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CASE REPORT

Silent sinus syndrome associated with natural childbirth

Zespół cichej zatoki związany z porodem siłami natury

Aleksandra Zofia Sobolewska[1,2], Pedro Clarós[1]

[1]Clinical Otolaryngological Department, 5th Military Clinical Hospital in Krakow, Poland
[2]Clarós Clinic, Barcelona, Spain

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ABSTRACT: Spontaneous, painless enophthalmos, hypoglobus with orbital floor resorption and maxillary sinus collapse on the ipsilateral side is recognised as a rare condition known as the silent sinus syndrome. This paper aimed to present an unusual association of natural childbirth and the onset of orbital floor displacement caused by silent sinus syndrome. We wanted to present a case of a 31-year-old woman who presented with a 3-month history of painless, progressive right enophthalmos otherwise utterly asymptomatic and who developed symptoms shortly after natural childbirth. That association has never been presented before in literature. We also wanted to discuss the pregnancy-related nasal congestion. We present our experience with this case treated with a single-stage procedure, focusing on the advantages of this one-step approach.

KEYWORDS: enophthalmos, hypoglobus, orbital floor displacement, childbirth, orbital reconstruction, silent sinus syndrome

STRESZCZENIE: Spontaniczne, bezbolesne zapadnięcie gałki ocznej (enophthalmos) z jej obniżeniem (hypoglobus), resorpcją ściany oczodołu i zapadnięciem się zatoki szczękowej po tej samej stronie stanowi rzadki zespół kliniczny określany jako zespół cichej zatoki (ang. silent sinus syndrome). Niniejszy artykuł ma na celu przedstawienie nietypowego związku między porodem siłami natury i przemieszczeniem ściany oczodołu w przebiegu zespołu cichej zatoki. Opisano przypadek 31-letniej kobiety, która zgłosiła się z trzymiesięcznym wywiadem bezbolesnego postępującego zapadnięcia gałki ocznej po stronie prawej bez żadnych innych dolegliwości, u której objawy rozwinęły się po porodzie siłami natury. Nigdy wcześniej nie opisywano w literaturze podobnego związku. W artykule omówiono również nieżyt nosa związany z ciążą. Przedstawiono doświadczenia autorów w leczeniu jednoetapowym z uwzględnieniem korzyści takiego podejścia.

SŁOWA KLUCZOWE: enophthalmos, hypoglobus, przemieszczenie ściany oczodołu, poród, rekonstrukcja oczodołu, zespół cichej zatoki

INTRODUCTION.

Spontaneous, painless enophthalmos, hypoglobus with orbital floor resorption and maxillary sinus collapse on the ipsilateral side was first defined by Soparkar as silent sinus syndrome (SSS) [1]. This rare condition was described 30 years earlier for the first time by Montgomery, in 1964 [2]. Formerly, SSS had been believed to be idiopathic. However, after clarification of the pathogenesis, iatrogenic and a post-traumatic origin is commonly accepted.

Enophthalmos and hypoglobus are the consequence of orbital floor collapse due to maxillary sinus atelectasis. Nowadays, researchers share the opinion that orbital floor bone resorption is a process caused by negative maxillary antral pressure because of obstruction of the infundibulum, which generates negative pressure.

According to literature, the syndrome occurs between the third and fifth decade of life but very rare among children, with no gender predilection [3][4]. Radiological features of this rare

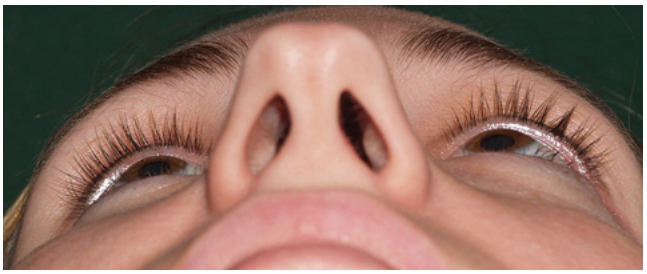

Fig. 1. Preoperative submental view: right enophthalmos, hypoglobus, slight ocular dystopia.

