

Frontal sinus mucocele with advanced orbital involvement – a case series

Śluzowiak zatoki czołowej z zaawansowanym zajęciem oczodołu – seria przypadków

Authors' Contribution:

A – Study Design
B – Data Collection
C – Statistical Analysis
D – Manuscript Preparation
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ABSTRACT:

Introduction: Mucoceles of the paranasal sinuses are more common in the frontal sinus than other sinuses due to multifactorial causes. Their close proximity to the orbit can lead to complications like diplopia, severe proptosis or even vision diminution. CT being the modality of choice, clearly demonstrates bony walls of paranasal sinuses and the extension of a mucocele into the surrounding structures. Mucoceles can either be excised endoscopically or by external approach.

Case report: We describe our experience of three frontal mucocele cases all of which showed advanced orbital involvement and were managed successfully by endoscopic approach with no complications or recurrence. All symptoms including diplopia and proptosis significantly improved in every case. Hence, we reiterate the management of paranasal sinus mucocele via endoscopic approach as the preferred surgical modality having advantage of shorter recovery time, easy access, lower morbidity and incidence of potential complications.

KEYWORDS:

endoscopic approach, frontal sinus, mucocele

STRESZCZENIE:

Wprowadzenie: Śluzowiaki (*mucocele*) zatok przynosowych z różnych przyczyn częściej niż w pozostałych zatokach występują w zatokach czołowych. Ich lokalizacja w bliskim sąsiedztwie oczodołów może prowadzić do powikłań, takich jak: diplopia, ciężka proptoza, a nawet osłabienie wzroku. Tomografia komputerowa, jako preferowana metoda obrazowania, pozwala na uwidocznienie kostnych ścian zatok przynosowych i zasięgu śluzowiaka w obrębie otaczających go struktur. Śluzowiaki można wycinać endoskopowo lub z podejścia zewnętrznego.

Opis przypadku: W artykule opisujemy nasze doświadczenie obejmujące trzy przypadki śluzowiaków charakteryzujących się zaawansowanym stopniem zajęcia oczodołu i poddanych skutecznemu leczeniu bez następstw w postaci powikłań lub nawrotów. We wszystkich doszło do znacznej poprawy w zakresie wszystkich objawów, w tym diplopii i proptozy. W świetle powyższego, ponownie podkreślamy preferencyjny charakter endoskopowego podejścia do leczenia śluzowiaków zatok przynosowych jako modalności charakteryzującej się: krótszym czasem rekonwalescencji, łatwością dostępu, niższą śmiertelnością i niższym odsetkiem potencjalnych powikłań.

SŁOWA KLUCZOWE: podejście endoskopowe, śluzowiak, zatoka czołowa

ABBREVIATIONS

CT – computed tomography
PNS – paranasal sinuses

INTRODUCTION

Mucoceles of the paranasal sinuses (PNS) are cystic expansile lesions first described by Lageback in 1820 [1]. The term mucocele was coined by Rollet in 1909. They are most commonly found in the frontal sinus

(60–65%) but can also occur in ethmoid sinuses (20–25%), maxillary sinus (10%), sphenoid sinus (1–2%). The etiopathogenesis of mucocele in the paranasal sinus is multifactorial with most common causes being inflammation, allergy, trauma or less commonly – secondary to neoplastic lesions in PNS obstructing sinus ostia [2–4].

Mucocele can be asymptomatic if small. If it grows however due to its expansile nature, ophthalmic and intracranial complications can sometimes occur as a result of close proximity of the paranasal sinus to the orbit and brain. Clinically, mucocele has to be differentiated from sinusitis and mucous retention cyst due to similar presentation.

