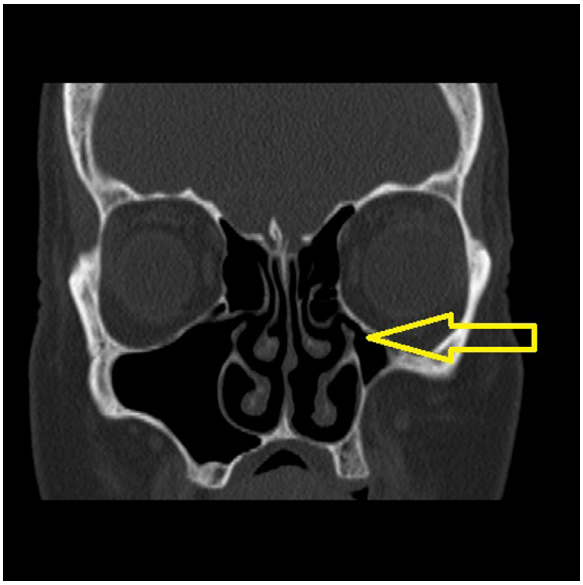

FIGURE 1. CT scan – silent sinus syndrome. Lateralization of uncinate process, maxillary sinus volume loss.

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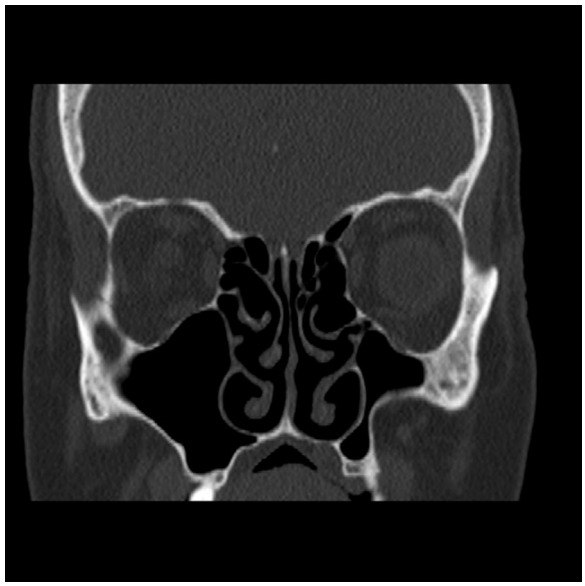

FIGURE 2. CT scan – silent sinus syndrome. Lateralization of uncinate process, maxillary sinus volume loss.

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Materials and Methods

The PubMed, Medline, and Science Direct data bases were used to search and review the most significant papers on the silent sinus syndrome. Subsequently, the same websites were used to search the case reports of the silent sinus syndrome. The inclusion criterion was the publication date, from January 2010 to October 2020. The exclusion criterion was the age, cases of patients below 18 years were not analysed. The key term used during the literature review was the "silent sinus syndrome."

Case reports found in the literature were analysed for sex, age of diagnosis, etiology, symptoms, and treatment.

Moreover, documentation of all patients undergoing endoscopic sinus surgery (ESS) at the Department of Laryngology and Laryngological Oncology in

Katowice-Ochojec due to sinuses diseases was reviewed in the period from September 2017 until December 2020. In this paper, the authors present cases of 7 patients treated with ESS due to SSS. The diagnosis of SSS and appropriate treatment were based on physical examination and CT scans.

In this paper, the authors also included 1 case of SSS reported in 2020 at the Department of Otorhinolaryngology and Oncological Laryngology in Zabrze.

Particular attention was paid to the bacterial flora of the sinuses. Bacteriological swab specimens were collected from the silent sinus during endoscopic surgery. Before the procedure, the nasal vestibule was cleaned and disinfected. The material was collected with a guarded swab stick with the AMIES medium and sent to the laboratory.

The exclusion criteria included patients under 18 years, sinuses without opacification in CT, congenital malformations, sinus hypoplasia, chronic maxillary atelectasis, facial trauma and postoperative status in the head/maxillofacial region.

