Posttraumatic Delayed Enophthalmos: Analogies with Silent Sinus Syndrome? Case Report and Literature Review

Gabriele Canzi, MD^{1,2} Valeria Morganti, MD² Giorgio Novelli, MD² Alberto Bozzetti, MD^{1,2}
Davide Sozzi, MD^{1,2}

¹ Emergency Department, Maxillofacial Surgery Unit, Milan, Italy

² Department of Surgery and Interdisciplinary Medicine, Maxillo-Facial Surgery Unit, University of Milano-Bicocca, Monza, Italy

Address for correspondence: Gabriele Canzi, MD, Maxillofacial Surgery Unit, Emergency Department, Ca' Granda Niguarda Hospital, 3 Piazza Ospedale Maggiore, 20162 Milan, Italy
(e-mail: gabriele.canzi@ospedaleniguarda.it).

Craniomaxillofac Trauma Reconstruction 2015;8:251–256

Abstract

Acute posttraumatic enophthalmos is a well-known symptom occurring in orbital blowout fractures. Its late onset in the absence of radiologic evidence of displaced fractures is rare and traditionally attributed to ischemic liponecrosis or fibrotic scarring of endo-orbital soft tissues. In this article, we describe a case of facial trauma, diagnosed and treated at the Maxillo-Facial Surgical Department of Hospital Ca' Granda Niguarda of Milan, in which delayed monolateral enophthalmos is associated with CT evidence of remodeling of orbital walls attributed to atelectasis of the maxillary sinus, as occurs spontaneously in patients suffering from silent sinus syndrome (SSS). Despite that classic criteria exclude traumatic etiology of SSS, recent literature suggests the possibility to include it. Our case is the first reported in literature supported by complete clinical and radiological documentation obtained before and after the condition established itself. The analogy with cases of spontaneous obstacle of aeration allows us to choose “two-step” surgical treatment with endoscopic uncinectomy and antrostomy and a delayed surgical correction of orbital volume to improve aesthetic results. The case described in this article and the review of the literature may focus physicians' attention on evaluating the possible traumatic changes in the physiologic sinus drainage system.

Keywords

- orbital fracture
- orbital wall deformities
- enophthalmos
- silent sinus syndrome
- sinus atelectasis

Acute posttraumatic enophthalmos, in absence of direct injuries of the globe is generally due to blowout fractures with herniation of the orbital soft tissues in the adjacent paranasal sinuses. It can be associated with visual deficits, diplopia, eye movement defects, hemosinus, and locoregional sensibility defect. In patients with radiologic evidence of orbital soft tissue herniation in the paranasal sinuses or increases in significant orbital volume, enophthalmos can be clinically recognized immediately or several days after periorbital swelling and/or emphysema reduction.^{1,2} In the absence of these radiological findings, enophthalmos is rare

and may be attributed to volumetric orbital content reduction induced by ischemic liponecrosis or posttraumatic fibrotic scarring of orbital soft tissues.^{1,3}

Late enophthalmos is uncommonly associated with the remodeling of orbital walls in nondisplaced fractures without primary orbital volume modifications. We present a case of bilateral multifragmentary composed orbitozygomatic fracture, which was diagnosed and treated at the Maxillo-Facial Surgical Department of Hospital Ca' Granda Niguarda of Milan, with delayed monolateral enophthalmos 2 months after trauma may be due to atelectasis of the maxillary sinus.

received

May 23, 2014

accepted after revision

July 27, 2014

published online

January 14, 2014

Copyright © 2015 by Thieme Medical Publishers, Inc., 333 Seventh Avenue, New York, NY 10001, USA.
Tel: +1(212) 584-4662.

DOI <http://dx.doi.org/10.1055/s-0034-1399799>.
ISSN 1943-3875.

Fig. 1 (a, b) Clinical view of the patient 5 days after the trauma, prior to bifocal mandible fracture surgery.

Case Report

The patient is a 27-year-old man who suffered facial trauma by crushing at the workplace (►Fig. 1a, b).

