Maxillary Sinus Mucocele as a Late Complication in Zygomatic-Orbital Complex Fracture

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Abstract

This article presents an unusual case of maxillary sinus mucocele as a late complication of zygomatic-orbital complex fracture, 23 years after the initial treatment. The patient was referring diplopia and decreased visual acuity with signs of dystopia, proptosis, and epiphora. Computed tomographic scan revealed an expansive lesion in the maxillary sinus with surrounding bone erosion and displacement of the eyeball. Treatment option was excisional biopsy and orbital floor reconstruction with titanium mesh restoring the appropriate orbital position. We propose that in the case of postoperative ophthalmological sequelae of facial fractures involving paranasal sinuses, mucocele should be considered in the differential diagnosis.

Keywords

- mucocele
- maxillary sinus
- proptosis

Mucocele is a chronic mucous-containing cystic lesion that, when affecting the paranasal sinuses, is lined by respiratory epithelium.¹ Paranasal sinus mucoceles may arise from inflammation, tumors, trauma, or surgical injury.² This pathology is usually related to a compromised sinus patency through blockage of the sinus outflow tract or entrapment of mucosa in a fracture line.³

Mucocele rarely involves the maxillary sinus and when it does, the most frequent etiology is either untreated trauma or history of prior sinus surgery. There are a few reports of mucocele secondary to facial fractures.^{4,5} Maxillary mucoceles can cause sinus expansion, bony wall resorption, and ultimately extension through the wall into the adjacent structures. Orbital involvement might occur due to the close anatomic relationship between the orbits and the paranasal sinuses, and thus mucocele may cause orbital displacement, proptosis, diplopia, ophthalmoplegia, and decreased visual acuity.^{6–8}

Although the mucocele is a benign lesion, the invasion of important anatomical structures can cause severe complications and permanent damage. Therefore, the prompt diagnosis and surgical enucleation is required. This study aims to

report an unusual presentation of maxillary sinus mucocele with orbital involvement.

Case Report

A 37-year-old male patient was referred to the Department of Diagnosis and Surgery, Division of Oral and Maxillofacial Surgery, Dental School at Araraquara (Unesp, Araraquara-SP, Brazil) with lower eyelid swelling, dystopia, proptosis and ipsilateral epiphora (► Fig. 1a, b), decreased visual acuity of the right eye with no diplopia, and slight restriction in infraversion. The patient reported surgical treatment of facial trauma 23 years before and the medical record review revealed the occurrence of unilateral transverse fracture of the maxilla and zygomatic complex with displacement and without rotation, including the infraorbital ridge and zygomatic buttress treated by open reduction and internal rigid fixation. In this occasion, any alteration in the maxillary sinus was observed.

The CT scan of the paranasal sinuses showed an extensive lesion with partial resorption of the right orbit floor (► Fig. 1c). The lesion was enucleated by the Caldwell-Luc

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Fig. 1 (a and b) Clinical aspect showing proptosis in the left eye (frontal and inferosuperior view). (c) Computed tomography revealing the presence of expansive lesion in the maxillary sinus causing orbital floor resorption, left side. (d) Excisional biopsy through subciliary approach.

approach and infraorbital incision. (→ **Fig. 1d**); reconstruction of the orbital floor was performed with an individually contoured titanium mesh (→ **Fig. 2a**). The wall of the cyst was dissected away from the orbital contents.

After reconstruction of the orbital floor, the forced duction test confirmed that there was no inferior orbital muscle impingement. The postoperative recovery progressed un-

eventfully, with absence of pain complaints, signs of infection, restriction of eye movements, proptosis, or diplopia (→ **Fig. 2b, c**).

Diagnosis of maxillary sinus mucocele was obtained after histological examination that presented ciliated stratified columnar respiratory epithelium with signs of chronic inflammation (→ **Fig. 2d**).

Fig. 2 (a) Orbital floor reconstruction with titanium mesh. (b and c) Complete tissue healing (frontal and inferosuperior view) on postoperative day 40. (d) Histological examination revealed columnar epithelium that was lining the cystic area and the submucosal tissue contained a moderate degree of lymphocytic inflammation.

Discussion

The mucocele is a unique pathology of the paranasal sinuses, which destroys the surrounding bony structure in spite of its benign nature.⁹ Maxillary mucoceles can expand to the inferior orbital rim causing ocular displacement, proptosis or enophthalmos, infraorbital nerve compression, lower lid distortion, epiphora, limitation of extraocular muscles, and ptosis.¹⁰ This patient had extension of the maxillary mucocele with orbital involvement and dysfunction.

In the early stages, the lesion appears radiographically as a nonspecific, uniformly clouded pathology with normal bony walls. In later stages, the sinus cavity appears expanded, and the walls are thinned and eventually perforated.¹ Once bone destruction has occurred, differential diagnosis should include malignant conditions, such as adenoid cystic carcinoma, squamous cell carcinoma, undifferentiated carcinoma, plasmacytoma, rhabdomyosarcoma, lymphoma, schwannoma, and tumors of dental origin, and a biopsy is required to confirm the diagnosis.^{11–13} CT scan of the paranasal sinuses in coronal and axial view is the gold standard investigation for confirmation and diagnosis and to know the extent of the lesion.⁹ The characteristic image findings on the CT scan for orbital mucoceles are typical space-occupying lesions from the paranasal sinus with surrounding bony erosion (eggshell bone erosion).¹⁰ In this case, CT scan showed a space-occupying lesion from the paranasal sinus with eggshell bone erosion.

Surgical treatment is indicated especially when the mucocele compromises adjacent structures to avoid severe complications and permanent damage. The treatment of choice for this condition is surgical enucleation through an open surgical field, endoscopy or a combined approach.^{14–16} Regarding the necessity of simultaneous lesion enucleation and orbital floor reconstruction with titanium mesh, an infraorbital incision and the Caldwell-Luc technique with total removal of the mucous membrane and inferior antrostomy were performed.

The prognosis for visual disturbances after operation seems to depend on the lapse of time from onset of mucocele until surgery and the severity of preoperative impairment of visual acuity. The gradual deterioration of visual acuity may result from ischemia of the optic nerve due to pressure from mucoceles.^{17–19} In our study, the ophthalmologic symptoms disappear immediately after surgery.

In the literature, the elapsed time to develop the mucocele after craniofacial trauma ranged from 1 to 35 years.^{3,19} In our study, the mucocele developed 23 years after trauma and surgical treatment. Based on these findings, we believe that the lesion occurs due to prior trauma/surgery. Patients who sustain craniofacial trauma may require regular term follow-up to evaluate for mucocele formation.

Conclusion

This case report highlights that maxillary sinus mucoceles grow slowly and may encroach the inferior orbit stealthily

with various ophthalmic manifestations. When patients with history of trauma involving the paranasal sinus area present ophthalmic symptoms, mucocele should be considered in the differential diagnosis. Appropriate and prompt treatment can lower morbidity especially in cases of lesions which involve relevant anatomical structures.

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