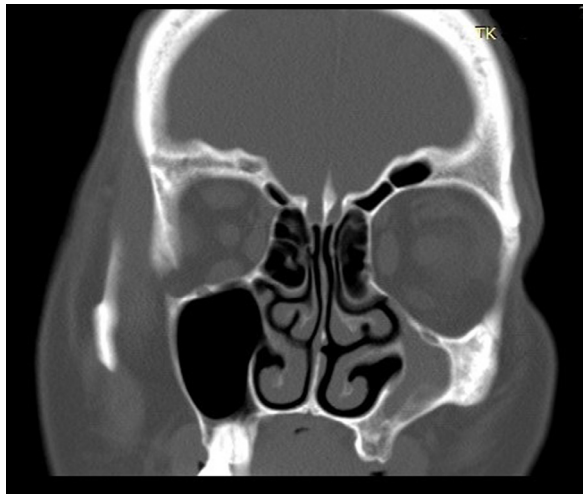

FIGURE 3. CT scan – silent sinus syndrome, radiological findings.
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Results

One hundred five publications were found, 63 of which were reviewed.[10-72] The remaining 42 papers did not contain case reports or were available only in a language other than English or did not have open access.

In 64 reviewed articles, a total of 147 cases of SSS were reported. One hundred and thirty-seven of them referred to the maxillary sinus, 1 – both the maxillary and ethmoid sinuses, 2 – the ethmoid sinus, 1 – the frontal sinus. Six authors described cases of the bilateral maxillary silent sinus syndrome. Among all the reported cases, there was no significant predominance of any side. Patients' ages varied from 19 to 78.

The authors were mainly interested in the medical history and potential etiology of SSS. Fifteen patients reported previous facial injury or surgery. One patient underwent orbital decompression due to Graves' disease. Eleven patients reported symptoms of sinusitis in their medical history. Forty-nine cases were qualified as idiopathic. In 64 cases, the patient's medical history regarding the potential etiology of SSS was not mentioned. Other authors associated the occurrence of SSS with: Botox treatment in the periorbital region, radix relicta, roller coaster ride, pregnancy, sinonasal malignancy, and thyroid eye disease. None of the authors focused on bacteriology. Two of them[14,24] reported no significant bacteriological growth.

Sixty-seven patients were treated only by endoscopy, 33 underwent endoscopy and orbital floor reconstruction, 19 - endoscopy and septoplasty. Other ways of treatment were orbital floor decompression only, hyaluronic gel injection, balloon sinuplasty, and myectomy of the inferior oblique muscle.

In the period from September 2017 to December 2020, 1,344 patients (580 women and 764 men) underwent ESS due to sinus problems at the Department of Laryngology and Laryngological Oncology in Katowice-Ochojec. The silent sinus syndrome was diagnosed in 7 patients, 4 men and 3 women. One case of SSS was diagnosed at the Department of Otorhinolaryngology and Oncological Laryngology in Zabrze. The average age of diagnosis was 45.4 years (the youngest patient was 31, the oldest was 75). All of the described SSS cases were idiopathic. Patients did not meet all the criteria of CRS according to EPOS 2012/2020.[73,74] Most frequent symptoms were: headaches, nasal congestion and facial asymmetry. CT scans revealed maxillary sinus volume loss with its opacification, lowering of the inferior orbital wall, lateral retraction and hypoplasia of the uncinate process and widening of the nasal meatus ipsilaterally, whereas other sinuses were intact. Lowering and destruction of the inferior orbital wall were observed. However, there were no vision disturbances. The decision for surgical intervention was made on the basis of radiographic findings (Fig. 3,4).

In each case, the treatment was limited to endoscopic sinus surgery (uncinectomy and antrostomy).