He was transported to the emergency department of the trauma center, Hospital Ca' Granda Niguarda of Milan. Clinical evaluation showed malocclusion with right mandibular parasymphysis mobility and lower jaw movement limitation, but without upper jaw mobility or nasal/oral bleeding; bilateral periorbital ecchymosis was present but without diplopia, bilateral ocular movement defects nor esoenophthalmos, or sensibility defects in the face.

Volumetric computed tomographic (CT) scan showed a displaced bifocal fracture of the mandible and a not displaced, bilateral and multifragmentary orbitozygomatic fracture with bilateral maxillary hemosinus and a nondisplaced fracture of the orbital floors without herniation (►Fig. 2a–c).

After 5 days the patient underwent surgery for bifocal mandible fracture open reduction and internal fixation. The operation was performed under general anesthesia with nasal intubation in the right nasal cavity. The middle third fractures were not indicated for surgical treatment. The postoperative CT scan showed good mandibular fracture reduction and stability of the other facial fractures. The patient had a regular postoperative follow-up and physiologic healing.

After 3 months, the patient went to the emergency unit because of self-observation of left enophthalmos with hypo-

globus and deepening of the upper homolateral superior palpebral sulcus, occurred in the previous 24 hours. No vision defects, diplopia, ocular movement deficits, or facial sensibility modifications were found (►Fig. 3a, b).

The patient did not report recent facial trauma or rhinosinus infection. Ophthalmological-orthoptic evaluation excluded ocular pathology with normal Hess-Lancaster test, no diplopia, and visual acuity problems. A maxillofacial CT revealed a complete occlusion of the left sinus meatus, monolateral atelectasis of maxillary sinus, presence of endosinus inflammatory material, and complete resolution of the contralateral maxillary hemosinus.

All the walls of the left maxillary sinus (in particular the lateral, posterior, and the orbital floor) presented a centripetal collapse with widening of the pterygopalatine and left infratemporal fossa, accentuation of the upper concavity of the orbital floor, and consequently augmentation of orbital volume with retroposition of the left eyeball (►Fig. 4a–c).

First, the patient was treated with antibiotics and steroids, then with functional endoscopic sinus surgery (FESS). An uncinectomy and antrostomy in the middle meatus were performed to aerate the left maxillary sinus.

Two months after surgery, a progressive, partial, and spontaneous left enophthalmos correction was achieved and then the clinical result was stabilized. No functional deficits occurred after surgery and a CT after 6 months showed the left sinus was aerated and the sinus walls less deformed (►Fig. 5a–c).

Fig. 2 Axial (a), coronal (b), and sagittal (c) CT scans of the fractures on the middle third of the face.

Fig. 3 (a, b) Clinical condition of the patient 3 months after the trauma; the occurrence of left enophthalmos, hypoglobus and deepening of the left superior palpebral sulcus can be noted.

Fig. 4 Axial (a), coronal (b), and sagittal (c) CT scans at 3-month follow-up after the trauma; the left maxillary sinus appears clearly atelectatic and completely filled by inflammatory material.

Fig. 5 Axial (a), coronal (b), and sagittal (c) CT scans at 6 months after endoscopic treatment. Notice the pneumatization of the left maxillary sinus, whereas acquired bone deformities remain stable.

Eighteen months after the endoscopic treatment, the patient underwent morphological surgical correction by left orbital floor reconstruction with a titanium mesh, through a transconjunctival approach. After 30 months, the patient now has satisfactory and stable results (►Fig. 6a, b and ►Fig. 7a, b).

Discussion

The clinical and radiological presentation of our patient is similar to SSS, but it does not support all the diagnostic criteria (►Figs. 8a, b). The first case of enophthalmos

after atelectasis of the maxillary sinus was described by Montgomery in 1964.⁴ In 1994, Soparkar et al described 19 cases of spontaneous monolateral enophthalmos with radiologic homolateral maxillary sinus atelectasis and classified them as “silent sinus syndrome” (SSS).⁵

The etiopathogenetic mechanism seems to derive from a maxillary sinus meatus obstruction with consequent hypoventilation, atelectasis, mucous retention and air reabsorption by the sinus mucosa with development of subatmospheric intrasinus pressure and consecutive centripetal sinus wall collapse.^{6–8}

Fig. 6 (a, b) Clinical view after left orbital floor reconstruction. Notice the repositioning of the left globe.

