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Chronic Maxillary Atelectasis and Silent Sinus Syndrom: Report of Three Cases and Literature Review

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1. Abstract

Chronic Maxillary Atelectasis (CMA) and Silent Sinus Syndrome (SSS) are conditions that may lie on the same clinical spectrum, since both have similarities in pathophysiology, clinical findings and treatment, although the absence of sinonasal symptoms is the main difference between them. In this study we present three different cases of CMA that were submitted to functional endoscopic sinus surgery to exemplify this condition and we made a literature review about both CMA and SSS for the most relevant and recent data about the subject. Most studies show that a negative pressure gradient may be the cause for the maxillary antral collapse and that best treatment is the endoscopic approach to reopen the sinus and restore its ventilation, orbital floor reconstruction is still debatable though. In conclusion, the understanding about both CMA and SSS seem to be increasing in the last years and that allows a better treatment and classification of the these conditions.

2. Introduction

Chronic maxillary atelectasis (CMA) and silente sinus syndrom (SSS) are two unusal clinical conditions whose denominations were coined at the end of the 20th century. Event though their names and even symptoms are different, recent studies have shown more and more that both situations seem to be in the same clinical spectrum [1, 2].

The main difference consists basically in the presence or abscence of sinonasal symptoms, which must be absent in the classic SSS [3]. Thus, isolated cases of SSS are tipically reported in the ophtalmological literature, while CMA, whose patients have similar complaints to chronic rhinosinusitis, is traditionally cited in otorhinolaryngological journals [1].

To demonstrate that situation, we reported three diferente cases of patients with CMA who were treated at an otolaryngology service and we made a literature review on both CMA and SSS to better caracterize these conditions and to show their proximity.

3. Case Report

3.1. Case 1

A 68-year-old female patient, hypertensive and dyslipidemic, referred hyposmia and hypogeusia that started two years before. She also mentioned itchy nose and sneezing when in contact with dust or smoke, but she denied rhinorrea, nasal obstruction, headache or facial pain.

Treatment with topical corticosteroids and nasal irrigation with saline was given. There was no improvement after three months and then a fibronasolaringoscopy and a computed tomography (CT) of the sinuses were requested. In the endoscopy, there was only signs of nonspecific rhinitis and pharyngolaryngeal reflux. While CT showed opacification and volume reduction of the right maxillary sinus, as well as a content with soft tissue density in sphenoid sinuses bilaterally with bone sclerosis and erosion of its walls (Figure 1).

The hypothesis of sphenoid fungal rhinosinusitis was proposed and the possibility of chronic maxillary atelectasis stage II was considered, since the patient had sinonasal symptoms, but she did not have facial asymmetry or orbital changes. Surgical treatment was then indicated for both conditions.

The patient underwent functional endoscopic sinus surgery (FESS). Anthrostomy of the right maxillary sinus was performed with a backbiter and a reduced size sinus was observed, with a "glue" type secretion inside and lowering of the orbital floor. Anthrostomy of the sphenoid sinuses was also performed, in which a friable and hypertrophied mucosa and a thick fungal content bilaterally were seen.

The following day, the patient was asymptomatic and was discharged home with a prescription for antibiotics and saline irrigation. She returned in a week in good general condition and she had excellent evolution of hyposmia and hypogeusia over the course of a year, despite occasionally having itchy nose and sneezing, probably due to allergic rhinitis. The anatomopathological result was compatible with non-invasive fungal rhinosinusitis, suggesting fungus ball, but there was no fungal growth in culture to identify its species.

3.2. Case 2

A 55-year-old male patient, previously healthy, reported a 15-year history of frontal headache, a feeling of pain and pressure in the face, postnasal drip and bilateral congestion, which worsened seasonally. On physical examination, he had septal deviation to the left, hypertrophy of the inferior turbinates and a slightly pale nasal mucosa. He had a CT scan of the sinus from three years ago showing opacification of the left maxillary sinus and its reduced dimensions, in addition to obliteration of the ostiomeatal complex by ipsilateral septal deviation (Figure 2).

After treatment with antibiotics and both systemic and topical corticosteroids for a few weeks, he repeated the CT scan, which maintained the deviation and maxillary sinusopathy with decreased sinus volume and now with lower displacement of the orbital floor. The patient denied visual symptoms or facial asymmetry, suggesting a diagnosis of stage II CMA.

He was then submitted to FESS, in which sectorial septoplasty was performed simultaneously with removal of the deviated septal cartilage to the left, bilateral turbinectomy and anthrostomy of the left maxillary sinus with backbiter. In the intraoperative period, a reduced size maxilallary sinus with “glue” secretion was seen and no natural or accessory ostium was identified.