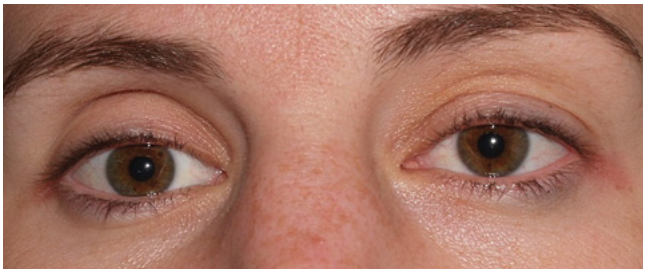

Fig. 2. Preoperative frontal view: right enophthalmos, deeper upper sulcus, slight ocular dystopia.

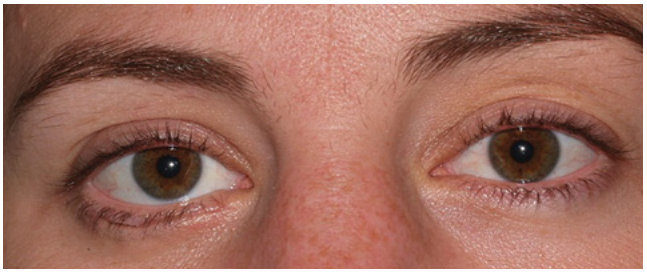

Fig. 3. Postoperative frontal view - 2 weeks after procedure.

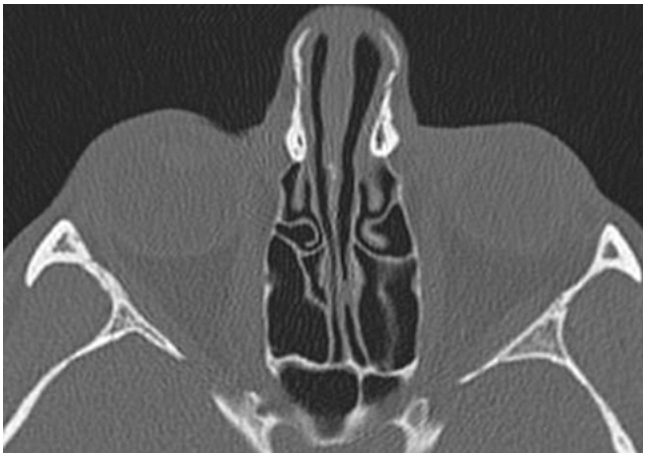

Fig. 4. CT image of June 2016 showing no apparent asymmetry of the eyeballs.

condition include inferior displacement of the orbital floor, collapsed maxillary sinus, infundibulum occlusion, uncinate process lateralisation, hypoplasia of the middle concha and the width of the middle meatus [5].

We would like to present our recent case of SSS. Patient's history is unique because of a well-established onset of the disease. Moreover, natural childbirth and nasal congestion during pregnancy could have been a possible trigger mechanism for the development of the silent sinus syndrome in that case.

To the best of authors' knowledge, an association between silent sinus syndrome and childbirth has never been reported before in literature.

We would like to share our experience on the treatment of the silent sinus syndrome combined with endoscopic sinus surgery and reconstruction of the orbital floor using Medpor® titanium orbital floor implant.

CASE REPORT

A 31-year-old woman (Fig. 1, 2) presented with a 3-month history of painless, progressive right enophthalmos, otherwise utterly asymptomatic, in August 2017.

Before this event, she had been a patient of one of the local otolaryngology clinics in June 2016, because of a sudden loss of olfaction. She underwent a standard ENT evaluation and CT imagining (Fig. 4, 5, 6), with no visible pathologies.