Fig. 7 (a, b) CT scans at 30 months of follow-up after left orbital floor reconstruction with titanium mesh show that the correct orbital volume has been regained.

Fig. 8 Pre- (a: 17836.61 mm³) and post- (b: 10849.27 mm³) atelectasis left maxillary sinus volume modification (analyzed with iPlan 3.0 Brainlab® software).

In literature the criteria for an SSS diagnosis include the following⁸:

1. Absence of chronic or acute rhinosinusitis in the 6 months before the appearance of clinical signs (enophthalmos, hypoglobus, and deepening of left superior palpebral sulcus).
2. Remodeling and depression of the orbital floor in coronal CT scans.
3. Absence of facial trauma, surgical procedures, and enophthalmos by another cause.
4. Absence of paranasal sinuses and congenital malformations of the nasal cavities.

According to this classification, the case reported in this work is not a classic SSS because it likely has a posttraumatic origin. In recent literature, the possibility is suggested to include posttraumatic cases in the SSS⁹ in the absence of primary traumatic orbital volume modification.

In 2004 Gagnon et al published the case of a 50-year-old man with a small displaced fracture of the orbital floor and medial wall with herniation of a modest amount of orbital soft tissue. In the absence of symptomatology, the patient underwent conservative treatment and 2 months later he developed a clinical and radiologic maxillary sinus atelectasis, similar to an SSS. The author attributed this to hypoventilation of the sinus due to maxillary meatus obstruction by orbital content herniation.³

In 2005 Ross and Kersten described a similar case in a 27-year-old man with enophthalmos, hypoglobus, accentuation of the superior palpebral sulcus, and radiological finding of atelectasis of the maxillary sinus 6 months after facial trauma with a moderately displaced fracture of the orbital floor, which was acutely asymptomatic and not surgically treated.¹⁰

Other authors described the role of the herniation of orbital content in the SSS. In 2003, Rose and Lund reported the effects of the obstruction of maxillary ostium by herniated orbital fat after orbital decompression in 6 patients with dysthyroid orbitopathy.¹¹

In 2008 Montezuma et al published two cases of enophthalmos respectively at 4 and 27 years after a facial trauma. The first patient, a 27-year-old man, had previously suffered a direct eye trauma. The second one, a 34-year-old woman, had a primary nasal fracture reduction and a secondary rhinoplasty (10 years later). The authors did not publish the diagnostic images acquired following the trauma to demonstrate the primary effects on orbital volumes. The CT images of the second patient were acquired after the suspected atelectasis of the maxillary sinus occurred. These show a modest thickening of the Schneider membrane without obliteration of the sinus cavity, which was aerated but atelectatic.¹²

Pawar et al reported a case of a 32-year-old patient with maxillary sinus atelectasis who underwent surgical reconstruction for a displaced fracture of the right orbital floor and medial wall. The author published a single CT image acquired after the occurrence of enophthalmos. This image shows hypoplasia of the right maxillary sinus, its obliteration by

inflammatory material, the presence of reconstructive lamina on the displaced floor fracture, and the persistence of an untreated displaced fracture of the medial wall.¹³

In 2004, Hobbs et al reported a 27-year-old patient with a clinical and radiological presentation compatible with SSS attributed to a trauma caused by nasal intubation performed 1 year previously. The authors thus proposed a new etiopathogenetic hypothesis for acquired atelectasis of the maxillary sinus.¹⁴

The case described in this article is the first reported in literature with complete radiological documentation of orbital trauma without primary orbital volume modification or soft tissue herniation. The analogy of the clinical-radiological findings with cases of spontaneous occurrence has allowed us to hypothesize that the etiopathogenesis is an obstacle to the aeration of the affected maxillary sinus. The patient therefore underwent endoscopic uncinectomy and antrostomy on the left maxillary sinus as indicated for patients suffering from SSS.