The patient had an episode of anterior epistaxis on the same day requiring anterior nasal packing for one day, being discharged on the second postoperative day with a prescription for oxymetazoline for three days and antibiotics.

He returned after two weeks, asymptomatic and without further bleeding, but with a large synechia of the septum with the lower shell on the left. Due to the extension and the good condition of the patient, the synechia was only undone three months later, when he first complained of nasal congestion after the surgery.

Over one year, he had a with a very significant improvement in his headache and good control of nasal symptoms. A postoperative CT showed an open and well-ventilated maxillary sinus, with excellent aspect.

3.3. Case 3

A 45-year-old female patient with a previous history of adenotonsillectomy and without comorbidities, referred having facial pain in the frontal and right retro-ocular areas for six years. She also complained of a long-standing ipsilateral nasal obstruction, hyaline rhinorrhea, frequent sneezing and itchy nose. There was a slight enophthalmos in the right eye, but the patient herself had not noticed it (Figure 3). In fibronasolaringoscopy, there was a slight septal deviation in “S” shape, affecting the right side anteriorly. Also, she presented lateralization of the medial wall of the right maxillary sinus.

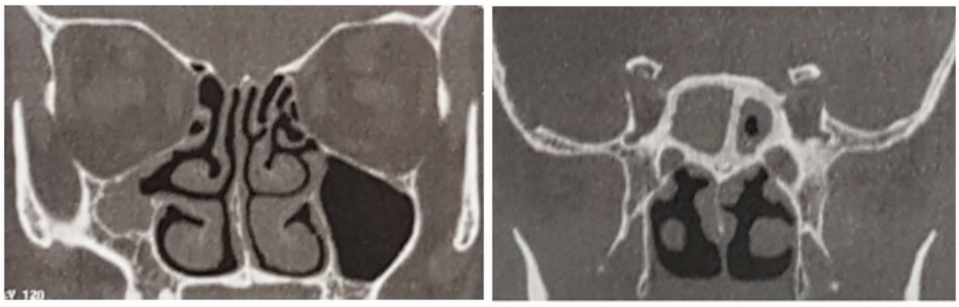

Figure 1: Coronal section CT of the face showing opacity and reduced volume of the right maxillary sinus, as well as filling the sphenoid sinuses bilaterally with surrounding bone erosion.

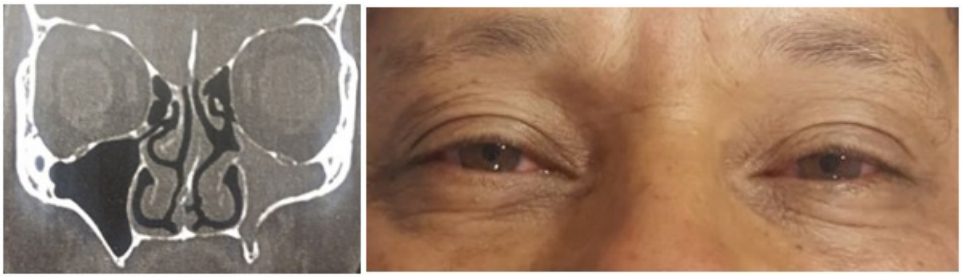

Figure 2: Coronal section CT of the face showing septal deviation to the left and opacity of the ispilateral maxillary sinus with atelectasis of its walls and increased orbital volume. Ectoscopy did not show enophthalmia or evident facial asymmetry.

Figure 3: Slight enophthalmos in the right eye.

In this case, CT of the sinuses was also requested. The scan showed lateralization of the uncinate process on the right and paradoxical medium turbinates (right one was bifid), but also atelectasis of the walls of the right maxillary sinus, resulting in a smaller sinus, filled by secretion, and lowering of the orbital floor, with enophthalmos and an increase right orbital volume (Figure 4). Based on this exam and the patients symptomps, the hypothesis of stage III CMA was proposed.