In the second half of 2016, the patient became pregnant for the first time in her life; the pregnancy course was normal. In April 2017 she gave birth to her first child. The natural labour was uneventful. The baby was born at full term and received 10 points in APGAR scale. Two weeks later, she experienced symptoms of mild rhinosinusitis.

Next month, she noticed a sudden difference in face symmetry. At the beginning of July, the patient decided to seek medical assistance because of apparent facial asymmetry. Detailed ophthalmic examination showed right enophthalmos with hypoglobus, giving a sunken sulcus appearance of the right upper lid. The rest of the ocular examination and orthoptic assessment was normal, no ocular movement impairment, no diplopia. Also, facial trauma, allergies and CRS were absent in her past medical history. Photographic documentation was obtained to evaluate facial symmetry.

Hertel exophthalmometer showed 15mm in the left eye and 12 mm in the right one, with hypoglobus of 5 mm. Endoscopic examination of the nasal cavity showed no pathological chan-

ges. CT imaging (Fig. 7, 8) revealed right enophthalmos, with inferiorly displaced and eroded orbital floor. Also, complete opacification and reduced volume of the right maxillary sinus was present. Partial opacification of anterior ethmoid cells and thinned lamina papyracea were noted. The patient was qualified for endoscopic drainage of the maxillary sinus, and reconstruction of the orbital floor. Endoscopic sinus surgery was performed by the right endonasal approach to the middle meatus. The concavity of the maxillary wall and lateralisation of the uncinate process was confirmed. Uncinectomy and antrostomy were carried out. Subcillary incision was used to approach the orbital floor. Medpor® titanium (Stryker®) implant was fixed and fitted with a proper aesthetic result (Fig. 3). The patient was discharged day after that. Her recovery was uneventful. The ophthalmic evaluation was scheduled one month after the surgery.

Hertel exophthalmometer results showed 15 mm on the left side, and 14 mm on the right side. The patient did not report any other complaints. Histopathology report described signs of compression atrophy of the respiratory mucosa, showing a compacted seromucous gland surrounded by stromal fibrosis (Fig. 11, 12). The superficial respiratory lining epithelium appeared discretely hyperplastic, showing slightly pleomorphic nuclei and eosinophilic apical cytoplasm, sometimes accompanied by cilia or muco-eosinophilic secretion. All this was accompanied by bony spicules of the wall of the maxillary sinus, with signs of atrophy. At its periphery, apoptotic cell remains were detected, with compact chromatin and certain nuclear pleomorphism surrounded by eosinophilic granular cytoplasm. Pathologist's findings were consistent with the silent sinus syndrome in the maxillary sinus. A control CT scan (Fig. 9, 10) six weeks after the procedure showed acceptable symmetry of the eyeballs. Also, slight thickening of mucosa with no exudate was present in the maxillary sinus and antrostomy was patent. No additional pathology was present. The patient had regular check-ups for over a year, reported no complains, with good toleration of the implant and satisfying aesthetic result. She was counselled that in case of a possible, next pregnancy, a Caesarean section will be advised for childbirth.

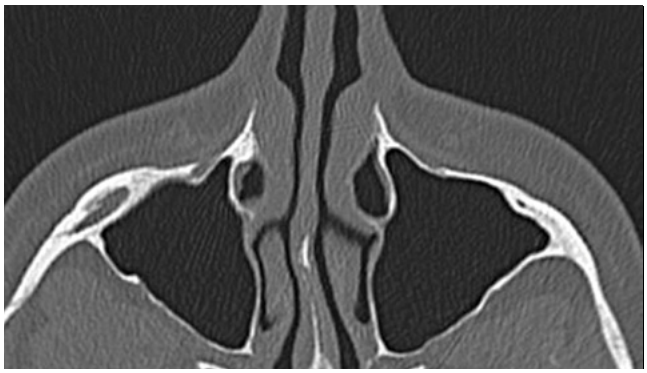

Fig. 5. CT image of June 2016 (axial view) showing a healthy and well-developed right maxillary sinus.