In agreement with literature,^{15,16} the histological and microbiological analyses of sinus content showed a nonspecific chronic flogosis, which was negative for bacteria and fungi at culture test. Endoscopic treatment is the first phase in the corrective procedure recommended by most literature. The second phase consists in surgical correction of the orbital volume and the aesthetic results.^{12,17-24} Some authors described the possibility of performing a one-step surgical operation to reestablish the correct maxillary sinus aeration and the right orbital floor morphology.^{3,8-10,16,25-29}

For our patient, we decided to first ensure maxillary sinus ventilation and to defer the orbital floor correction in absence of any functional disorder. This reduced the risk of infective complications and allowed the maxillary sinus to spontaneously remodel after the etiological cause was removed by endoscopy.^{12,17-24}

The recommended follow up before the second surgical step varies between 2 and 12 month; in literature the complete resolution of clinical symptoms after first procedure is described.^{12,17-24}

The uniqueness of this case is the late occurrence of enophthalmos after a composed orbital fracture without soft tissue herniation, due to atelectasis and deformity of the maxillary sinus.

This etiopathogenetic hypothesis is supported by complete clinical and radiological documentation obtained before and after the condition established itself. None of the cases previously reported in literature was completely documented with a CT scan and clinical pictures.

The presence of rim fractures could have decreased the resistance of the orbital floor, which would contribute to accelerating the deforming effect of negative intrasinus pressure, probably caused by direct damage to the ostiomeatal complex. This case, although closely connected to traditional diagnostic criteria, cannot be strictly attributed to SSS.⁸ The absence of symptoms, the clinical-radiological analogy, and the common pathogenetic mechanism have provided the rationale for the endoscopic treatment and the later surgical correction of residual morpho-aesthetic defects.

Conclusion

The case described in this article and the review of the literature do not change the conservative view for asymptomatic patients with nondisplaced orbital fractures. However, physicians' attention on coexistent traumatic changes in the physiologic sinus drainage system can indicate a longer period of clinical follow-up to catch possible late-onset complications. By the way, this can also be the starting point to reconsider true diagnostic criteria for SSS, which traditionally excludes traumatic cases.

Conflict of Interest

None declared.

