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Post-Traumatic Giant Frontal Sinus Mucopyocele: A Case Report

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Abstract

Paranasal sinus mucocele is a benign cystic lesion, of chronic nature, lined with mucosa and filled with mucous content. Obstruction of the frontonasal duct, where frontal sinus drainage occur, leads to mucocele formation, which grows slowly, expanding into the surrounding bony walls with subsequent bone destruction and resorption. Mucocele expansion tends to occur in the direction with the least resistance, thus frontal sinus mucocele has the tendency to expand inferiorly, eroding the superior orbital wall and causing inferior displacement of the globe with consequent ophthalmic symptoms such as: Diplopia, ptosis, proptosis, epiphora or amaurosis in cases of optic nerve compression by the mass effect of mucocele. This case report presents a 42-year-old male with 18 years giant post-traumatic frontal sinus mucocele with orbital involvement and extensive right eye proptosis, where conservative pharmacological treatment was unsuccessful and surgical treatment was favored with good post-operative evolution.

Keywords: Mucocele • Frontal sinus • Proptosis • External approach • Job tasks

Introduction

Paranasal sinus mucocele is a benign cystic lesion, of chronic nature, lined with mucosa and filled with mucous content. Incidence of mucoceles varies in the literature from being divided equally between males and females to a ratio of 7:1 with a predominance at the 3rd and 4th decades of adult age 6-7. Drainage of the frontal sinus is achieved via the corresponding frontonasal duct bilaterally. Obstruction of this drainage leads to mucocele formation, which grows slowly, expanding into the surrounding bony walls with subsequent bone destruction and resorption [1]. Mucocele expansion tends to occur in the direction with the least resistance, thus frontal sinus mucocele has the tendency to expand inferiorly, eroding the superior orbital wall and causing inferior displacement of the globe with consequent ophthalmic symptoms such as diplopia, ptosis, proptosis, epiphora or amaurosis in cases of optic nerve compression by the mass effect of mucocele. Serious complications include mucopyocele, bony erosion with orbital or encephalic involvement. Mucocele formation maybe attributed to inflammation, allergy, tumors, previous sinus surgery and trauma to the frontal sinus, all of which are conditions that lead to obstruction of the frontonasal duct. A mucopyocele or pyocele is the name given to infected mucocele, where the cyst is completely or partially filled with purulent material [2].

Case Presentation

A 42 years old male presented to the emergency department at the hospital of craniofacial anomalies rehabilitation in Bauru, Sao Paulo, presenting a decrease in visual acuity and progressive orbital proptosis with an onset of 2 months. He has a history of cranioencephalic trauma after an automotive accident occurring 18 years ago, with a fracture of the bony walls of the right frontal sinus wall treated by osteosynthesis in another hospital.

Upon physical examination, presents an extensive proptosis of the right eye with a loss of the eye contour, conjunctival edema, corneal opacity and visual acuity limited to light perception in the right eye (Figure 1) [3].

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Figure 1. A) Pre-operative right eye proptosis; B) Immediate proptosis regression post-operatively; C) Coronal flap; D) Coronal flap+frontal sinus defect.

Left eye is normal. Computerized tomography of the face shows hypoattenuous lesion occupying the frontal sinus, orbit and ethmoidal sinus on the right side causing partial obliteration of the intraconal fat with inferomedial displacement of orbital structures and subsequent protrusion of the right ocular globe (Figure 2) [4].

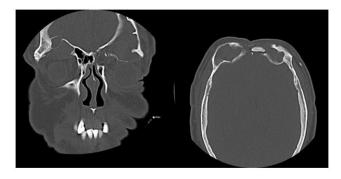


Figure 2. Coronal and axial pre-operative CT image.

Pharmacological treatment with ciprofloxacin shows no improvement. Surgical intervention was the treatment of choice *via* coronal access, with an access to the frontal sinus *via* anterior bony wall, that was found necrotic. Large amounts of purulent material were drained and osteosynthesis material that was found inside the sinus was removed (Figure 3) [5].

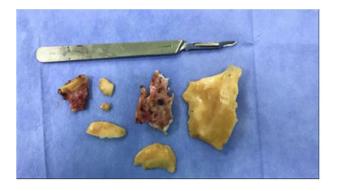


Figure 3. Frontal bone fragments.

After excessive irrigation with normal saline, the anterior wall was reconstructed with periosteal flap. Immediate post-operative evaluation showed a large degree of proptosis regression [6]. The patient made an uneventful recovery and presented improvements in ocular proptosis after discharge from hospital. Eighteen months follow-up had shown no signs of recurrence, good healing aspect with no limitation of extrinsic ocular muscles, slight dystopia is observed which resulted in facial asymmetry, gross visual acuity at left eye was 20/20 and right eye 20/200 (Figures 4 and 5) [7].