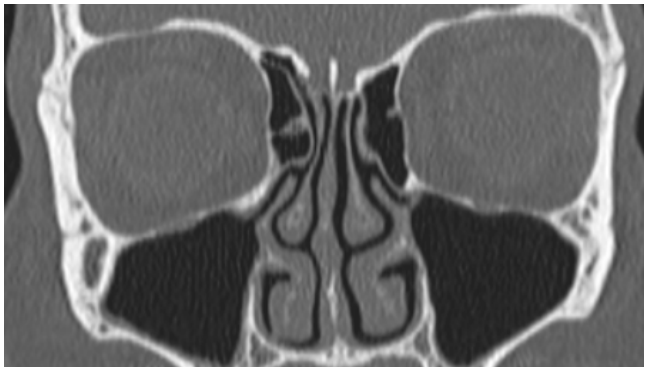

Fig. 6. CT image of June 2016 (frontal view) showing normal maxillary sinuses with patent infundibulum and no asymmetry of the orbital floor.

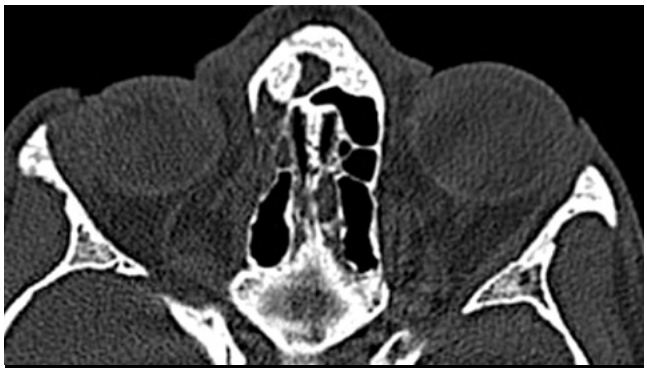

Fig. 7. Preoperative CT scan (axial view) showing hypoglobus, thinned lamina papyracea and opacification of anterior ethmoid cells.

DISCUSSION

Silent sinus syndrome has a distinctive clinical presentation and characteristic diagnostic features. Sudden orbital floor displacement with no history of typical symptoms of chronic sinusitis but with distinctive computed tomography imaging usually should allow for a proper diagnosis. Silent sinus syndrome remains an important concept in neuro-ophthalmology. Despite being extensively described in the past 20 years, the pathophysiology of this syndrome has not been explained yet. Several possible mechanisms have been pointed to.

The „flow obstruction," theory states that during maxillary ostium occlusion, trapped secretion leads to inflammation and erosion of the sinus walls. Additionally, mucosal gas absorption generates negative pressure which causes inferior displacement of the orbital floor [6].

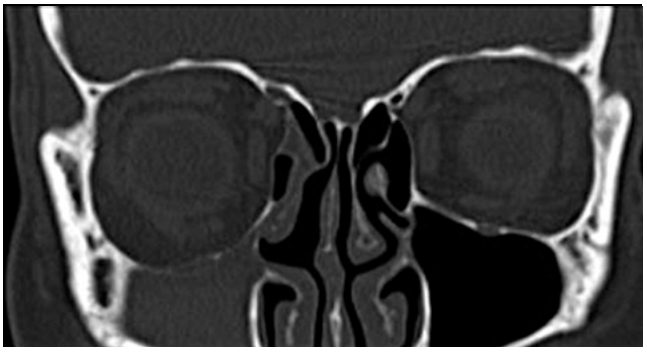

Fig. 8. Preoperative CT scan (frontal view) showing inferior displacement of the orbital floor, lateralisation of the middle turbinate and opacification of the maxillary sinus.