In all patients, the bacterial flora of the silent sinus was examined. In 3 cases, Gram-positive bacteria growth was observed (*S. aureus, S. epidermitis*). In 4 cases, Gram-negative bacteria were identified (*Klebsiella oxytoca, E. coli, Pseudomonas aeruginosa*). In 1 case there was no significant bacterial growth.

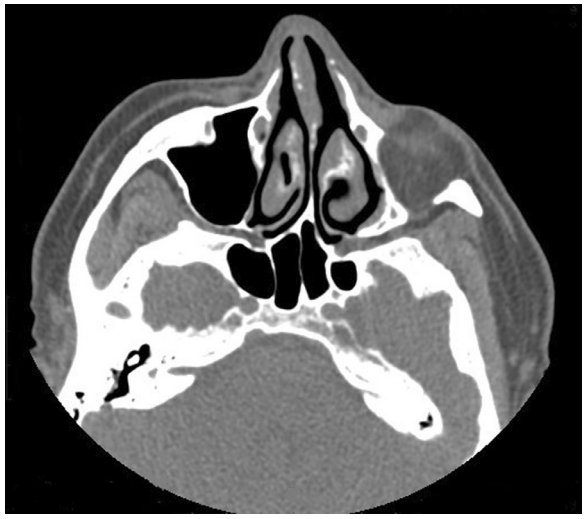

FIGURE 4. CT scan – silent sinus syndrome, radiological findings.
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In Table 1 we present all data about patients with SSS treated in Katowice-Ochojec and Zabrze.

Discussion

The pathophysiology of SSS remains uncertain. It was originally suggested to be an idiopathic disorder. Currently, it is believed that the cause is the obstruction of osteomeatal complex. Narrowing of the natural opening of the maxillary sinus results in sinus hypoventilation and discharge. The resorption of secretion leads to the generation of subatmospheric pressure in the maxillary antrum causing maxillary sinus collapse.[4, 8, 75-77]

It should be remembered that the collapse of the eye in the orbit may occur as a result of physiological processes such as dehydration or loss of fat tissue in the orbit, resulting from pathology such as orbital or eye trauma, in Wegener's granulomatosis and systemic disease (sclerodermia), post radiotherapy atrophy, neoplastic metastases or Parry-Romberg syndrome.

To diagnose SSS, the following criteria from anamnesis could be useful[78]:

- no episodes of acute sinusitis in the past 6 months and no history of chronic sinusitis (CRS)
- no data suggesting a history of orbital trauma (including surgery) or enophthalmos of a different etiology,
- no data suggesting congenital deformities of the nasal cavity and sinuses.

The ophthalmological and radiological examination features of SSS are presented in Table 2.[3,79]

In contrast to the criteria mentioned above, Sopokar and Buono suggest that 36% of patients with SSS suffered from CRS.[2,3] In the literature reviewed in this paper, sinusitis symptoms were present in 11 cases reported as SSS. Moreover, 15 cases of SSS from the reviewed literature were associated with previous maxillofacial/orbital trauma or surgery. This may suggest that the criteria of SSS remain unclear or misunderstood.

SSS most frequently concerns the maxillary sinus, without the predominance of either the left or right side. There are single descriptions of the bilateral silent sinus syndrome in the literature.[28, 67, 52] Brown and McArdle[14,69] reported 2 cases of the silent ethmoid sinus syndrome and Naik described 1 patient with the silent frontal sinus.[16]

The available literature does not provide a detailed analysis of the bacterial flora of the operated silent sinuses. It has been found that bacterial cultures are usually negative.[10, 14, 24] In the authors' own material, *S. aureus, S. epidermitis, E. coli* and *Klebsiella oxytoca* were found.

The rare prevalence of SSS makes it difficult to assess the impact on the etiology and course of SSS depending on the appearance of the bacteria cultures, and requires further research.

