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Direttore Nicola Picardi

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Nicola Lombardo*, Pierpaolo Ferrise*, Giovanna Lucia Piazzetta*, Donatella Bruzzichessi**, Domenico Testa***, Pasquale Viola*, Corrado Pelaia****, Gaetano Motta***

**Department of Medical and Surgical Sciences, ENT Unit, University "Magna Græcia" of Catanzaro, Catanzaro, Italy*
***Department of Clinical and Experimental Medicine, University "Magna Græcia" of Catanzaro, Catanzaro, Italy*
****Department of Anesthesiologic, Surgical and Emergency Sciences, Otolaryngology, Head and Neck Surgery Unit, Second University of Naples, Naples, Italy*
*****Department of Medical and Surgical Sciences, Section of Respiratory Diseases, University "Magna Græcia" of Catanzaro, Catanzaro, Italy*

Maxillary sinus mucocele with orbital complications

INTRODUCTION: *Maxillary sinus mucocele is an unusual clinic entity (incidence of 3-10%), represented by mucous secretion lined by respiratory stratified pavemented epithelium and its origin is mainly secondary to infections, inflammations, surgery, trauma or neoplasia.*
CASE REPORT: *We present a case of maxillary sinus mucocele. Subject of the study is a 74 aged patient referring positive anamnesis for nasal cavity surgery. His symptoms appeared six months ago and he complainted vertical ocular diplopia and ocular tension sensation, without rhinologic complaints. Oculistic examination confirmed an isolated left inferior rectus palsy, and CT showed mucous secretion occupying left maxillary sinus, with bone erosion to the orbital floor until orbital muscles. MRI is the gold standard for differential diagnosis with neoplastic lesions but the patient refused to perform this radio-diagnostic exam. He underwent to Endoscopic sinus surgery, consisting in middle meatal antrostomy and removal of the mucocele. Four months later the endoscopic follow-up showed regular maxillary cavity and regular ocular motility.*
CONCLUSION: *In our opinion the rebuilding of the eroded bone is not mandatory if the integrity of the maxillary upper wall mucosa is respected, and antrostomy with drainage allows to recover ocular and paranasal sinus function.*

KEY WORDS:, Diplopia, Endoscopic sinus surgery

Introduction

Maxillary sinus mucocele is an unusual clinic entity, represented by mucous secretion piled up in the maxillary sinus, lined by respiratory stratified pavemented epithelium. Mucoceles may be caused by an obstructed mucous gland's duct in the maxillary sinus wall, or secondary to chronic infection, allergic sinonasal disease, surgery on paranasal sinuses, maxillo-facial injury, sinonasal cavity's neoplasia which obstruct the natural sinus' ostium[1]. Generally they have a slow spread in many directions eroding bone walls. Asymptomatic during many years, it manifests itself with an obstructive nasal symptomatology, heaviness, cheek's soft tissues swelling or, rarely, with ocular symptoms.

Case Presentation

Subject of the study is a 74 aged patient referring positive anamnesis for nasal cavity surgery, not better specified, when he was 20 years old, to remove a sinonasal polypus and doubtful anamnesis for asthma. He has got good health up to six months earlier, when he began to complaint new symptoms. His first complaints were vertical binocular diplopia, left eye's proptosis, ocular tension sensation and infraorbital swelling, without pain or rhinologic complaints.

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Correspondence to: Dr. Nicola Lombardo ENT Unit Director, Department of Medical and Surgical Sciences, University "Magna Græcia" of Catanzaro, Viale Europa 100, 88100 Catanzaro Italy; (e-mail: nlombardo@unicz.it)

Dyplopia was due to mucocele's extension to orbital muscles. Therefore he underwent to oculistic examination.
The ophthalmological evaluation revelead a normal visual acuity (0.0 logmar in both eyes) and a normal fundus. Hertel's exophthalmometry showed 3 mm of left-side exophthalmos. Results of motility examination showed a 5 PD left hypertropia in primary position that increased to 15 in left downgaze. Downward saccadic velocity was clinical normal in each eye. The ductions and versions' evaluation showed underaction of the Left Inferior Rectus. The Hess–Lancaster test confirmed an isolated left inferior rectus palsy.
We perform our diagnostic protocol for allergic asthma, an iter made by spirometry, Skin Prick Test, total (PRIST) and specific IgE level (RAST). He resulted negative for allergic asthma [2].

