Case Report:

Frontal Mucocele With Proptosis: DrafIIa Approach

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Abstract:

Paranasal sinus mucocele commonly involved fronto-ethmoidal region rather than other due to its narrow anatomical drainage outflow, which put them at a higher tendency to get obstructed. Usually, it arises from an identifiable cause such as a history of endonasal surgery, facial trauma or background of nasal allergy or rhinosinusitis. Rarely patient presented with primary frontal mucocele, and its presentation depending on the mucocele location and extension with surrounding mass effect. Treatment is based on restoration of frontal sinus drainage and ventilation or towards a more radical and definitive approach which is sinus obliteration. Risk and benefits between these two need to be taken into consideration. We present a case of primary frontal mucocele with gradual onset of unilateral eye proptosis which first presented solely with ophthalmic symptoms.

Keywords frontal mucocele, drafIIa, eye proptosis, marsupialization

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Introduction

Mucocele of paranasal sinuses is a benign lesion yet having the expansile capability of all dimension[1]. It can occur secondary to facial trauma, previous surgery, or spontaneous obstruction of sinus tract outflow[2]. It can also occur in a patient with a background of nasal allergy and chronic sinusitis[3]. Primary mucocele is when there is no causative factor identified, whereas in secondary mucocele, it is due from any reason that hinders the normal mucociliary drainage through its ostium. Primary mucocele commonly arose from an ethmoid sinus (45.5%), followed by maxillary sinus (18.2%)[2]. In contrast to secondary mucocele, it is vice versa[2]. In frontoethmoidal mucocele, there is no known causes identified and it is the commonest anatomical region to be involved possibly due to intrinsic narrowing and tortuosity of the frontonasal duct, whereby even a minor obstruction due to minimal inflammatory process leads to full-blown outflow obstruction[4]. The pressure from continuous accumulation of fluid inside its mucocele wall can cause a bony reaction to the surrounding bones, leading to the softening of the bony wall. Frontal sinus mucocele commonly extends laterally into orbit, superiorly into anterior cranial fossa, anteriorly into the frontal outer table and forehead, and posteriorly into ethmoidal sinuses.

Case Report

20-year-old Malay female, a Thalassemic trait patient, presented with a persistent left throbbing frontal headache for one year. She has no nausea, vomiting or bodily weakness or numbness. There was no history of trauma to the forehead or orbital area and no history of sinus surgery before. She has no chronic rhinosinusitis symptoms. However, she noticed her left eye was gradually proptosed about a year ago. She never seeks any medical attention until about one month before the presentation,

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where she started to have double vision, and this had brought her to see an ophthalmologist. There was no ophthalmoplegia, eye discharge or redness. Otherwise, there was no tuberculosis contact and no history of chronic inhalation of wood dust or chemical fumes. Eye assessment was done. Subsequently she was referred to otorhinolaryngologist to rule out paranasal sinus disease.

Examination revealed a unilateral left eye proptosis (Figure 1) which was not pulsatile. Otherwise, no bulging of the forehead or medial canthal seen. There was no paranasal tenderness. Cranial nerve examinations were normal except for left III, IV and VI, whereby the eyeball was unable to turn laterally and upwards due to downward and lateral displacement from the mass effect causing her to have diplopia on looking at superolateral aspect. The rigid nasendoscopy examinations of the bilateral nasal cavity were normal. There was no palpable neck node. The oral cavity and the oropharynx were normal.

Figure 1: Pre-operative image. Note the left eye proptosis.

A Contrast-enhanced computed tomography scan was done to evaluate the lesion and its relationship with the surrounding structures (Figure 2-4).

