

CASE REPORT**Silent sinus syndrome - Case presentation****Veronica Trombitas¹, Alina Nagy¹, Diana Vlad¹, Aranka Ilea², Silviu Albu¹**¹II-nd Department of Otolaryngology, "Iuliu Hatieganu" University of Medicine and Pharmacy Cluj-Napoca, Romania²Department of Oral Rehabilitation, Oral Health and Dental Office Management, University of Medicine and Pharmacy Cluj-Napoca, Romania**ABSTRACT**

BACKGROUND. Silent sinus syndrome (SSS) is known as a relatively new pathology with clinical characterizations, represented by the facial asymmetry determined by progressive enophthalmos and hypoglobus and the absence of nasal symptoms. The theory that sustained this pathology is based on the negative maxillary pressure and the chronic evolution of symptomatology.

CASE REPORT. A 49-year-old white female presented in our clinic for facial asymmetry, right facial pressure, right depression of the orbital floor and hypoglobus, without diplopia. The CT scan showed an opaque right maxillary sinus with the depression of the orbital floor.

CONCLUSION. The pathogenesis, clinical and imagistic features are the most important in the SSS for diagnosis and correct treatment.

KEYWORDS: silent sinus syndrome, enophthalmos, facial asymmetry, chronic maxillary atelectasis.

INTRODUCTION

SSS was described in literature fairly recently, the first case being reported in 1964 by Montgomery¹, but the name was coined 30 years later by Soparker et al.^{2,3}. The syndrome is a rare pathology characterized by unilateral progressive enophthalmos and/or hypoglobus⁴, which consists in a unilateral painless facial asymmetry⁵, not associated with orbital trauma or surgery, nor with congenital facial asymmetry or rhinosinusitis symptoms or history⁶, but with a chronic evolution of weeks or months. The pathogenesis consists in a maxillary hypoplasia with negative maxillary pressure, determined by the obstruction of the infundibulum⁷ associated with the inflammation of sinus mucosa¹ and a subsequent association of the orbital floor collapse³ and pseudoretraction of the upper eyelid. The majority of patients, mostly in their third to fifth decades of life⁷, address firstly to the ophthalmologist and sec-

ondary to the ENT physician and the diagnosis is established clinically and imagistically⁵. CT findings consist in the unilateral maxillary sinus opacification with or without fluid level, infundibular obstruction⁸, maxillary atelectasis and orbital floor erosion with collapse of the orbital contents^{6,7}.

The aim of this case report is to highlight the importance of the correct management of the SSS, starting with a suitable diagnosis.

CASE REPORT

We present the case of 49-year-old white female, with a six-month history of right facial asymmetry and right facial pressure, without diplopia, diagnosed by the ophthalmologist as upper eyelid ptosis and proposed for ophthalmology surgery. The patient presented in our department (ENT Department at CF



Figure 1 CT-scan coronal section reveals the complete opacification in the right maxillary sinus, retraction to the orbital floor, lateralization of the uncinete

Hospital, Cluj-Napoca) for the aforementioned signs, without rhinosinusitis symptoms or history of injury or surgery. The ENT examination revealed facial asymmetry determined by right enophthalmos and an upper eyelid pseudoptosis; also, nasal endoscopy with 0 and 30° rigid endoscope showed a moderate lateralization of the middle turbinate, lateralization of the intersinonasal wall and uncinete process raising the suspicion of an inflammatory process.

The CT scan revealed important modifications: a complete opacification of the maxillary sinus in the *coronal sections*, maxillary sinus asymmetry, retraction to the orbital floor, lateralization of the uncinete (Figure 1); the *axial sections* showed retraction of the right maxillary posterior wall and maxillary sinus hypoplasia (Figure 2).



Figure 3 Maxillary antrostomy aspect

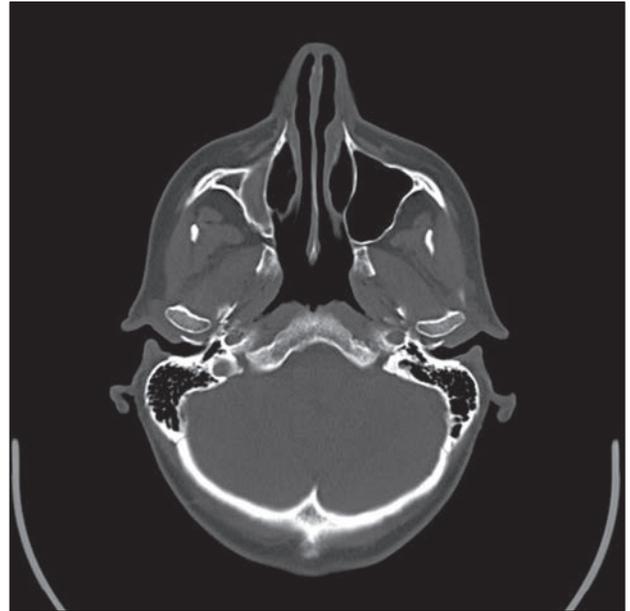


Figure 2 CT-scan axial section shows retraction of the right maxillary posterior wall and maxillary sinus hypoplasia