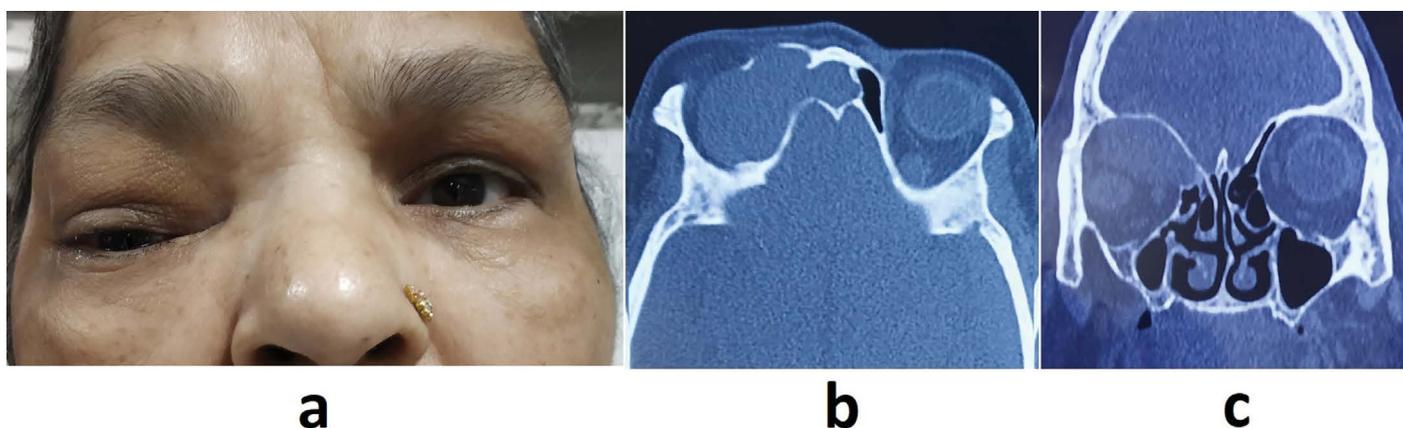


Fig. 1. Clinical (a.) and radiological (b., c.) image of case 1.

Computed tomography (CT) scan of paranasal sinuses can help in this differentiation [3].

Management of mucocele is only surgical with two main approaches. It is either excised endoscopically or by external approach. Both approaches have their advantages and disadvantages and decision to use a particular approach is based on the site, extent of mucocele and whether it is recurrent or not.

In this series we describe a detailed ocular and sinonasal examination including visual acuity before and after surgery, imaging findings, treatment and outcome of treatment. All three cases of mucocele were successfully operated on in our institution endoscopically.

The authors declare that they followed the protocols of their work center on the publication of patient data and that all the patients included in the study received sufficient information and gave their informed consent in writing to participate in that study.

PATIENTS AND METHODOLOGY

Patient 1.

A 58-year-old female presented with complaints of painless supra-orbital swelling with proptosis of the right eye and double vision slowly progressing for the past seven years. She had already undergone an endoscopic procedure for the same problem three years earlier but had recurrence of symptoms 4 months later. On palpation the swelling was diffuse, puffy and non-tender. The right eyeball was pushed laterally and inferiorly by the swelling causing restrictions of the eyeball movements. Vision in the affected eye was 6/18. Endoscopic examination revealed adhesions between the middle turbinate and the orbital medial wall causing obstruction to the frontal recess outflow pathway. CT imaging revealed a frontal mucopyocele causing bone thinning of the posterior and anterior wall of the frontal sinus extending laterally up to the lateral orbital wall (Fig. 1.).

Patient 2.

A 42-year-old female presented with swelling in the medial canthal region of the right eye progressively increasing in size for the past

3 years. The swelling was painless though it was associated with recurrent frontal headaches during episodes of upper respiratory tract infections. On examination the swelling was compressible and non-tender. Proptosis was present pushing the right eyeball infero-laterally and causing restriction of vertical eye movements leading to diplopia but vision was normal. Nasal endoscopy revealed inaccessible right frontal recess area due to lateralization of the middle turbinate. CT imaging showed an expansile cystic swelling in the right frontoethmoidal region pushing the globe inferolaterally without any bony erosions (Fig. 2.).

Patient 1.

A 36-year-old female presented with left eye movements being affected due to extreme proptosis pushing the eyeball infero-laterally. On examination, the affected eye had exposure keratitis. Vision was 6/24. There was a swelling above the medial canthus of the eye which was soft, compressible and non-tender on palpation. CT imaging revealed a swelling occupying the frontal sinus and the ethmoidal region with loss of the anterior wall of the frontal sinus and extending into the orbit causing proptosis (Fig. 3.).

All the patients underwent endoscopic sinus surgery and the frontal recess area was uncapped and opened up. In all cases, around 20–30 mL of pus was drained and the sinus was cleared. The middle turbinate was trimmed and adequately medialized, and the frontal sinus opening was widened and packed with anterior nasal packing. Pack removal was carried out after 48 hours. The patients were followed after 1 week, 2 weeks and then monthly for a period of 6 months. They remained recurrence-free.

DISCUSSION

Paranasal sinus mucocoele is one of the most common benign expansile lesions of the PNS. The incidence of mucocoele in PNS is going down probably due to early and more effective treatment of sinusitis, resulting in lower incidence of complications. Mucocele primarily develops when the sinus ostium is obstructed either due to inflammation, trauma, allergy, presence of paranasal sinus, polyposis, tumors, previous sinus surgery or any other congenital anomaly [5].