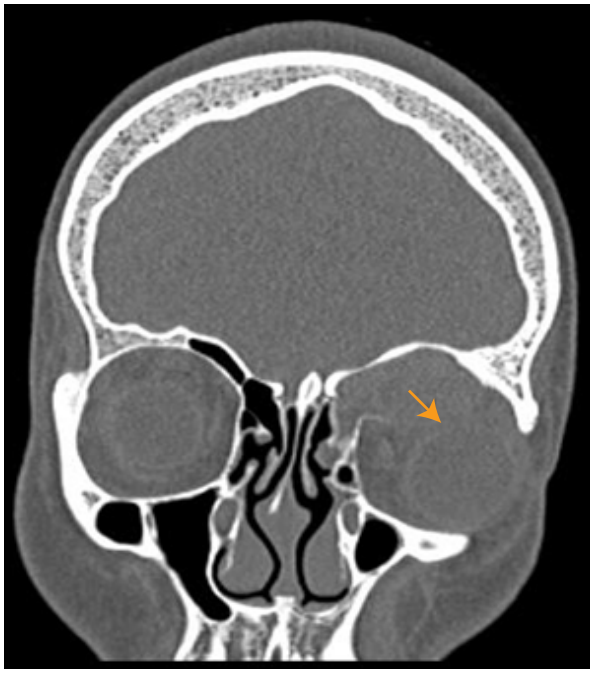

Figure 2: Coronal view (bone window) of Paranasal Sinuses showing poorly enhancing soft tissue mass occupying the left frontal sinus causing bony expansion. The surrounding bone seems thinned, with areas of bony discontinuity near the roof of left nasal cavity and roof of left orbital rim. The frontal mass measured 3.9cm x 2.3cm x 2.6cm (width x anteroposterior x cranio-caudal) in dimension. It was homogenously seen without any calcification. The mass extends into left orbital area, causing displacement of the left eye globe infero-anteriorly without infiltration into the eye globe (red arrow).

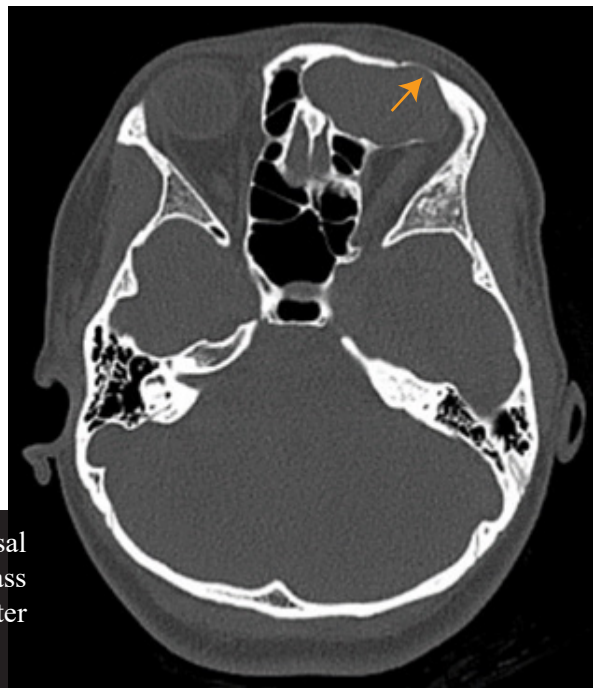

Figure 3 : Axial view (bone window) of Paranasal sinus, showed left frontal homogenous mass extending into left orbit, with papery thin outer table (red arrow).

Figure 4 :Parasagittal view (bone window) of Paranasal sinus, showed left frontal homogenous mass with thinning of anterior cranial fossa bone with the cortex continuity still intact. No intracranial extension seen (red arrow).

Therefore, the diagnosis of the frontal mucocele with mass effect (left proptosis) was made. The patient underwent left anterior functional endoscopic sinus surgery with marsupialization of the left frontal sinus mucocele.

Intraoperatively, the left maxillary sinus showed healthy mucosa, and there was no pus or polyps seen. The left agger nasi was small. The left bony wall of bulla ethmoidalis was sclerotic and appeared thickened. Upon uncapping and removal of the bony wall of bulla ethmoidalis, there was a yellowish cyst seen which became more bulge upon pressing from the left supraorbital rim. The mucocele was then marsupialized. About 4mls of thick yellowish curd-like mucopus drained. The frontal sinus ostium was visualized using a 70-degree rigid nasoendoscope and the surrounding bone was left untouched i.e. no curettage or bony scraping done. After ensuring the frontal sinus ostium was patent and the mucocele lining properly marsupialized into the nasal cavity, the bulla ethmoidalis cavity was then packed with modified sinus packing, using a trimmed merocele that can be fitted into the frontal recess. This packing is chosen as it provides good expansion therefore maintaining the patency of the sinus outflow tract, as compared to other nasal packing.