References

- Hamedani M, Pournaras JA, Goldblum D. Diagnosis and management of enophthalmos. *Surv Ophthalmol* 2007;52(5):457–473
- Whitehouse RW, Batterbury M, Jackson A, Noble JL. Prediction of enophthalmos by computed tomography after 'blow out' orbital fracture. *Br J Ophthalmol* 1994;78(8):618–620
- Gagnon MR, Yeatts RP, Williams Z, Matthews B. Delayed enophthalmos following a minimally displaced orbital floor fracture. *Ophthalm Plast Reconstr Surg* 2004;20(3):241–243
- Montgomery WW. Mucocoele of the maxillary sinus causing enophthalmos. *Eye Ear Nose Throat Mon* 1964;43:41–44
- Soparkar CNS, Patrinely JR, Cuaycong MJ, et al. The silent sinus syndrome. A cause of spontaneous enophthalmos. *Ophthalmology* 1994;101(4):772–778
- Hunt SM, Tami TA. Sinusitis-induced enophthalmos: the silent sinus syndrome. *Ear Nose Throat J* 2000;79(8):576, 579–581, 584
- Davidson JK, Soparkar CNS, Williams JB, Patrinely JR. Negative sinus pressure and normal predisease imaging in silent sinus syndrome. *Arch Ophthalmol* 1999;117(12):1653–1654
- Vander Meer JB, Harris G, Toohill RJ, Smith TL. The silent sinus syndrome: a case series and literature review. *Laryngoscope* 2001;111(6):975–978
- Cobb AR, Murthy R, Cousin GC, et al. Silent sinus syndrome. *Br J Oral Maxillofac Surg* 2012;50(6):e81–e85
- Ross JJ, Kersten RC. Late enophthalmos mimicking silent sinus syndrome secondary to orbital trauma. *J Craniofac Surg* 2005;16(5):840–843
- Rose GE, Lund VJ. Clinical features and treatment of late enophthalmos after orbital decompression. *Ophthalmology* 2003;110:819–826
- Montezuma SR, Gopal H, Savar A, Turalba A, Cestari DM, Torun N. Silent sinus syndrome presenting as enophthalmos long after orbital trauma. *J Neuroophthalmol* 2008;28(2):107–110
- Pawar SS, Hong S, Poetker DM. Delayed presentation of silent sinus syndrome after orbital trauma. *Am J Otolaryngol* 2010;31(1):61–63
- Hobbs CGL, Saunders MW, Potts MJ. "Imploding antrum" or silent sinus syndrome following naso-tracheal intubation. *Br J Ophthalmol* 2004;88(7):974–975
- Wilkins RB, Kulwin DR. Spontaneous enophthalmos associated with chronic maxillary sinusitis. *Ophthalmology* 1981;88(9):981–985
- Boyd JH, Yaffee K, Holds J. Maxillary sinus atelectasis with enophthalmos. *Ann Otol Rhinol Laryngol* 1998;107(1):34–39
- Thomas RD, Graham SM, Carter KD, Nerad JA. Management of the orbital floor in silent sinus syndrome. *Am J Rhinol* 2003;17(2):97–100
- Babin E, Bertrand M, Retout A, Gardea E, Choussy O. Silent sinus syndrome with spontaneous orbital floor reconstruction. *B-ENT* 2009;5(2):125–128
- Nkenke E, Alexiou C, Iro H, et al. Management of spontaneous enophthalmos due to silent sinus syndrome: a case report. *Int J Oral Maxillofac Surg* 2005;34(7):809–811
- Braganza A, Khooshabeh R. Ethmoidal involvement in "imploding" (silent) sinus syndrome. *Ophthalm Plast Reconstr Surg* 2005;21(4):305–307
- Babiński D, Skorek A, Stankiewicz C. Chronic maxillary atelectasis (silent sinus syndrome) [in Polish]. *Otolaryngol Pol* 2006;60(6):929–933
- Sciarretta V, Pasquini E, Tesei F, Modugno GC, Farneti G. Endoscopic sinus surgery for the treatment of maxillary sinus atelectasis and silent sinus syndrome. *J Otolaryngol* 2006;35(1):60–64
- Facon F, Eloy P, Brasseur P, Collet S, Bertrand B. The silent sinus syndrome. *Eur Arch Otorhinolaryngol* 2006;263(6):567–571
- Sivasubramaniam R, Sacks R, Thornton M. Silent sinus syndrome: dynamic changes in the position of the orbital floor after restoration of normal sinus pressure. *J Laryngol Otol* 2011;125(12):1239–1243
- Beasley NJ, Jones NS, Downes RN. Enophthalmos secondary to maxillary sinus disease: single-stage operative management. *J Laryngol Otol* 1995;109(9):868–870
- Blackwell KE, Goldberg RA, Calcaterra TC. Atelectasis of the maxillary sinus with enophthalmos and midface depression. *Ann Otol Rhinol Laryngol* 1993;102(6):429–432
- Ando A, Cruz AA. Management of enophthalmos and superior sulcus deformity induced by the silent sinus syndrome. *Aesthetic Plast Surg* 2005;29(2):74–77
- Dinis PB, Branco C, Subtil J. Endonasal endoscopic repair of the orbital floor defect in the silent sinus syndrome. *Otolaryngol Head Neck Surg* 2006;135(5):800–802
- Sesenna E, Oretti G, Anghinoni ML, Ferri A. Simultaneous management of the enophthalmos and sinus pathology in silent sinus syndrome: a report of three cases. *J Craniomaxillofac Surg* 2010;38(6):469–472