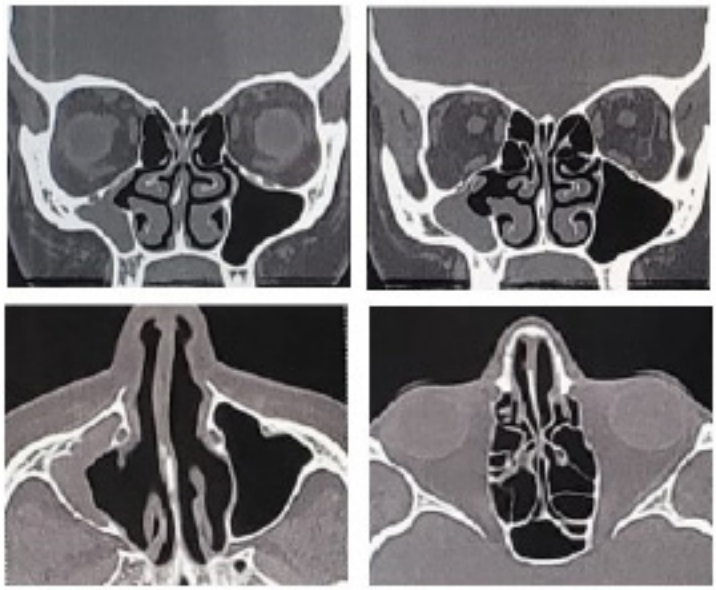

Figure 4: CT coronal sections show opacity and reduction of the right maxillary sinus with paradoxical middle turbinates. In axial sections, it is possible to notice lateral retraction of the medial sinus wall and slight enophthalmia in the right eye.

The patient was then submitted to FESS, in which an anthrostomy of the right maxillary sinus was performed with a backbiter, with no complications, and she was discharged with nasal corticosteroid and and instructed to perform nasal saline irrigation. After one week, she stated partial improvement in her pain symptoms and mild nasal congestion on the right side, being completely asymptomatic after thirty days of surgery.

4. Discussion

In 1964, Montgomery described two cases of unilateral enophthalmos in patients with supposed maxillary "mucoceles" [4]. Thirty years later, Soparkar et al. created the term "silent sinus syndrome" to refer to nineteen patients with spontaneous enophthalmos and collapse of the maxillary sinus, but with no sinus-related symptoms [3]. Shortly thereafter, in 1997, Kass et. al. also described the process of spontaneous maxillary sinus implosion associated with sinonasal symptoms, using the term "chronic maxillary atelectasis" [5].

Thus, SSS can be defined by the criteria devised by Soparkar as a change in facial appearance, enophthalmia and/or hypoglobus associated with reduction of maxillary sinus volume in the absence of sinonasal symptoms [6]. CMA is also described as the decrease in the sinus volume secondary to displacement of its walls and associated with symptoms such as congestion, rhinorrhea, headache, facial pressure, postnasal

discharge, and/or dental or facial pain. It can also be divided into: stage I, when there is only membranous deformity; stage II, if bone deformity; and stage III, when there is facial deformity and asymmetry [5]. Considering this, we can classify our patients here described as having CMA (cases 1 and 2 as stage II and case 3 as stage III), since two of them had significant complaints of facial pain and one of hyposmia.

Therefore, it is clear that sinonasal symptoms have a fundamental role in this differentiation, which even justifies the "silent" course of SSS [7]. However, it is observed that at least one-half of the cases described as CMA or SSS have a similar history of mild rhinosinusitis, showing a lack of padronization in the literature [1].

Brandt and Wright, in 2008, made a systematic review on all cases published so far and they saw that the distinction between the two diseases appeared to be a nomenclature problem, as they realized that 85% of SSS cases also met the criteria for CMA. They conclude that both diseases are likely a spectrum of the same clinical condition and that SSS may be the final pathway of CMA in the absence of sinonasal symptoms, which is perfectly possible to occur, as there is a poor relationship between symptoms and late-stage maxillary antral collapse [1].

CMA pathophysiology is not yet fully understood. Initially, there was the hypothesis that it was linked with hypoplasia of the maxillary [8]. This theory proved to be inconsistent after several studies showed normal sinus anatomy many years before the onset of symptoms, which favors that CMA is an acquired and progressive condition [9].

There are currently at least two theories for the pathogenesis of CMA: the obstruction of outflow theory, that posits that the causal fact is an acquired obstruction of the infundibulum generating hypoventilation; and mechanical theory, in which the contraction and relaxation of the masticatory muscles might cause aspiration of a closed maxillary antrum [10]. The obstruction of outflow theory has found some good support in the literature, since CMA may be associated with nasal anatomical variation, such as septal deviation, concha bullosa, or even other obstructive conditions (occlusion of the ostium by fat after orbital decompression, trauma, tumor or foreign body) [2]. In the three cases presented, one of them was submitted to septoplasty due to significant deviation and another had paradoxical medium turbinate, which may be associated factors for obstruction of outflow.