The patient received surgical treatment via Functional Endoscopic Sinus Surgery (FESS)⁹ and maxillary antrostomy was performed (Figure 3); the right nasal cavity was packed using resorbable packing (Meroce1)¹⁰. The surgical treatment was associated with antibiotics and saline lavage for 7 days. Mucosa samples were collected around the antrostomy for a complex examination, which showed the presence of inflammatory markers (Goblet cells, eosinophils, macrophages, fibrosis, epithelial hypertrophy, subepithelial hypertrophy and edema) (Figure 4).

The first follow-up visit was one week after surgery, for removing clots, crusts and secretions; the second visit was after one month, when the nasal wound healing was completely achieved (Figure 5) and the facial asymmetry was improved.

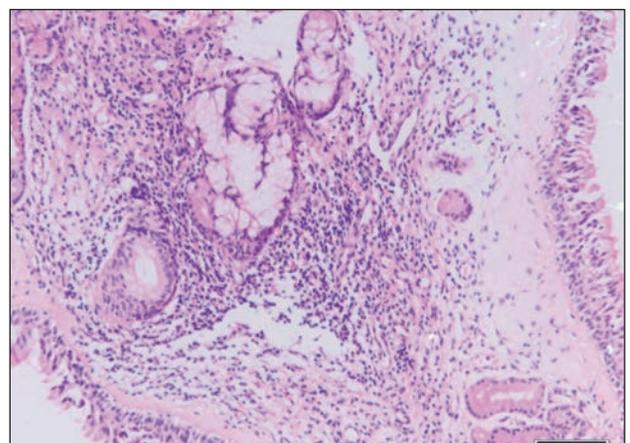


Figure 4 Sinus mucosa with inflammatory aspect (H&E staining, 10X)

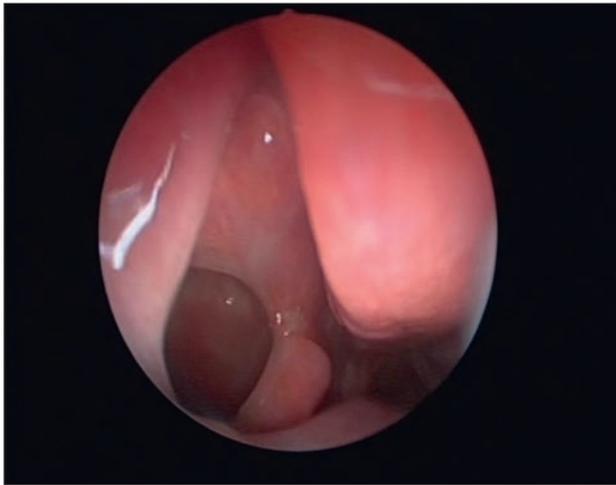


Figure 5 Nasal endoscopy aspect. Follow-up visit, one month after surgery

DISCUSSIONS

The changes in the structure of maxillary and orbital walls are frequently met and most often these changes are accompanied by nasal blockage, rhinorrhea and headache, facial or dental pain^{11,12}. SSS is a challenging diagnosis because it can be diagnosed in a subgroup of patients, who have the sinus and orbital changes, but no specific symptoms of chronic rhinosinusitis. Kass et al.¹³ demonstrated in their study, comparing the pressure of the chronic maxillary atelectasis sinus with chronic maxillary sinusitis sinus and with a normal maxillary sinus, that the negative pressure is the basis of the chronic maxillary atelectasis. The negative pressure originates in the osteomeatal complex obstruction, determines bone remodeling and one of the most important signs, enophthalmos^{2,6,7}.

The SSS have some important characteristics; several of these represent diagnosis criteria:

1. Unilaterality of issues, sustained by the CT scan,
2. Absence of documented nasosinus or orbital trauma, surgery or congenital abnormality, no history of chronic rhinosinusitis^{3,4,7},
3. Poverty in local symptoms, rarely found facial or dental pressure, diplopia,
4. The notable changes are facial asymmetry, enophthalmos, hypoglobus,
5. Radiologic examination is essential, CT scan changes are pathognomonic,
6. Histologic examination highlights the inflammatory character of nasal mucosa and excludes purulent inflammation or malignancy⁴.

