THE SILENT SINUS SYNDROME: A MISDIAGNOSED PTERYGO-MAXILLARY FOSSA TUMOUR - CASE REPORT

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Abstract: *This entity is relatively new in sino-nasal pathology, characterized by a chronic unilateral maxillary sinusitis, without specific symptoms. Like atelectatic otitis caused by the middle ear negative pressure dysfunction of the Eustachion tube, in silent sinus syndrome, a slight thickening of the sinus walls appears, caused by chronic inflammation and negative intrasinusal pressure due to the osteo-meatal dysfunction, with the onset of indolent enophthalmos. Materials and methods: We present a 38-year-old man misdiagnosed with right pterygomaxillar fossa tumor, suffering from fluctuant migrena, partial nasal obstruction onset 3 years earlier, associated in the last 3 months with unilateral indolent enophtalmia, unilateral vision problem and facial asymmetry. Clinic and paraclinic examinations showed a likely silent sinus syndrome. In this case we performed sino-nasal surgical treatment. Conclusion: SSS is a rare pathology, characterised by lack of pain and progressive enophtalmos, being necessary a multidiscipline outcome.*

INTRODUCTION

The silent sinus syndrome (SSS) is a relatively new entity, first described in 1964 as the chronic maxillary atelectasis by Montgomery (1), but named as such by Soparkar in 1994.(2) This disease is characterized by painless enophtalmos, sometimes the onset of diplopia, with facial deformity and signs of chronic maxillary sinusitis on paraclinic examination. These symptoms are caused by the unaerated unilateral maxillary sinus due to trauma, anatomic development, post-surgery or other unknown factors. Brandt et al (3) found 105 cases from 1964-2006, some of them under the name of chronic maxillary atelectasis (CMA), and proposed that the name of silent sinus syndrome is a stage of the CMA.

We found no reports regarding misdiagnosed silent sinus syndrome (SSS) with pterygo-maxillary fossa tumour, so be believed it would be interesting to present the case in order to clarify the entity and specify the symptomatology and treatment.

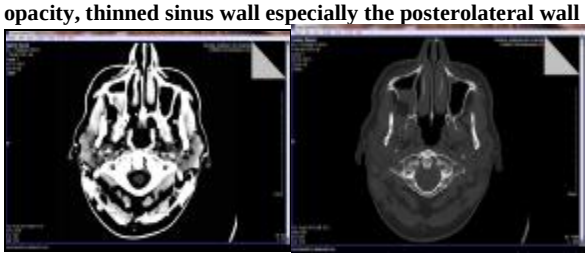
CASE REPORT

A 38-year old man was sent to our clinic for suspicion of right pterygomaxillary fossa tumour. He complained of headache for about 3 years, treated with anti-inflammatory once in a while and right painless progressive enophtalmos for the last 3 months with inconstant vision disturbances. The patient did not complain of mucopurulent rhinorhea, teeth pain, diplopia or hemi facial pressure. He remembered a minor facial trauma 20 years ago during a football game, which did not require any treatment. He has worked for 15 years in a factory with exposure to pollutants. He is a non-smoker, with a history of general rush after antibiotic administration. The clinic examination revealed right painless enophtalmia, with upper lid downward, deepened upper lid sulcus and facial asymmetry. The patient was seen by an ophthalmologist, who noticed no vision disturbances or diplopia. He also consulted a neurologist for his headache, who decided to perform an MRI and discovered a right pterygomallar fossa tumour sending him to an OMF surgeon. The latter decided to make a CT scan which revealed right maxillary sinus volume loss, with opacity, thinned sinus wall, especially the posterolateral wall and the superior wall. The nasal endoscopy results were indicative of chronic hypertrophic nasal mucosa, lateralization of the middle turbinate and obstruction of the air space reduced on the right side.

Figure no. 1. Frontal view of the patient shows enophtalmos, facial asymmetry and deep upper lid sulcus on right side

The CT scan revealed right maxillary sinus volume loss, with opacity, thinned sinus wall especially the posterolateral wall and the superior wall, increased volume of the orbit due to retraction of the sinusal walls and increased fat tissue in the pterygomaxillar fossa.

Figure no. 2. Axial CT scan shows sinus volume loss, with opacity, thinned sinus wall especially the posterolateral wall



We diagnosed the patient with silent sinus syndrome on the right side and excluded the tumour suspicion based on the

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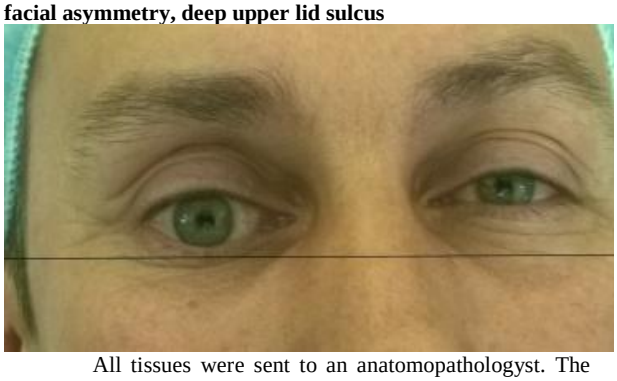
characteristics on the CT (the same density as fat on the IRM localised in the pterygomaxillar fossa).

The patient was processed for surgery and we conducted a functional endoscopy sinus surgery with medialization of the middle turbinate, uncinectomy, large antrostomy extended to anterior part of the internal sinus wall, lavage of the sinus cavity (dark-yellow viscous mucus, with black centre). Using a 45-degree endoscope, we visualised the interior cavity of the sinus - smaller than on the opposite side, with central retraction of the posterior and superior wall and no changes to the mucosa.