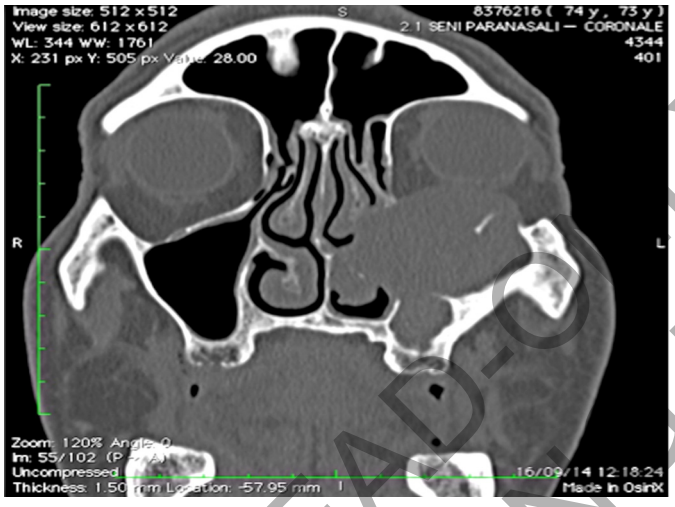

Fig. 1: A coronal CT scan of a left maxillary mucocele eroding orbital floor and medial maxillary sinus wall.

The patient was inserted in a study protocol for unilateral lesions of paranasal sinuses and PET in order to differentiate an inverted papilloma (IP) [3].
CT imaging of the paranasal sinuses (Fig. 1), showed mucous secretion occupying left maxillary sinus, with bulging of the medial wall and narrowing of the osteomeatal complex. In addition, CT images highlighted expansion with bone erosion to the orbital floor until orbital muscles, without clear cleavage plan.
Content density was 10 units HH lower than that of masticatory muscles (orientating for a solid content of the lesion).
Fiberoptic nasal endoscopy showed obstruction of the left osteomeatal complex caused by bulging of the medial wall of the maxillary sinus, without mucous secretion in nasal cavity. We decided to subject the patient to endoscopic sinus surgery in general anesthesia, consisting in removing middle turbinate's lateral side, middle meatal antrostomy and marsupialization with drainage of the mucocele (Fig. 2). We make also a biopsy of the lesion. The diagnosis of a mucocele was confirmed histologically. Patient immediately regain visual function, the post-surgery course was regular. He repeated ocular motility examen and Hess-Lancaster test showing normal result. Endoscopic follow-up showed regular maxillary cavity, no relapse of the disease. Two months later the patient didn't complaint disorder of ocular motility or ocular tension sensation and heaviness to the ipsilateral emiface. The patient has given his informed consent to data treatment.

Discussion

Maxillary sinus mucoceles are rare events, with an incidence of 3-10% in english literature [4,5,6], while in asian literature they are more frequent [7,8]. This surprising difference isn't yet explainable. A reason may be found in the progressive abandonment of the open surgery tec-

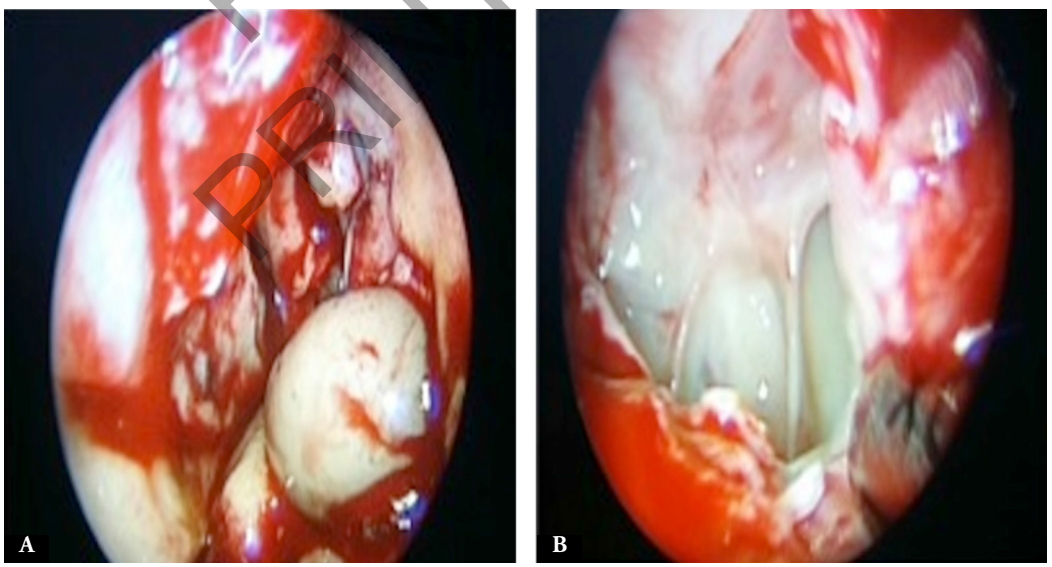

Fig. 2: A) Left maxillary sinus mucocele arising from its medial wall. B) Left maxillary sinus after middle meatal antrostomy and mucocele's removal.