The management of SSS follows two objectives: maxillary sinus drainage and orbital restoration. These goals are achieved by surgical treatment, especially FESS. Although the surgery by Caldwell-Luc technique could obtain good results, the high associated morbidity

invalidates this approach¹⁴. Because the antrostomy (FESS) is a minimally invasive procedure and restoring a positive maxillary pressure could resolve the orbital changes, it should be indicated as the primary option. The orbital reconstruction is controversial in literature, some reports indicating a single-stage operation with simultaneous maxillary antrostomy and orbital implants^{4,6,7,15}, but a complete remodeling of the implicated bone is achieved two months after surgery, so that the orbital defect could be resolved at a later time if necessary⁴.

CONCLUSIONS

Starting right from the name, SSS is a controversial pathology, with many pathogenesis theories and many strategies for treatment. A careful diagnosis could direct to a correct management of SSS.

Conflict of interest: The authors have no conflict of interest.

Contribution of authors: All authors have equally contributed to this work.

REFERENCES

1. Dumitrescu D., Fanuta B., Stepan A.E., Fronie A.I., Dumitrescu C.I., Martu M.C., Surlin P., Surlin V., Popescu M. - Silent sinus syndrome—report of a case. *Rom J Morphol Embryol.*, 2015;56(1):229-237.
2. Soparkar C.N., Patrinely J.R., Cuaycong M.J., Dailey R.A., Kersten R.C., Rubin P.A., Linberg J.V., Howard G.R., Donovan D.T., Matoba A.Y., et al. - The silent sinus syndrome. A cause of spontaneous enophthalmos. *Ophthalmology.*, 1994;101(4):772-778.
3. Numa W.A., Desai U., Gold D.R., Heher K.L., Annino D.J. - Silent sinus syndrome: a case presentation and comprehensive review of all 84 reported cases. *Ann Otol Rhinol Laryngol.*, 2005;114(9):688-694.
4. Nkenke E., Alexiou C., Iro H., Amann K., Kirchner T., Häusler G., Neukam F.W., Holbach L.M. - Management of spontaneous enophthalmos due to silent sinus syndrome: a case report. *Int J Oral Maxillofac Surg.*, 2005;34(7):809-811.
5. Illner A., Davidson H.C., Harnsberger H.R., Hoffman J. - The silent sinus syndrome: clinical and radiographic findings. *AJR Am J Roentgenol.*, 2002;178(2):503-506.
6. Vander Meer J.B., Harris G., Toohill R.J., Smith T.L. - The silent sinus syndrome: a case series and literature review. *Laryngoscope.*, 2001;111(6):975-978.
7. Cobb A.R., Murthy R., Cousin G.C., El-Rasheed A., Toma A., Uddin J., Manisali M. - Silent sinus syndrome. *Br J Oral Maxillofac Surg.*, 2012;50(6):e81-5. doi: 10.1016/j.bjoms.2011.10.001. Epub 2011 Nov 1.
8. Gocea A., Carlos P., Albu S. - Silent Sinus Syndrome: a case presentation and literature review. *ORL.roVol II. 2/2009:37-41CNCSIS C.*
9. Kennedy D.W. - Functional endoscopic sinus surgery. *Technique. Arch Otolaryngol.*, 1985;111(10):643-649.
10. Konstantinidis I., Tsakiropoulou E., Vital I., Triaridis S., Vital V., Konstantinidis J. - Intra- and postoperative application of Mitomycin C in

- the middle meatus reduces adhesions and antrostomy stenosis after FESS. *Rhinology.*, 2008;46(2):107-111.
11. Boyd J.H., Yaffee K., Holds J. - Maxillary sinus atelectasis with enophthalmos. *Ann Otol Rhinol Laryngol.*, 1998;107(1):34-39.
 12. Albu S., Trombitas V., Vlad D., Emanuelli E. - The influence of spray cryotherapy on wound healing following endoscopic sinus surgery in chronic rhinosinusitis. *Laryngoscope.*, 2015 Mar 17. doi: 10.1002/lary.25257.
 13. Kass E.S., Salman S., Montgomery W.W. - Manometric study of complete ostial occlusion in chronic maxillary atelectasis. *Laryngoscope.*, 1996;106(10):1255-1258.
 14. Kass E.S., Salman S., Rubin P.A., Weber A.L., Montgomery W.W. - Chronic maxillary atelectasis. *Ann Otol Rhinol Laryngol.*, 1997;106(2):109-116.
 15. Sesenna E., Oretti G., Anghinoni M.L., Ferri A. - Simultaneous management of the enophthalmos and sinus pathology in silent sinus syndrome: a report of three cases. *J Craniomaxillofac Surg.*, 2010;38:469-472.