Figure 4. No limitation of extrinsic eye movements was observed 18-months post- operatively.



Figure 5. Face contour of the patient 18 months post-operatively.

Post-operative CT scans were obtained, showing no recurrence. Ophthalmologic follow-up due to corneal ulceration and visual acuity reduction are indicated. The patient is currently on periodic postoperative follow-ups (Figure 6) [8].

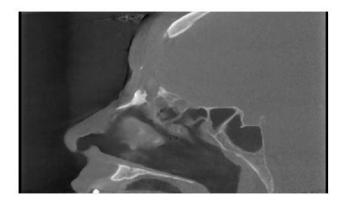


Figure 6. Sagittal image for 18 months post-operative CT scan.

Results and Discussion

Paranasal mucoceles are benign, epithelium-lined cysts filled with mucoid material. Mucoceles develop when the sinus ostium is obstructed resulting in progressive accumulation of secretions and epithelial cells in the sinus cavity, with subsequent expansion of the involved sinus. Sinus expansion is a necessary key to finding in the diagnosis of mucocele. In the absence of expansion, the term sinus obstruction should be used. The etiology of obstruction in mucocele is variable and includes inflammation, trauma and tumor. Predisposing factors are summarized. The frontal and ethmoid paranasal sinuses are involved in up to 90% of cases. The maxillary sinus is affected less frequently (10%) and the sphenoid sinus only rarely. In children, an unusual variant can involve the nasolacrimal duct. In some cases with extensive osteolytic destruction of the surrounding anatomical structures, the primary site of the mucocele cannot be determined. Young adults (20-40 years) are most commonly affected [9].

Mucoceles that are sufficiently large may exert mass effect on the surrounding anatomic structures. The clinical symptoms vary according to the location. A literature search for cases of giant frontal mucoceles yielded 13 relevant articles. Giant mucoceles of other paranasal sinuses are beyond the scope of this article. Cases were included as long as they presented scientific rigour and relevant bibliographic sources. Eight cases were excluded due to a lack of descriptive parameters and/or images of the mucoceles [10]. The remaining five cases are summarized. In the current literature there are no criteria regarding the use of the term giant mucocele. Based on the reported dimensions in other case reports, we propose 5 cm as a cut-off value of the lesion size. CT and MRI are complementary when imaging mucoceles. CT depicts an expansile, homogenous mass with remodeling of the adjacent bone. Occasionally, a mucocele may cause bone destruction simulating an aggressive neoplasm [11].

In the three cases where contrast was administered, subtle peripheral enhancement was seen, similar to characteristics of smaller mucoceles described in the literature. MRI can be helpful when differentiating mucoceles from other aggressive lesions. Signal intensity on T1-WI is variable (low in case of low protein content and high in case of high protein content). All reviewed giant mucoceles were bright on T2-WI, which may be explained by their high fluid content. However, desiccation of the mucocele contents has been

described in chronic non-giant cases, resulting in decreased signal intensity on T_1 and T_2 weighted images [12].

Mucoceles are expanding masses, usually mucus-filled, when infected they are called mucopyoceles. Differential diagnosis of mucocele is obtained according to inflammatory, genetic, cystic and neoplastic considerations, for the likes of Rathke cleft cyst, dermoid cyst, pituitary adenoma, craniopharyngioma, optic glioma and neoplastic lesions of the cranial base, facial sinuses and nasopharynx. In malignant tumors of paranasal sinuses, ocular proptosis occurs in 4.5% to 38% of the cases. Computerized tomography is considered the exam of choice as it allows visualization of the degree of sinus expansion, bony destruction and involvement of neighboring structures. Occasionally, frontal mucoceles can simulate sinus/orbital neoplasms in CT scans [13].

Conclusion

The use of heterologous graft for the osteosynthesis of frontal sinus, like hydroxyapatite or methyl methacrylate, can be associated to the development of mucopyocele. Due to the harder consistency of the graft material as compared to frontal bone, infection can spread easily to the orbit, in addition to making the surgical access to the frontal sinus ostium more challenging. Naso-orbital duct obstruction subsequent to frontal trauma or to obstruction by heterologous graft material without adequate sinus follow-up is considered the most probable cause of the mucocele formation in frontal sinus.

Conflict of Interest

The authors report no financial support and no conflicts of interest.

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