One month post operativly, (Figure 5) the patient's eye were symmetrically levelled and no more evidence of proptosis seen. The eye movement was full in all direction and there was no symptom of diplopia upon looking superolaterally. From rigid nasondoscopy, it showed a patent frontal recess with minimal synechiae.

Figure 5: Post-operative day 30. Note the symmetrical eye level. Resolved proptosis.

Discussion

The mucocele content can be liquid, or once there is a superimposed infection, it can lead to mucopyocele. The mucopyocele of the frontoethmoidal sinus can lead to orbital infection(5, 6) mimicking orbital complications of an acute sinusitis. Mucocele is a slow-growing mass. At its early stage a small, localized frontal mucocele usually does not cause any symptom but at the later stage it will eventually expand enough to cause an effect to its surrounding structure. Symptoms that may arise due to this local extension of the mucocele includes frontal headache[1,3], diplopia[1,3,7], proptosis[1,3,8], frontal mass[7], craniofacial disfigurement[9] and nasal blockage[3,9].

The preferred imaging modalities for paranasal sinus mucocele is Computed Tomography (CT). CT provides an excellent diagnostic tool, as well as to radiologically mapped the surgical landmark. CT scan of mucocele will show smooth rounded bony remodelling in response to compression pressure exerted by the expanding mucocele. The content of the mucocele will be seen as a homogenous opacification and its attenuation is dependent on its content. Mucoid content will show a 10-18 Hounsfield Unit (HU), while chronic mucocele with proteinaceous content will show a

20-40 HU range[10]. This information reflects the chronicity of the disease.

Contrast media is not administered routinely, but when administered, it can light up the mucocele lining[10],indicating a soft tissue extension of the frontal mucocele. Magnetic Resonance Imaging (MRI) can also aid in delineating the mucocele lining. Apart from that, MRI is best reserved for mucocele, suspected to arise secondary to a sinonasal tumor[10]. MRI can also provide extra information pertaining the orbit when the cause of proptosis or diplopia is unexplainable[8].

The definite management of frontal mucocele is still debatable whether to go for a conservative approach to restore the frontal sinus drainage or to go for a more radical procedure i.e. the obliteration of the frontal sinus[11]. The latter is spared as a last resort, when the endonasal and external approach are not feasible, as well as failed previous conservative measures[12]. Even though the controversy between these two have their pros and cons, some author advocates that obliteration of frontal sinus is the definite management for frontal sinus disease[12].

Marsupialization of the frontal sinus mucocele with orbital extension can be approached via a transnasal endoscopic fronto-ethmoidectomy[8]. While a frontal mucocele with intracranial extension is usually approached externally with the aim of a better surgical visualization, access and ease for reconstruction[1]. This approach is via coronal incision, frontotemporal craniotomy, removal of the posterior table of the frontal sinus, separating mucocele from extradural frontal lobe and removal of mucocele. The reconstruction can be achieved by harvesting a pericranial flap to obliterate the frontal sinus and its foramen followed by a dural repair[1]. However, the patient should be well explained that this procedure is more radical and has a higher morbidity[12]. In our case, the frontal mucocele was decompressed and marsupialized after the uncapping of the frontal sinus ostium through the removal of the bulla ethmoidalis bony wall. This method is called Draf Type II surgery[13]. The surgery was decided as it has a lower morbidity, while maintaining the forehead sensation, and providing adequate postoperative nasoendoscopy surveillance. However, this approach has the disadvantage of an increased restenosis rate, prolonged follow-up, and the possibility of hardships encountered during revision surgeries[12].

In the early days, frontal mucocele with orbital invasion is almost always treated via an open external approach[14]. Using the Killian's method, the surgeon will performed external frontal sinusotomy where intraoperatively, it will able to confirmed that frontal mucocele compressing the optic canal[14]. Postoperatively the exophthalmos will be resolved but some of the patient will have atrophic optic disc[14].

