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Unusual ocular manifestations of silent sinus syndrome

Manifestações oculares incomuns na síndrome do seio silencioso

Fabricio Lopes da Fonseca[1] ,Luciana Mazoti[2] ,Mariza Polati[3]

ABSTRACT

Silent sinus syndrome is an acquired condition in which there is a gradual collapse of the orbital floor and inward retraction of the maxillary sinus (atelectasis of the maxillary sinus). This in turn may cause associated ocular occurrences of enophthalmos and hypotropia. This is a report of an 8 year-old boy with silent sinus syndrome and associated ocular motility disorders. The association between silent sinus syndrome and ocular motility disturbance has been recently described in the literature. However, this is an infrequent association, mainly in childhood.

Keywords: Paranasal sinus diseases; Maxillary sinus; Orbit; Strabismus; Case reports

RESUMO

A síndrome do seio silencioso é afecção adquirida em que há colapso gradual do assoalho orbital e do seio maxilar (atelectasia do seio maxilar), o que pode acarretar alterações orbitárias e oculares associadas, como enoftalmia e hipotropia. Relatamos o caso de um paciente de 8 anos de idade com síndrome do seio silencioso e distúrbios da motilidade ocular. A associação entre a síndrome do seio silencioso e alterações da motilidade ocular extrínseca tem sido descrita na literatura. No entanto, esta é uma associação pouco frequente, principalmente na infância.

Descritores: *Doenças dos seios paranasais; Seio maxilar; Órbita; Estrabismo; Relatos de casos*

[1]Ophthalmology and Otorhinolaryngology Department, Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo - USP – São Paulo (SP), Brazil.

[2]Ophthalmology and Otorhinolaryngology Department, Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo - USP – São Paulo (SP), Brazil.

[3]Head of Strabismus Department. Ophthalmology and Otorhinolaryngology Department, Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo - USP – São Paulo (SP), Brazil.

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Introduction

The silent sinus syndrome (SSS) is a rare disorder, first described by Montgomery[1] in 1964 and named by Soparkar et al.[2] in 1994, characterized more often by enophthalmos and hypotropia and increased ipsilateral orbital volume secondary to maxillary sinus atelectasis.

The typical syndrome occurs between the third and fifth decades of life[3], with equal distribution between genders[3,4] and unilateral symptoms, most commonly reported as enophthalmos of the ipsilateral eye, proptosis of the contralateral eye or changing of facial appearance[1,5-7]. Orbital asymmetry, palpebral retraction[1,6,7] and deepening of the superior eyelid sulcus[5] are more specific symptoms. The ocular motility is frequently preserved[8-10], although there are recent articles relating diplopia[11,12], even in children[13].

The most obvious radiological signs are clouding of the maxillary sinus[10], loss of the maxillary sinus roof convexity and uprising and thinning of the orbital floor[8,10], nasal septum deviation at the affected side[2,10] and lateralization of the ipsilateral middle turbinate[5,10]. Increased orbital volume and atelectasis of the maxillary sinus are required findings for the diagnosis[8]. The classical treatment aims to restore normal sinus aeration and to correct enophthalmos and hypotropia with an orbital implant[4]. The disease progression is interrupted with sinus aeration[10].

Report of case

The case of an 8 year-old boy with exotropia and amblyopia of the left eye since birth. Treatment was patching for 2 years, which was discontinued 3 years ago, when he was started on glasses. There is also no history of trauma or surgery.

The ophthalmological exam revealed a visual acuity OD 1,0 and OS 0,2 (LH Symbols; Snellen equivalent OD 20/20 and OS 20/100), under cycloplegic refraction of +1.25 -1.50 x 180 and +3.00 -2.00 x 160, respectively; far and near left exotropia and torticollis (tilts and rotates the head to the left) with an angle of turn of 15 degrees. There was a Hertel enophthalmos of 4 mm.

The Krimsky test showed an incomitance: at distance, right eye fixating exotropia of 35 prism diopters and left hypotropia of 4 prism diopters; with left eye fixating, an exotropia of 50 prism diopters and left hypotropia of 6 prism diopters. At near distance, right eye fixating exotropia of 30 prism diopters and left hypotropia of 6 prism diopters; with left eye fixating, an exotropia of 50 prism diopters and left hypotropia of 6 prism diopters.

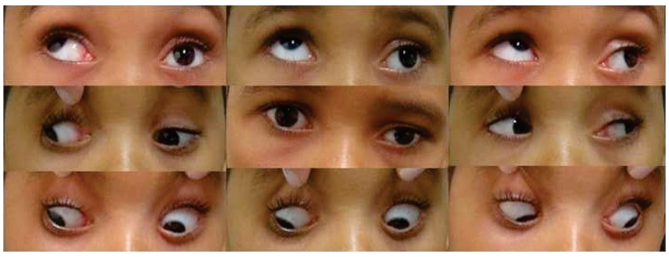

Figure 1. Facial asymmetry, enophthalmos and hypotropia on the left; gaze restriction.