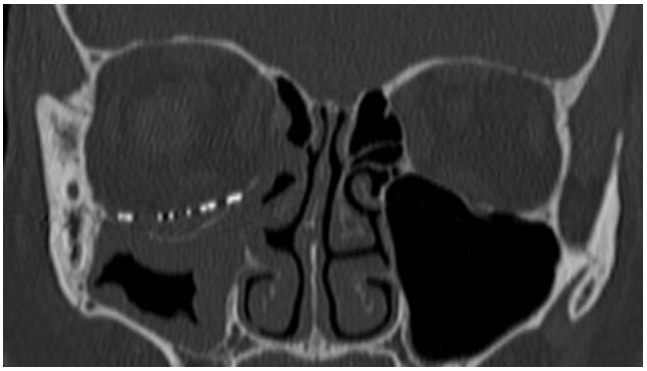

Fig. 9. Postoperative CT scan (frontal view) showing placement of an orbital implant. Slight thickening of mucosa with no opacification of the maxillary sinus.

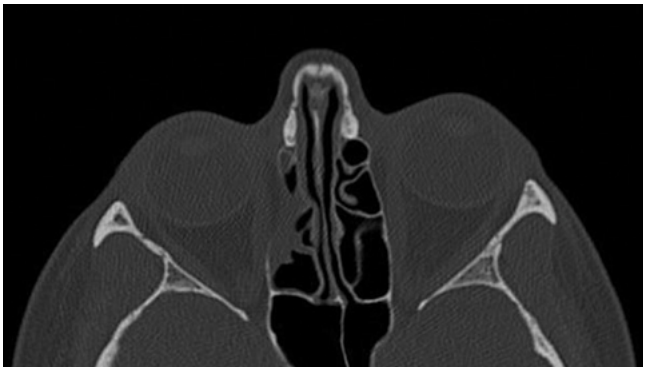

Fig.10. Postoperative CT scan (axial view) showing symmetry of eyeballs.

Another possible mechanism involves disturbance of communication between the sinus and the pterygopalatine fossa, which generates pressure gradient during the act of mastication. Constriction and relaxation of masticatory muscles may cause aspiration and implosion of the sinus walls [7].

We aimed to present this particular case because the reason for "flow obstruction" in the maxillary sinus ostium may be nasal congestion caused by hormonal changes during pregnancy and extensive effort during labour. Characteristic, in that case, was a short period between natural childbirth and the onset of the disease.

Nasal congestion that is not present before pregnancy represents a separate clinical entity, called "pregnancy-related rhinitis", defined as nasal congestion present in the last 6 or more weeks of pregnancy without other signs of respiratory tract infection and with no known allergic cause, disappearing completely within two weeks after delivery [8].

According to the literature, this condition was reported to affect 20% to 40% of pregnant women in all trimesters [8][9].

Elevated oestrogen levels cause nasal congestion. This explanation follows from the studies of Toppozada et al. based on nasal mucosa biopsies from women taking contraceptive pills and from pregnant women with or without nasal congestion [10].

In more recent studies several mechanisms were proposed; these include the effects of pregnancy hormones through the shift of increased plasma volume to extracellular space [11], enhanced expression of H1 receptors on nasal epithelial cells [12], increased gland secretion, and vasodilation caused by both vasoactive intestinal peptide and cholinergic action [13].

In the presented case, one of possible explanations for the development of SSS was mild nasal congestion due to pregnancy which led to obstruction of the infundibulum. Extensive effort during natural childbirth may be considered as a triggering mechanism for the development of maxillary sinus occlusion.

First CT imaging performed one year earlier showed no sign of rhinitis or any symptom of altered orbital floor anatomy. Probably, the maxillary sinus ostium occlusion induced by hormonal changes due to pregnancy could have impaired sinus ventilation and led to entrapment of the secretion causing inflammation and bone resorption. However, the event of natural childbirth led to rapid displacement of the orbital floor. The very sudden onset of the disease is characteristic and was commonly reported in the literature [14].

Association of pregnancy rhinitis and natural childbirth with silent sinus syndrome has not been reported before. Nevertheless, hormonal changes in the nasal mucosa due to gestation, extensive effort during natural childbirth and unfavourable nasal anatomy of the ostiomeatal complex provide a reasonable answer. We consider these facts to play a role in the development of silent sinus syndrome symptoms in our case.