The same author also describes another case series he encountered whereby the frontal mucocele had invaded the retrobulbar portion of the optic nerve, and the mucocele was successfully marsupialized via transnasal approach. Post-surgery, the ophthalmic complaint resolved; however, the author encountered the same complication which is the atrophic optic disc. All these 3 cases involved elderly patients with acute and a long-standing history of ophthalmic complaints[14]. The aetiology of optic nerve disturbance was believed mainly contributed from the compressive effect of the mucocele, leading to disruption of blood flow to the orbit, as well as the inflammatory process that was spreading to the orbital content[14]. Complete resolution of proptosis and orbital symptoms may be achieved within six to twelve months after the surgery to the frontal mucocele[1,8]. However, in cases of the optic disc atrophy, it was irreversible damage and no treatment can be offered whereby the patient might progress to vision loss and eventually blindness[15]. Optic disc atrophy usually preceded by optic disc oedema, and in the latter condition, the vision might be preserved as there is role of steroid described in the literature[15]. Patient with optic nerve compression can experienced reduce central and peripheral vision with visual field defect[15]. Therefore, it is important for early intervention of suspicion of optic nerve compression which diagnosis can be established by doing simple bed side ophthalmoscopy.

Loo JL et.al in their case series, found that the risk of optic neuropathy and poorer visual outcome is higher when there is the involvement of sphenoid sinus and Onodi cell mucocele, possibly due to close relationship with the optic nerve[16]. If the concurrent ethmoidal air cells involved are the anterior group, the risk of vision compromise is much lesser[16].

Some authors suggested a few parameters to predict the outcome of endonasal and external approaches of frontal mucocele marsupialization. The parameters include the type of obstruction

(membranous vs. bony), the number of mucocele (single vs. multiple), site of mucocele (medial vs. lateral) and size of mucocele less or more than 20mm). Although they found that none of these parameters correlates with the outcomes of frontal sinus outflow patency, they observe that membranous type, solitary mucocele and location nearer to frontal outflow tract showed better outcome[11].

While there is no study done to evaluate orbital recurrence in cases of post endoscopic marsupialization of frontal mucocele, there were 2 isolated cases reported regarding this disease recurrence. The first case was disease recurrence following an endoscopic nasal surgery whereby the approach used was bicoronal flap and facial craniotomy, followed by frontal sinus obliteration. The other case was disease recurrence after two times endoscopic nasal surgery, where the approached was through the external Lynch Howarth incision, In that case, the mucocele was carefully dissected away from the periorbita completely, then followed by frontal sinus obliteration[17].

Frontal mucocele is well known to be a highly recurrent type of disease, therefore regular follow up is important to ensure frontal sinus ostium is patent and there is no restenosis happen[11]. The recurrence rate reported was 14% for open surgical approach to frontal mucocele[11]. However, there was no data available to evaluate outcome of surgery for endoscopic technique as the restenosis of frontal outflow tract which cause frontal sinus symptom are often delayed by more than 10 years[18], but the study available is within shorter period of follow up[11].

Conclusion

Proptosed eye without any sign of intracranial space-occupying lesions warranted an evaluation by ENT to further seek the cause due to its proximity to the nasal cavity and sinuses. Benign lesions such as sinus mucocele can cause local mass effects. Endoscopic approaches, external approach or combination of both can be used to remove the mucocele that hinders the outflow tract. In frontal mucocele, even though the definite management is frontal sinus obliteration, a more conservative method to restore the frontal sinus drainage and ventilations still offer good outcomes.

Source of fund (if any)

None to declared.

Conflict of interest

None to declared.

Ethical clearance

No ethical approval needed for this case report writing.

Informed consent was obtained from the patient for this case report.

Authors' contribution

1. Dr Siti Nazira binti Abdullah (First Author) – contributed in writing the manuscript, collection and complication of the data needed.
2. Mr (Dr) Mohamad Azizul Fitri Khalid– contributed in original concept of the case report and critical review of the manuscript.
3. Associate Professor (Dr) Ramiza Ramza Ramli (Corresponding Author) – critical review of the manuscript and preparing for the final submission of the manuscript.

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