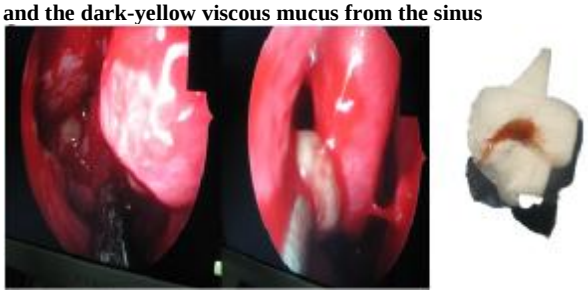
Figure no. 3. Frontal view of the patient shows enophtalmos, facial asymmetry, deep upper lid sulcus



All tissues were sent to an anatomopathologyst. The microscopic alterations were chronic inflammatory on the mucosa of sinonasal mucosa. There was no bacteria culture present. Regarding the enophtalmos, we performed no surgery, but waiting for the results of the 6 month - recovery.

The 3-month follow-up revealed significant changes: reduced asymmetry of the face, no visual complaints and reduced enophtalmia. The rhinoscopy showed large osteomeatal complex formed after surgery with normal discharged maxillary cavity.

Figure no. 4. Intraoperative images from the middle meatus and the dark-yellow viscous mucus from the sinus



DISCUSSIONS

Misdiagnosing a silent sinus syndrome changes the course of treatment. A pterygopalatine fossa tumour comes with late presentation at the physician because of absence of symptoms and usually, imaging showing extension in the area around, cases when surgery is impossible to perform.(4) Differential diagnosis is made with other cause of indolent enophtalmia, appearance of hipoplastic sinus (in syndromic pacients), sdr. Parry Romberg, linear scleroma, trauma, tumours, diffuse facial lipodistrophy.(5)

In the literature, there is described the confusion of painless enophtalmos and facial asymmetry with Bell's palsy. The patient underwent specific treatment with no response. After CT examination the maxillary sinus was hypoplastic, with centripet modification of the superior and posterior wall, enophtalmos, being diagnosed with silent sinus syndrome.(6)

The medical history plays an important role in the appearance of SSS; it is known that facial trauma, surgery for sinonasal malignancies, history of chronic rhinosinusitis and orbital decompression produce changes in sinonasal anatomy predisposing to develop SSS.(7)

There is no gender predilection, the typical patient is in 3-rd to 4-th decade, non-smoker, apparently with no history of trauma or chronic sinuses discharge.

Hobbs et al described a 27-year old female who developed this condition after nasotracheal intubation for a routine general anaesthesia. Also, iatrogenic version of the disease occurring after bilateral orbital decompression is described. Usually, it is unilateral, but bilateral iatrogenic SSS was described (7) as well.

Usually, an anatomical condition is necessary to appear in this disease like septal deviation, big Haller cell, concha bullosa, medialisation of middle turbinate (which moves the uncinate process blocking the ostium, even in the presence of an accesory ostium), presence of mucocel, nasal polyps. Some of them are visible on CT scan or MRI, which are the main investigations necessary to confirm the disease.(8)

The pathogenesis of SSS is still unknown, but some theories argue that the hypoventilation of the sinus, due to the anatomical middle meatus modification, causes sinus atelectasis. Other specialists maintain the idea that chronic sinusitis with anaerobe bacteria causes viscous mucus accumulation, osteoclastic modification to the sinus walls and onset sinus atelectasis.(9)

The surgical treatment is the best option for this kind of patients: large antrostomy with drainage of the mucous. The dilemma is if we have to repair the inferior orbital wall or we should wait and see (but how long should we wait?). Sesenna et al presented three cases treated with a single stage procedure, endoscopy and subcilliar approach, obtaining very good results, with no relapse of the disease.(8) Numa et al presented a SSS case who underwent surgical treatment, and 14-th month follow-up revealed normal eye line.(10) Some authors had good results in resolving the aeration of the sinus and the follow-up for resolution of the enophtalmos from 2 to 2 year.(11) We chose to wait for 6 months for the resolution of the enophtalmos, but at the 3 month-follow-up, there was a very good improvement.

Children are not predisposed to develop SSS, but nonsyndromic pediatric patients can develop SSS due to maxillary sinus hypoplasia, even if the sinus is not fully developed until 15-18 years of age. It is difficult to diagnose SSS in a maxillary sinus hypoplasia without any previous imaging studies.(12)

Figure no. 1. Mangussi classification of chronic maxillary atelectasis

Chronic Maxillary Atelectasis				
Terminology	Stage I	Stage II	Stage III	SSS
Deformity	Membranous (soft medial wall)	Bony (osseous walls)	Clinical (enophthalmos, hypoglobus, facial asymmetry)	
Nasosinusal symptoms	+++	++	+	–

Mangussi et al proposed a classification of Chronic Maxillary Atelectasis and SSS, considering that the latter is a stage of first. Also Hypoplastic maxillary sinus should be differentiate from SSS.(13,14)

CONCLUSIONS

Differential diagnosis between silent sinus syndrome and

pterygopalatine fossa is very important, because the course of treatment is different. The scan imaging and clinical complaints showed are very useful, aiding the clinician to propose the specific treatment. Being a new entity, SSS should be taken into consideration when patients come with progressive enophtalmos, subclinical signs of rinosinusitis and specific scan imaging.

The endoscopic treatment associated with the subcilliar approach for enophtalmos (15) should be individualised for every patient, depending on his wishes.

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