Furthermore, an essential factor here was proper orbital reconstruction because of patient's high expectations regarding the aesthetic results.

Treatment of the silent sinus syndrome requires maxillary sinus drainage and usually an approach to restoration the orbital anatomy. In the literature, various materials were recommended for reconstruction, such as autogenous grafts, like fragments of the iliac bone or artificial material, e.g. porous polyethene implants. Usually, material preferences are a matter of surgeon's personal experience. Our preferred method is the implementation of titanium implants which allows for satisfying aesthetic results and shorter time of the procedure, with acceptable patient's tolerance. The use of pre-prepared biocompatible implants allows for a satisfactory aesthetic result, favours shorter time of surgery, and reduces blood loss during the procedure. Surgical treatment of the silent sinus syndrome should be based on the multidisciplinary experience of otolaryngologists anf ophthalmologists.

CONCLUSION

The silent sinus syndrome is described as spontaneous enophthalmos, orbital floor resorption and maxillary sinus collapse on the affected side with a characteristic radiological presentation. Therefore, it should always be included in the differential diagnosis of enophthalmos.

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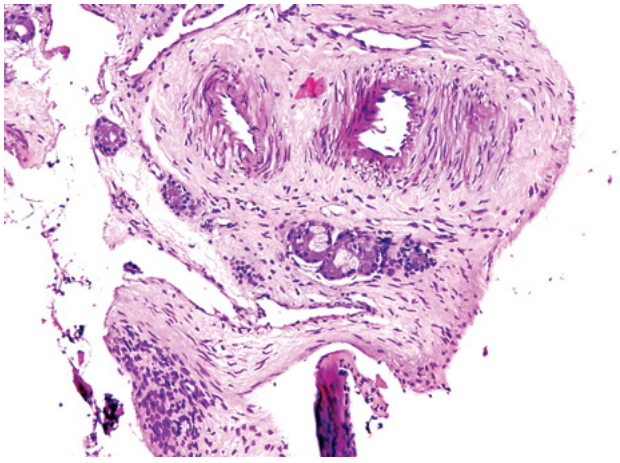

Fig. 11. Small magnification. Hematoxylin and eosin stain. Transversal section of two arterioles with a thickened wall, surrounding three atrophic glands. At the bottom, centrally, a vertical spicule of atrophic bone.

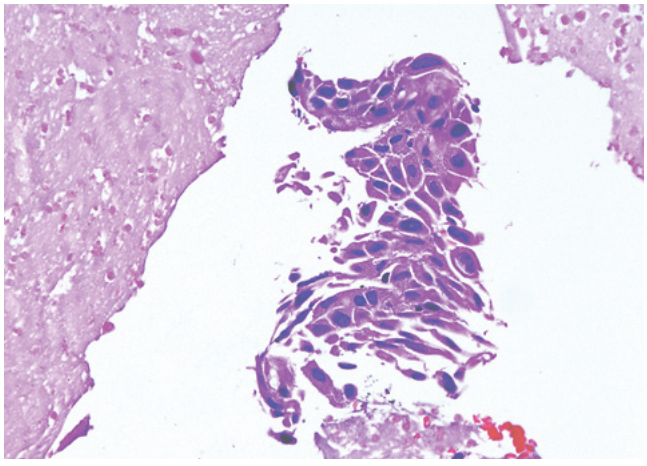

Fig. 12. Small magnification. Hematoxylin and eosin stain. Margin of the central necrotic ball with a neighboring cluster of apoptotic cells.

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Corresponding author: Aleksandra Sobolewska; Clinical Otolaryngological Department, 5th Military Clinical Hospital in Krakow, Poland, Wrocławska Street 1-3, 30-901 Krakow, Poland; E-mail: olka.sobolewska@doctoral.uj.edu.pl

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