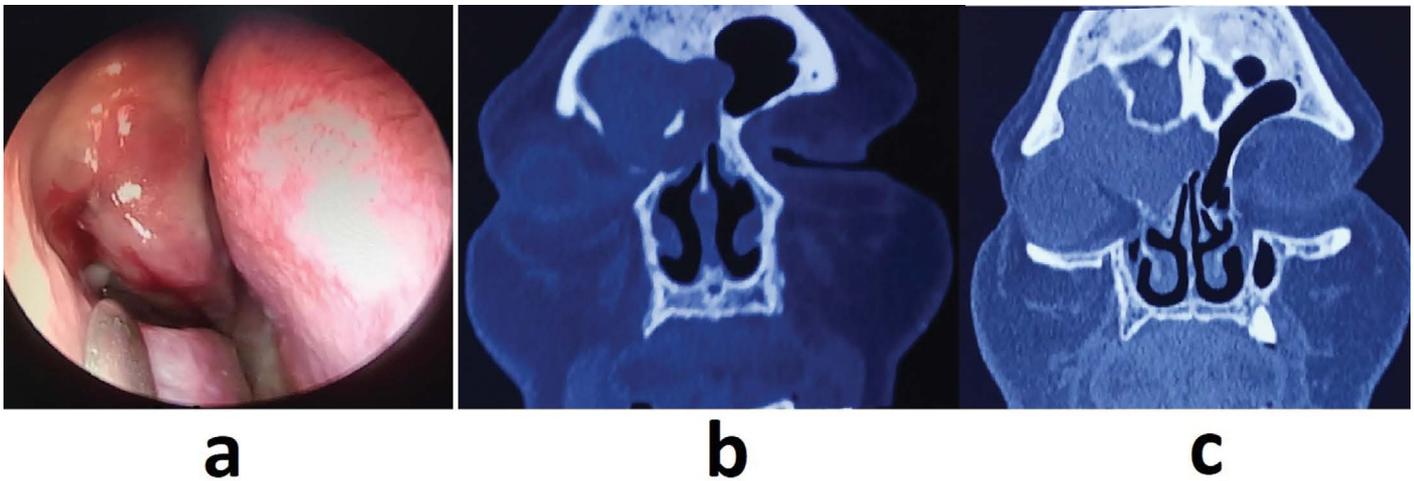


Fig. 2. Endoscopic (a.) and radiological (b., c.) image of case 2.

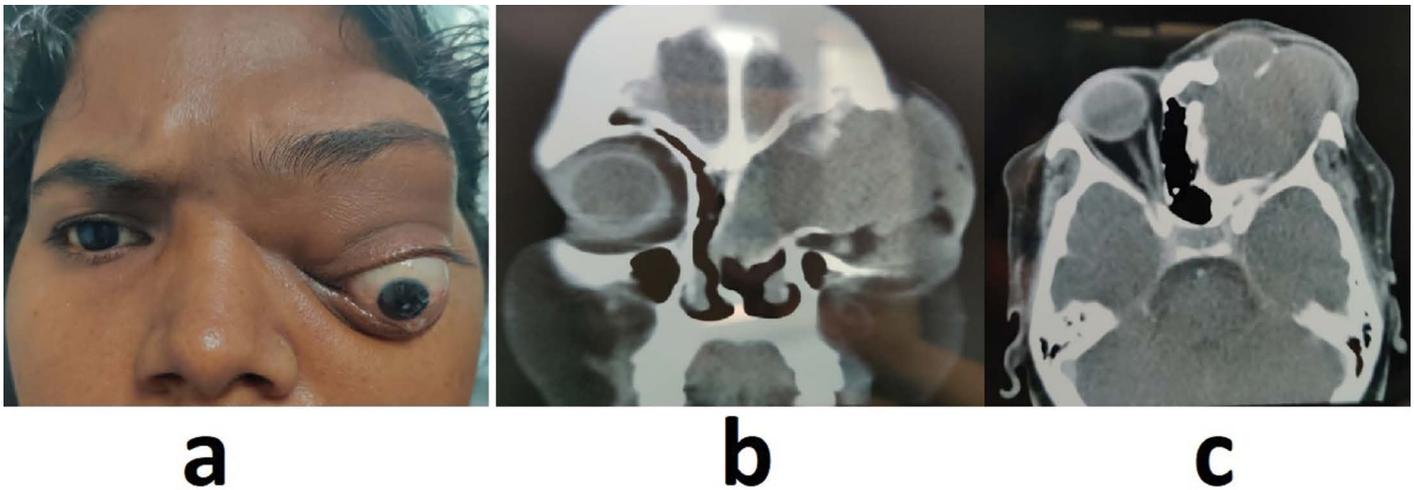


Fig. 3. Clinical (a.) and radiological (b., c.) image of case 3 showing extreme proptosis.

In our series only one case had a history of trauma, 7 years ago. Literature reports that the mean time to develop mucocele is 5 years [6] but this case probably developed due to allergy or inflammation. A predisposing factor could be identified in more than 50% of patients out of which only 45% were primary in a study carried out by Maria et al. [7]. Bockmöhlet et al. and Kyung Lee et al. reported 66% of secondary mucocele and 64.5% of secondary mucocele respectively [8, 9]. Mucous accumulation in a mucocele leads to increase in the size of mass with bony wall expansion due to pressure which is further enhanced by prostaglandins and collagenase which aid in osteolysis [10–12].

