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Case Report

Frontal mucocele causing ophthalmoplegia and proptosis

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ABSTRACT

Mucoceles are known to occur as a result of chronic accumulation of mucoid secretions within a sinus as a result of long-standing outflow obstruction secondary to inflammation. This can lead to extension into the orbital and intracranial cavities. It can also get infected to cause mucopyocele. The frontal sinus is the most common site for mucocele. Long-standing symptoms of chronic rhinosinusitis, if ignored, can lead to such complications. Clinical features include frontal pain and swelling. It may displace the orbit inferiorly and laterally. If neglected, it can progress to cause osteomyelitis and erode the posterior wall of the sinus to cause intracranial complications. Imaging is paramount in assessing the lesion's type and extent. Computed tomography can be used to delineate the bony erosions, whereas Magnetic Resonance Imaging helps characterize the lesion's nature. Surgery remains the mainstay of treatment with the aim of draining the mucocele and removing the mucosa to prevent a recurrence. Surgical approaches can be both endoscopic and open, with the advantages and disadvantages of each.

1. Introduction

Langenbeck first described paranasal sinus mucoceles with the name of hydatides in 1820. The term "mucocele" was given by Rollet in 1909. Mucocele can be defined as an epithelial-lined mucus-containing sac.[1] It is cystic in consistency and has expansile nature. Although it in itself does not have destructive properties, it can cause bony erosion due to pressure over a prolonged period. Mucocele can have multiple etiologies, including inflammation, trauma, allergy, previous surgery, anatomical anomalies, and bony lesions such as osteoma or fibrous dysplasia. All etiologies ultimately converge, causing obstruction of the natural Ostia leading to impaired sinus drainage.

The most common site of mucocele is the frontal sinus (60-89%), followed by the ethmoid sinus (8-30%) and maxillary sinus (5%). Mucoceles have rarely been reported in the sphenoid sinus. The most common age of presentation is 40 to 60 years, with no sex predilection.[2]

As a result of obstructed drainage, continuous negative pressure builds in the sinus, leading to mucin accumulation over time. The resultant inflammation results in the recruitment of lymphocytes and fibroblasts. This results in bony remodelling and resorption.

Various theories of the pathogenesis of mucocele have been described in the literature. The theory of cystic degeneration of seromucinous glands by Batsakis et al. states that the pseudostratified columnar epithelium gets compressed to cuboidal epithelium due to pressure. This supports the theory of pressure erosion. However, the cuboidal epithelium was rarely seen in mucoceles. Lund and Milroy conducted a histopathological comparison between the epithelium lining of mucocele and healthy

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sinus mucosa. The mucocele epithelium had inflammatory infiltrates, cytokines, PGE2, collagenases and fibroblasts. These stimulate osteoblasts and lead to bony expansion and remodelling.[3]

Superadded infection in these mucoceles can lead to increased inflammatory cytokines. This further accelerates the inflammation and formation of purulence, which can lead to mucopyocele as seen in our case. Most commonly isolated organisms in culture are Staphylococcus aureus followed by alpha hemolytic streptococcus. Hemophilus, gram-negative bacilli and anaerobes such as Peptostreptococcus are also occasionally isolated.[4]

The clinical presentation largely depends on the site of origin. Frontal mucocele can present with frontal headache, asymmetry of face, frontal swelling. These are generally followed by orbital complaints, the most common being proptosis (83%) and diplopia (45%). Other orbital complaints can be reduced ocular mobility (as in our case) and reduced visual acuity. Erosion of the anterior wall can present as frontal swelling, whereas posterior wall erosion can lead to meningitis and even CSF fistula. The direction of proptosis helps in localising the lesion. Axial proptosis is seen with an orbital apex lesion. Ethmoidal lesions displace the orbit laterally, whereas frontal lesions displace the orbit laterally and inferiorly.[5,6]

Imaging with Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) is of paramount importance in differentiating mucocele from other pathologies such as neoplasms, in assessing the extent of the disease and also in determining the integrity of bony walls of orbit and frontal sinus (anterior and posterior tables). Mucoceles on CT are seen as well-defined lesions which are isodense.