Both explanations seem to be moving towards a common denominator, which is the negative pressure gradient. In these situations of chronic poor ventilation, secretion is reabsorbed and negative pressure is generated within the sinus [10]. Simultaneously, osteolysis of its walls occurs through bone resorption (osteopenia due to increased osteoclastic activity) in response to the lowgrade inflammatory process, making them thinner and more fragile [11]. These two conditions favor the implosion of the maxillary sinus, with retraction of its walls and the arching of the orbital floor [8]. In this stage, CMA can already be classified into stage II, as in cases 1 and 2, or stage III if facial asymmetry, as in case 3.

The negative pressure theory has already been demonstrated in animals with occluded maxillary ostium and in humans [2, 12]. In the study by Kass et al., negative antral pressure was detected in the affected sinus of patients undergoing surgery, while the contralateral antral pressure was equal to atmospheric pressure [13]. In addition, Gillman et al. linked a parallel between this phenomena to auditory tube dysfunction, generating tympanic membrane retraction also through hypoventilation [14].

The clinical findings in SSS covers only ophthalmological signs and symptoms, the main ones being painless enophthalmos (98%), hypoglobus (53%) and mediofacial asymmetry or deformity (69%). Diplopia may occur (up to 65%), but eye acuity and motility are almost always preserved [1]. In our study, only patient 3 had mild enophthalmos. Another possible finding is retraction of the upper eyelid and Graefe's pseudo-sign, which is the illusion of the lack of convergence of the upper eyelids when looking down due to unilteral hypoglobus [9]. Stage III CMA may also have these manifestations, so as sinonasal symptoms.

Imaging exams are essential for diagnosis, since they prove the acquired reduction in the volume of the maxillary sinus, as seen in the three cases. Computed tomography (CT) of sinuses remains the gold standard for better visualization of bone structures and also for documenting enophthalmos and increased orbital volume [10]. Other key findings include total or partial sinus opacification, loss of the maxillary roof convexity, uprising and thinning of the orbital floor, septal deviation on the affected side and lateralization of the ipsilateral middle turbinate (obstructive factors) [8].

In all cases presented, CT was essential for diagnosis, showing

unilateral maxillary sinus disease with secretion and reduction in sinus volume. In case 2, the bowing of the orbital floor was more evident, increasing the orbital volume. Case 1 also had bilateral sphenoid fungal rhinosinusitis, possibly related to the patient's nasal symptoms.

The treatment of these cases is basically through surgery, consisting of reopening the maxillary sinus and removing the obstructive factor to restore ventilation and intrasinusal pressure [8]. Historically, the Caldwell-Luc procedure for maxillary sinus had been used in the past, but today, with advances in functional endoscopic sinus surgery (FESS), anthrostomy with uncinectomy or total ethmoidectomy has been stated as the best treatment option [2, 7]. If septal deviation, septoplasty can be performed in the same act, as demonstrated by Darghal et al. in four of the eleven patients presented [11], and as was done in our patient in Case 2. Intraoperatively, much attention is needed when there is a lateralisation of the uncinate process when there is hypoglobus, as its incorrect identification can lead to eye damage [7]. Fortunately, none of the reported patients had ocular complications, only one of them had mild epistaxis and synechia, that were resolved. Some authors have also demonstrated the use of balloon sinuplasty with better cost-benefit [15, 16].

Another reason for debate is the need and the moment to perform surgical correction of the orbital floor, which is done via transconjunctival or subciliary for insertion of autogenous or alloplastic subperiosteal implant, such as Medpore implants (high density porous polyethylene material), silicone blocks or Teflon sheets. [1, 10, 11]. This procedure can be done simultaneously with FESS or 6 months later. Doing both in the same hospitalization has many advantages over the risk of another general anesthesia and the costs, but many authors argue that doing so increases the risk of orbital infection. In addition, the evolution of CMA is interrupted with FESS and there is some degree of sinus expansion and even resolution of the disease in some cases [2, 10]. As an example, Babar-Graig et al. only had to do such intervention in two out of sixteen patients (12.5%)and in the study by Dorlodót et al. only one out of eighteen (5.5%) [2, 17]. In a 2008 study, 34% of surgeons did not reconstruct the orbital floor in a simultaneous treatment [1]. As only patient 3 had enophthalmia, but it was still mild, none of our cases underwent such procedure.

The distinction between CMA and SSS is tenuous and involves much more a nomenclature question and classification, since there is a lot of similarity in pathophysiology, imaging findings, evolution and treatment. There are still controversies regarding the classification, some aspects of the surgical technique and even the pathophysiology, but as more studies are published, the more these diseases seem to move towards a common denominator.

5. Conclusion

CMA and SSS are two pathologies that have several similarities, and may even be contained in the same clinical spectrum of disase according to the last studies. Even so, it is necessary to know the definition of the two conditions so that there is a more accurate classification in the future, allowing a better treatment and follow-up.

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