The authors of the analysed papers agree on the management of SSS. The most important is to improve the drainage of the sick sinus. Endoscopic surgery of the maxillary sinus is proposed in cases without facial deformity and diplopia.[19,20] Depending on the degree of enophthalmos and hypoglobus, simultaneous, or delayed reconstruction of the superior wall of the maxillary sinus is considered.[10, 22, 28, 31, 44, 65, 66, 80]

In the authors' material of 1,344 patients who underwent endoscopic sinus surgery only 7 patients were diagnosed with SSS. This may suggest that those with affected vision, diplopia or significant enophthalmos first turn to ophthalmologists or maxillofacial surgeons instead of otorhinolaryngologists. It is important for all those specialists to be aware of this syndrome and its typical symptoms.

The presented criteria of SSS should shorten the diagnostic process in order to prevent patients from developing changes in the orbit, vision disorders or face deformation. Early diagnosis gives a chance for less invasive surgery limited to antrostomy, which is relatively safe.

In conclusión, early diagnosis of SSS may help avoid orbital complications and may limit surgical intervention including endoscopic surgery. Careful analysis of imaging examinations facilitates safe opening of the silent sinus. Further research into bacteriology may

Table 1. PATIENTS WITH SSS TREATED IN OUR DEPARTMENTS – DATA SUMMARY

Patient	Age	Sex	Symptoms	Ophtalmological Symptops	Comorbidities	Permanent Treatment	Medical History	Allergies	Cigarette Smoking	Sinus Culture	Post-Operative Effect
1	33	F	Recurring headaches Running nose	-	Chronic glomerulonephritis	-	Broken nose (more than 10 yr before) Tooth (18 and 28) extraction Tonsillectomy (10 yr before) Sinus puncture (11 yr before)	-	No	Escherichia coli	Positive
2	40	M	Headaches Nasal congestion	-	-	-	Not relevant	Wasp and bee venom	No	Klebsiella oxytoca	Positive
3	31	M	Headaches Nasal congestion	Orbital asymmetry	Depression	Escitalopram	Septoplasty (12 yr before) Tonsillectomy (in childhood)	Acari, dogs, cats, birch	No	Staphylococcus aureus	Partially positive
4	55	F	Headaches Snooring	-	Osteoarthritis	-	Broken nose and septoplasty in childhood Tonsillectomy (35 yr before)	Acari	Yes	Escherichia coli Klebsiella oxytoca	
5	33	F	Maxilla pain	Eyelid asymmetry	Reactive arthritis Metylopredniozolone	Septoplasty (4 months before)	Artemisia	Yes	Staphylococcus epidermitis	Positive	
6	75	F	Headaches Nasal congestion	-	Peripheral facial nerve palsy	-	Cataract operation (4 yr before) Carcinoma basocellulace of nose skin (5 yr before)	Penicillin	yes	Pseudomonas aeruginosa	Partially positive
7	46	M	Headaches Nasal congestion Running nose	-	-	-	-	-	No	-	Positive
8	43	F	Headaches	-	-	-	Sinus puncture in childhood Head trauma in childhood (without any fracture)	-	No	Staphylococcus aureus	Partially posotive

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Table 2. THE OPHTHALMOLOGICAL AND RADIOLOGICAL EXAMINATION FEATURES OF SILENT SINUS SYNDROME

Ophthalmological	Radiological
• enophthalmos ranging from 1 to 6 mm (Hertel exophthalmometry is proposed for objective evaluation of enophthalmos) • hypoglobus ranging from 0 to 6 mm • apparent retraction of the eyelid • lagophthalmos • normal eye mobility and visual function • sinking of the eye • orbital asymmetry • deepening of the superior sulcus • others (eg, diplopia, ptosis, blepharoptosis or oscillopsia)	• maxillary sinus volume loss • lateral retraction of the uncinate process • partial or complete opacification of the maxillary sinus with / without air-fluid levels and thickened mucosa • lowering of the inferior orbital wall, often with osteopenia, partial or complete resorption • inward retraction of other sinus walls • nasal septum deviation often towards the involved sinus • lateralization of the ipsilateral middle nasal concha

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help to understand the pathophysiology of the silent sinus syndrome.

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