Mucocele affects all age groups but it is most common in the 3rd and 4th decade of life. In our series, all 3 cases were females, aged between 36–58 years. A lot of authors have reported a similar higher female predilection [13, 14] in their series while others reported the opposite [10, 15].

In our series, the frontal sinus was the most common site involved along with the ethmoid sinus, a finding which is corroborated by most of earlier studies [10, 15, 16]. Very rarely cases have been described in the concha bullosa, lateral sphenoid recess and orbital floor [17].

Signs and symptoms of mucocele are variable as they depend on the site affected. They can usually cause headache, peri-orbital pain, and decreased/blurred vision. Frontal ethmoid mucocele due to its pressure exerted on the orbit, usually presents with proptosis, diplopia, peri-orbital swelling and hypophthalmus [11, 18, 19].

In our series the most common symptom was diplopia. Decreased vision is most commonly associated with posterior ethmoid, sphenoid mucocele as lamina papyracea may be pushed into the optic canal leading to optic neuropathy [18]. Decreased vision can also be present in exposure keratopathy due to proptosis. In our series proptosis was present in all cases with exposure keratitis in one patient. On the contrary, Jaswal et al. reported proptosis in 1/3rd of cases only [14]. Literature also shows that around 14% of mucoceles are often asymptomatic.

If a mucocele becomes infected it might also present with symptoms of sinusitis, fever, erythema and orbital cellulitis [10,20]. Very rarely intracranial involvement, and possible complications were reported on, with meningitis, subdural or cerebral abscess or pneumocephalus [21]. Sometimes the 3., 4., 5., or 6. cranial nerve and rarely the pituitary gland can be involved [19].

CT scan is the imaging modality of choice for PNS mucocele as it clearly demonstrates bony walls of PNS with the extent of the mucocele into the surrounding structures [9, 16]. Mucocele is seen as an airless non-enhancing iso-dense/mildly hyper-dense homogenous mass on CT [3]. Van tassel et al. reported a 66% incidence of hyper-dense mucocele on CT in their study [22]. Mucopyocele may appear as denser and peripherally enhanced. MRI might be needed in cases of CT showing intracranial extension and partial loss of vision to identify brain tissue and orbit involvement, and to differentiate mucocele from other soft tissues neoplasms [23, 24].

Treatment of mucocele is only surgical, aimed at restoring sinus drainage. It can either be approached endoscopically or through conventional external approach. In patients with decreased vision, urgent surgery is needed, preferably within 24 hours after the onset of visual disturbances. Endoscopic Endonasal Approach is the most favored approach these days due to short recovery time, easy access, lower morbidity and lesser incidence of potential complications compared to the intracranial route [25]. External approach is usually reserved for a frontal mucocele which only affects the most external and postero-superior region of the sinus, or in recurrent cases or mucocele secondary to osteoma or orbital herniation. External approach has the advantage of exposure of a complete sinus and provides opportunity for obliteration of the sinus to reduce recurrence. It also allows for curettage under vision of the exposed dura [26].

Mucocele should be widely opened and an adequate part of its wall should be excised to ensure sufficient drainage and ventilation towards the nostril. Periosteum should be left intact to allow normal osteogenesis and bone remodeling [27]. Emphasis should be given to preserve normal mucosa and complete mucocele mucosa removal

is not required. Also, it is important to carefully assess mucocele mucosa close to the orbit and skull base, especially if dura or periorbital dura is exposed, if possible preserve it [5], as mucocele mucosa acts as a cover to preserve vital structures underneath. Adequate marsupialization is the key to prevent recurrence and in some cases to improve vision [23]. However, in case of complete preoperative loss of vision, the prognosis is slightly poor. In our series, we approached all the cases successfully with endonasal endoscopic approach even though one of the cases was recurrent. Vision improvement was seen in all our patients.

Recurrence rate as high as 23% has been reported on in a series of fronto-ethmoidal mucoceles. This was probably due to a long follow-up, which was around 7 years. This author also reveals that recurrence can occur as long as after 41 years, thus explaining the importance of long-term follow-up [27]. Courson et al. [28] published a series of 957 fronto-ethmoidal mucoceles in which he reported that the recurrence rate of patients operated endoscopically and externally was similar. We found no recurrence. However, we had a follow-up of only around 6 months. Long-term follow-up is required in our study for validation of our results.

CONCLUSION

Paranasal sinus mucocele primarily affects the frontoethmoidal complex. Endoscopic endonasal approach is the treatment of choice as this is a safe and effective technique in most of the cases of mucocele. Immediate surgery can avoid permanent visual impairment and other sequelae. Surgery within 24 hours of onset of decreased vision is usually recommended.

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