2. Case Presentation

An otherwise healthy 62-year-old male patient presented with complaints of swelling above the left eye for one year, which was insidious in onset and gradually progressive. The swelling was associated with eye protrusion and double vision for the past six months. However, the patient has no symptoms of chronic rhinosinusitis, including nasal obstruction, nasal discharge, facial pain or smell abnormalities. On clinical examination, a 3 x 2 cm solitary swelling was seen above the medial canthus causing non-axial proptosis, pushing the orbit laterally, forwards and inferiorly. The swelling was firm in consistency with a smooth surface, regular border, non-tender and not fluctuant. The overlying skin was normal, and there was no local temperature rise. The extraocular movement of the left eye was restricted in the superior gaze. The pupillary reflex, the light accommodation reflex and the fundus examination were normal. The septum was deviated towards the left side; however bilateral airway patency was maintained. The nasal mucosa was normal.

A non-contrast computed tomography showed expansile soft tissue contents occupying the left frontal sinus, measuring 2.5 x 4.4 x 2.4 cm, with bony remodelling of the outer and the inner table. However, no calcifications were seen. The contents were hyperintense on T2 weighted sequence with no post-contrast enhancement. There was no evidence of orbital or intracranial extension.

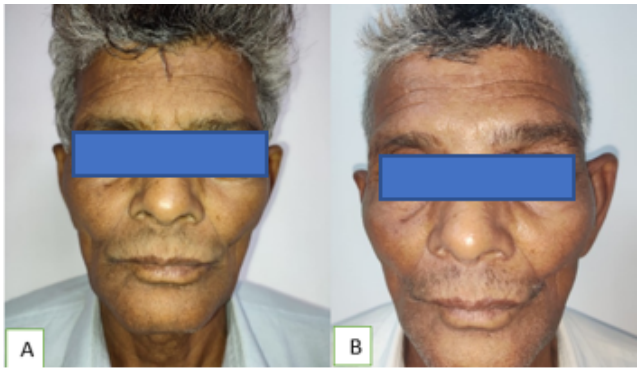

Fig. 1: a: Preoperative picture showing swelling in the left medial canthus region with proptosis of the left eye **b:** Postoperative picture showing postoperative resolution of the swelling with the left eye in normal position.

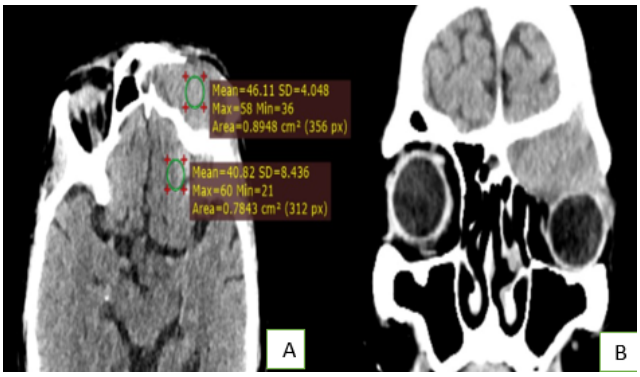

Fig. 2: a: Preoperative Non-contrast computed tomography of the nose and Paranasal sinuses (Axial view) showing expansile soft tissue contents occupying the left frontal sinus, measuring 2.5 x 4.4 x 2.4 cm, with the bony remodelling of the outer and the inner table **b:** Preoperative Non-contrast computed tomography of the nose and Paranasal sinuses (Coronal view) showing hypertense expansile lesion filling the left frontal sinus with supra-orbital extension and left eye proptosis.

A combined external and endoscopic approach was planned for the frontal sinus. Lynch Howarth incision was given midway between the medial canthus and the midline dorsum. This was extended inferiorly for 1 cm and extended superior for 1 cm, curving below the superior brow. The incision was deepened, and the periosteum was elevated to expose the frontal process of the maxilla inferiorly and the floor of the frontal sinus superiorly.

Fig. 3: a: Preoperative T2 weighted Magnetic resonance imaging of the nose and Paranasal sinuses (Axial view) showing hyperintense lesion filling the left frontal sinus with no postcontrast enhancement **b:** Preoperative contrast enhanced T1 weighted Magnetic resonance imaging of the nose and Paranasal sinuses (Axial view) showing non-enhancing lesion filling the left frontal sinus.

Fig. 4: Intraoperative imaging showing Lynch Howarth incision given midway between the medial canthus and the midline dorsum to expose the frontal process of the maxilla inferiorly and the frontal sinus floor superiorly. The frontal sinus floor was found to be dehiscent and approximately 10 mL of mucin was evacuated.