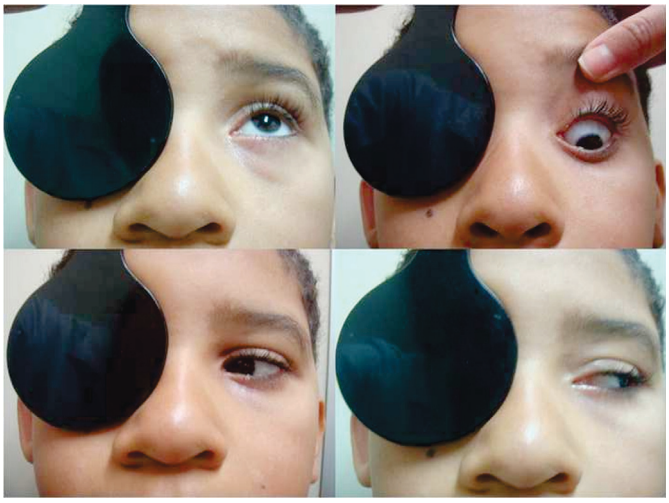

Figure 2. Ductions, showing no adduction deficit

All the left extraocular muscles had a marked underaction, except for the lateral rectus muscle. It seems an adduction deficit (Figure 1), that is not present in ductions (Figure 2). These findings would be secondary to a large exotropia. The upper eyelid did not follow the left eye when it moved in a downward position. There was also a manifest constant pendular nystagmus, low

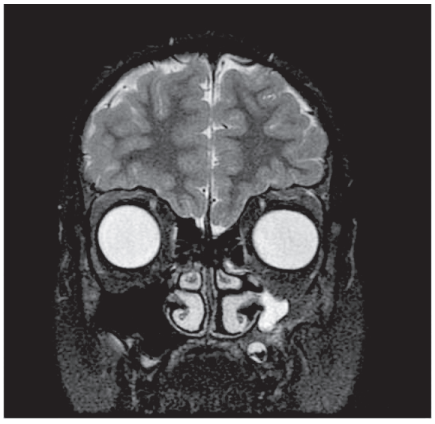

Figure 3. T2 image showing the maxillary sinus of reduced size, with elevation of its floor and lowering the orbit floor. It is also observed the ipsilateral middle turbinate laterally displaced

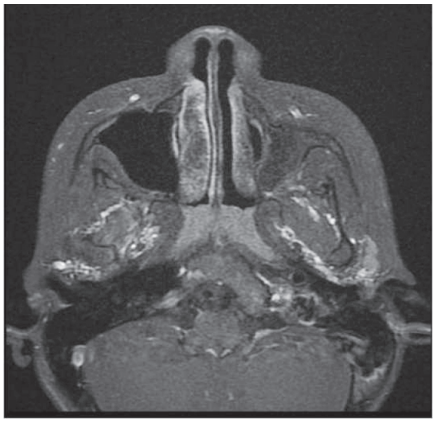

Figure 4. Increased ipsilateral middle meatus due to the lateralization of the uncinate process in T1 image

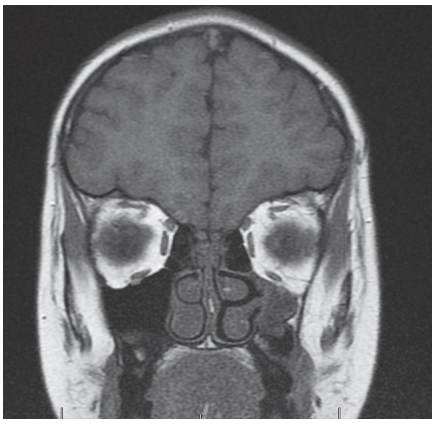

Figure 5. T1 image showing displacement of left inferior rectus muscle

amplitude and frequency, no null point, observed in all gaze positions, with upper eyelid fasciculation.

In an attempt to open the eyes, they converged.

There was anisocoria of 2 mm, left pupil greater. This condition was absent in the dark. Unfortunately, we do not have measurements in both bright and dim illumination. The left eye was amblyopic. A orbital MRI scan showed an anomalous left maxillary sinus featuring small size, associated with elevation of its floor and lowering of orbital floor, and hyperintense T2 filling content. A slight deviation was also noted of the nasal septum to the affected side, and lateralization of the uncinate process and left middle turbinate (Figures 3 and 4). A coronal section of the MRI showed inferior displacement of extra-ocular muscles positioning, mainly the inferior rectus (Figure 5).

The patient was treated with glasses and referred to Otorrhinolaringology Department for endoscopic maxillary antrostomy. After 12 months of former evaluation, visual acuity and ocular motility were unchanged.

Discussion

The exact pathogenetic mechanism leading to SSS is not clear[14]. Initially, the hypothesis that chronic obstructive sinusitis coexisted with a primary condition of sinus hypoplasia was formulated; however, several authors demonstrated the absolute normality of the maxillary sinus walls before clinical and radiological findings of SSS[14,15] which confirms that it is an acquired condition. Studies identified infundibular obstruction as a probable pathogenetic event, leading to sinus hypoventilation and development of negative pressure. Persistence of negative pressure seems to produce slow maxillary sinus atelectasis. Furthermore, progressive osteomalacia[14] or probably reduced osteoblastic activity[15] is thought to contribute to inward bowing of the sinus bone and to orbital floor osteomalacia.

This case presents some unfrequent features associated with silent sinus syndrome. The occurrence of symptoms in this age group is unusual. Although diplopia and gaze restriction are seen in some cases, its presentation is unusual, mainly in this age range. The motility issues is due to a mechanical restriction, as showed by MRI scan and related in previous studies[11-13]. The most common findings suggest, with hypotropia and enophthalmos, as well as the characteristic radiological criteria, the diagnosis of framework compatible with silent sinus syndrome, however, with unusual epidemiological and clinical characteristics. The treatment of SSS typically consists in functional endoscopic surgery to remove the obstruction and restore positive pressure. Orbital floor lifting and reconstruction may be performed during the same session or at a later date[16]. Surgical intervention is useful to interrupt progression of maxillary and orbital changes and to correct enophthalmos or facial deformity, although it does not seem to produce a significant restoration of orbital muscle function and, consequently, of diplopia[15,17].

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Corresponding author:
A/C Fabricio Lopes da Fonseca
Rua Xavier de Almeida, 1135 apto 121 – Ipiranga
CEP 04211001 - São Paulo (SP), Brasil.
E-mail: fabricio89@gmail.com