nique (like Caldwell-Luc) and the more frequent recourse to endoscopic tecnique's, that expose frontal and ethmoidal sinuses to a greater incidence of secondary mucoceles.
Mucoceles of maxillary sinus are in most cases secondary to maxillary ostium's obstruction and they are more rarely primitives. The extension of the maxillary mucocele to orbit is very unusual, in fact its spread is usually toward nasal cavity, cheek's soft tissues or toward oral cavity up to expulsion of dental elements [9,10,11]. In our case, the time elapsed between the initial procedure and the development of symptoms was almost 50 years, much more time than other series [12]. In our case diplopia was the first complaint, caused by involvement of inferior rectus muscle. CT study of the paranasal sinuses is gold standard imaging technique, while MRI is more useful in making differential diagnosis with neoplastic lesions. CT scan will show mucocele as an homogenous lesion, which is isodense with brain [5] and contrast enhancement is not necessary, but in our case content density was 10 units HH lower than that of masticatory muscles (orientating for a solid content of the lesion).
Then, in our case, CT leaves us some doubts on lesion's nature that we solved during surgery. The MRI wasn't performed because patient refused.
Today endoscopic treatment of maxillary sinus mucocele is the gold standard [12,13] leaving a role to Caldwell-Luc tecnique only when a combined approach is necessary as in case of extension to face's soft tissues [13]. Generally middle antrostomy, marsupialization and drainage of the mucocele are adequate to solve this pathology [11-15] and we didn't resect the thin wall of the periorbital tissue, to avoid iatrogenic ocular lesions [16]. Orbital floor, showed eroded on TC images, didn't need rebuilding. The immediate resolution of the symptoms after surgery support our choice to not perform bone rebuilding, and literature data[16] showed that bone remodelling occurs spontaneously in the postsurgery scarring process.

Conclusion

Mucocele of the maxillary sinus is a rare condition that requires special attention. The peculiarity of our case is in the ocular symptomatology that can at first mislead the diagnosis. The gold standard treatment is FESS and, in this case, the erosion of the orbital wall did not require reconstruction. In our opinion, in fact, mucocele's drainage following a large antrostomy allows complete recovery of ocular and maxillary function by respecting integrity of the maxillary sinus upper wall mucosa in order to avoid damage to the ocular cavity.

Riassunto

Il mucocele del seno mascellare rappresenta un'entità clinica rara. La sua origine è più frequentemente secondaria a pregressi interventi di chirurgia dei seni paranasali, infezioni o patologia allergica delle fosse nasali, traumi facciali, neoplasie dei seni paranasali. Raramente è di origine primitiva. La relativa maggiore frequenza di casi nella letteratura asiatica è spiegabile col recente abbandono della tecnica Caldwell-Luc a favore dell'endoscopia chirurgica. La particolarità di questo caso che presentiamo è la pressochè completa assenza di sintomatologia naso-sinusale, a favore di sintomatologia oftalmica data dall'erosione del pavimento orbitale e da fenomeni di compressione. La compressione ha determinato un deficit della contrazione del muscolo retto inferiore di sinistra determinando diplopia. L'erosione del pavimento orbitale è stato dimostrato tramite TC.
Il trattamento è stato eseguito con tecnica endoscopica effettuando un'ampia antrostomia e drenando il seno mascellare. Abbiamo notato il completo recupero della funzionalità oculare e dell'integrità anatomica e funzionale del seno mascellare già dopo due mesi, senza il bisogno di procedere alla ricostruzione. Ancora oggi il paziente non ha più presentato segni di recidiva né di alterata motilità oculare.
Concludendo abbiamo voluto porre attenzione sulla particolare sintomatologia del caso e, grazie ad un approccio diagnostico multidisciplinare abbiamo notato che non è sempre obbligatorio ricostruire la lesione ossea ma è sufficiente procedere ad un'adeguata antrostomia e al drenaggio del mucocele, nel rispetto del sottile tessuto che ricopre la parete superiore del seno mascellare per ottenere il completo recupero della normalità anatomo-funzionale della cavità paranasale.

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