The floor of the sinus was found to be dehiscent and approximately 10 mL of mucin was evacuated. The sinus was then inspected with a 4 mm 0-degree endoscope and all the remaining walls were intact. The polypoidal mucosa of the sinus was then removed. Further, the frontal process of maxilla was drilling to exposure the frontal recess, from which purulent discharge was expressed. Left frontal sinusotomy (Draf IIa) was done using intact bulla technique after uncinectomy. Curved suction inserted trans nasally was then seen through the external incision ensuring the patency of frontal recess. Left uncinate was polypoidal and was seen to be obstructing the frontal recess. The wound was then closed in 2 layers and the left nasal cavity was packed.

Fig. 5: Intraoperative imaging showing the evaluation of the patency of the frontal recess by inserting a curved suction trans-nasally which was then seen through the external incision.

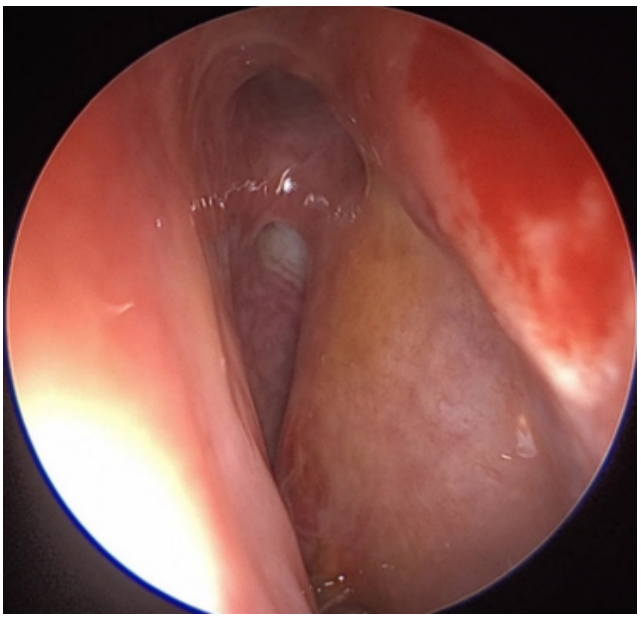

Fig. 6: 6-months postoperative Diagnostic nasal endoscopy shows a patent frontal recess with normal mucosa.

The post operative period was uneventful. Nasal pack was removed on post operative day 2 and sutures were removed on post operative day 7. Proptosis and superior gaze restriction resolved completely on 2^{nd} follow up which was 2 weeks post operatively. Vision and extra ocular movements were normal. The patient has been asymptomatic till 6 months post operatively. Nasal endoscopy shows a patent frontal recess with normal mucosa.

3. Discussion

Mucocele is diagnosed using the triad of clinical history, physical examination and radiology. The characteristic radiological findings include isodense soft tissue with regular margins and osteolysis. It is hyperintense on T2 weighted images owing to its fluid content. Protein and blood contents of the lesion appear hyperintense on T1 weighted images.

Gold standard management of mucoceles is surgery. The surgical approach to the frontal sinus is tailor made to each case depending on the size, site and the location of the mucocele. The aim of the surgery is to drain the mucocele and establish definitive ventilation of the sinus to prevent recurrence. A landmark paradigm shift has been observed over the past few decades from the traditional radical open approach to the more recent endoscopic mucosal preserving functional surgery. Age old controversy regarding the merits and demerits of stenting has had mixed opinions, with the proponents arguing on lesser chances of stenosis and the opponents claiming higher chances of stenosis and granulations secondary to stenting.

Various approached to the frontal sinus have been described in literature. The Lynch approach has disadvantage of poor cosmesis and neuralgic pain. The bicoronal osteoplastic flap offers excellent exposure to all walls of the sinus but comes at the cost of scalp numbness, frontalis palsy, longer surgical time and poor cosmesis. The transcarancular transconjuncti val approach avoids facial incision and hence has better cosmesis. But it has limited exposure of the lateral part of the sinus and involves removing lamina papyracea. The orbital fat can prolapse through this defect and obstruct the sinus outflow tract, leading to long term failure.[7]

4. Conclusion

Mucoceles are known to occur as a result of chronic accumulation of mucoid secretions within a sinus as a result of long-standing outflow obstruction secondary to inflammation. The surgical treatment plan should be individualized as per the extent of the disease. The combined in our case offered the advantage of wide exposure with limited facial scarring. The incision in our case did not involve the superior orbital rim, and hence preserved the supra orbital and supra trochlear neurovascular bundle.

5. Source of Funding

None.

6. Conflict of Interest

None.

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