PARANASAL SINUS MUCOCELE AND ITS COMPLICATIONS: RETROSPECTIVE ANALYSIS AND LITERATURE REVIEW

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ABSTRACT

BACKGROUND: Mucocele is a true cystic expansile mass filled with mucus resulting from obstruction of the sinus ostia. Prompt diagnosis and management are needed to avoid complications due to the intracranial or intra-orbital extension.

OBJECTIVES: To audit data of mucocele of the paranasal sinuses in a tertiary center, to review the literature regarding the clinical features and the management of mucocele in the paranasal sinus and to emphasize the iatrogenic causes of mucocele after the development of Endoscopic Skull Base Surgeries.

MATERIALS AND METHODS: A retrospective analysis of etiology, clinical presentation, sinuses involved, operative technique, complication, recurrence were interpreted.

RESULTS: The mean age of 16 patients managed is 49. Location:37% frontal,19% ethmoidal,19% frontoethmoidal,19% sphenoidal,6% maxillary. The iatrogenic cause was 44%. Symptoms included proptosis, external swelling, nasal block, and discharge, and vision loss. Complications were orbital abscess and skull base defect. 14 patients underwent endoscopic procedures-excision or marsupialization. One patient underwent craniotomy; one patient combined approach was performed. One patient had a recurrence.

CONCLUSION: Mucocele of the paranasal sinuses is still a rare clinical scenario and it is imperative to note that any delay in diagnosis and management can lead to expansion and erosion of the bony wall of the sinuses and causing complications.

KEYWORDS

Mucocele of Paranasal sinuses; Marsupialization; Iatrogenic causes of Mucocele; Skull base surgery; Vascularized flap

INTRODUCTION:

Wilkerson Jr had elaborately reviewed the literature on mucocele that was available before 1945. He quotes the definition of a mucocele as written by McKenzie in 1928 "the modern opinion is that a mucocele is a mucus cyst (and in turn a form of adenoma) which, undergoing active enlargement leads to distention of the bony walls of sinus and their slow atrophy". He goes on to describe the writing of Boenninghaus (1898-1899) on one of the discovered skulls at the famous chapel of Tschernbeney, this skull had a large defect in the anterior wall of the frontal sinus and the cerebral wall, which was likely to be the result of a frontal sinus mucocele¹.

Berthon (1880) has been credited as the first physician to record cases of other physicians in his thesis. Some of the reports by Berthon were taken from the Dezeirmeris that was published in 1839. Dated as early as 1725, this thesis had reports on treatment of mucocele². Prof. Langenbeck (1820) has been credited with publishing the first authoritative report of a mucocele of the frontal sinus but he mentioned it as a Hydatid Cyst. It was Rollet in 1909 who mentioned the name

Mucocele³. It is a cystic mass with mucus retention from the respiratory epithelium with a propensity to expand. Mucocele is similar to a space-occupying lesion and can present as a benign lesion with the possibility of causing serious complications due to bony erosions. Since then numerous case reports, case series, and reviews have been published in literature adding a few additional learning points to identify and manage these lesions better.

This study aims to retrospectively analyze data with regards to the demographic features, etiology, clinical presentation, complications with treatment options, and surgical practices to decrease the recurrence.

MATERIALS AND METHODS

A retrospective analysis of data of patients managed for paranasal sinus mucocele in a tertiary hospital between July 2015 and July 2019 was included in the study (TABLE 1). Well explained Informed Consent was taken before surgery in all cases. CT scan finding was also collected for all and the MRI findings for indicated patients were noted.

TABLE 1:

No	Age	Presenting Symptoms	Location	Surgery	Complication	Previous Surgery
1	39/M	Diminished vision with proptosis x 2 years	Left anterior ethmoid with orbital extension	Endoscopic marsupialization of mucocele with orbital decompression	Orbital involvement	Nil
2	57/F	Diminished vision with proptosis and nasal bleed x 1 year	Bilateral frontal mucocele	Endoscopic marsupialization	Orbital abscess	Nil
3	70/M	Headache x 3 years	Left sphenoid	Endoscopic marsupialization	Skull base defect	Nil
4	7/M	Proptosis And diminished vision x 6 months	Right ethmoid	Endoscopic marsupialization	Nil	Nil
5	29/M	Right frontal headache and pain x 4 months	Right frontal	Endoscopic marsupialization	Nil	4 times operated for recurrent mucocele
6	59/F	Bleeding from nasal cavity and headache x 3 months	Left frontal	Endoscopic Marsupialization	Nil	Pituitary macroadenoma excision
7	50/F	Headache and eye pain and proptosis x 2 months	Right frontal	Endoscopic marsupialization	Nil	CSF leak repair
8	42/M	Headache and proptosis x 1 month	Right frontal	Craniotomy and endoscopic excision	Nil	Nil
9	72/F	Nasal obstruction proptosis x 7 months	Ethmoidal right	Endoscopic marsupialization	Nil	CSF leak repair

10	59/M	External frontal swelling and head ache x 1 yea	Right fronto ethmoidal	Endoscopic marsupialization	Skull base defect	Nil
11	62/M	Headache x 1 year	Sphenoid mucocoele	Endoscopic marsupialization	Optic nerve compression	Pituitary macroadenoma excision
12	47/F	Headache x 1 year	Sphenoid	Endoscopic marsupialization	Nil	Pituitary macroadenoma excision
13	70/F	Pain and swelling in the brow area and headache x 6 month	Right frontoethmoid	Endoscopic marsupialization	Skull base defect	Nil
14	24/M	Headache X 6 months	Right frontal	Combined endoscopic excision with bicoronal approach	Nil	Nil
15	66/F	Diplopia x 3 months	Right fronto ethmoid	Endoscopic marsupialization	Nil	FESS with septoplasty
16	45/M	Pain in left cheek and hard palate x 3months	Left maxillary	Endoscopic marsupialization	Nil	Post tooth extraction

RESULTS:

In this series, cases have been seen between 7-70 with a mean age of 49 years. There were 16 patients and 9 were male patients. There were 6 frontal, 3 frontoethmoidal, 3 ethmoidal, 3 sphenoidal and 1 maxillary. The symptoms depended on the sinus involved. In the case of frontoethmoid involvement, there was proptosis due to the expansion of the orbit along with headache. It was associated with headache and external swelling. Diminished or double vision was seen in such mucocoele due to the orbital involvement and the compression of the optic nerve in case of sphenoid. In the case of maxillary involvement, there was cheek swelling and facial pain. Other associated symptoms were nasal block, discharge, and nasal bleeding. CT scan revealed an expansile homogenous well-defined mass with the remodeling of adjacent bones and Magnetic Resonance Imaging was done in patients with the intracranial or intra-orbital extension (FIG 1, 2). 14 patients underwent endoscopic procedures which included endoscopic excision or marsupialization. One patient underwent craniotomy and in one patient combined approach was performed.

FIGURE 1.

FIGURE 2.

Complications of mucocoele seen in our study were orbital abscess and skull base defect with intracranial and extradural involvement. Seven cases of mucocoele had a history of previous surgery. Three following pituitary macroadenoma excision, two following CSF leak repair, one following functional endoscopic sinus surgery, and one following tooth extraction. Average interval between surgery and incidence of mucocoele was 1-8 years. Recurrence was seen in one patient.

DISCUSSION:

Paranasal sinus mucocoeles is an expansile cystic lesion that occurs due to mucus retention^{4,5}. These mucocoeles expand causing bony

expansion, resorption and erosion⁶. They are commonly seen in the third and fourth decade of life but can be reported across all age groups. The etiopathogenesis of this lesion could be due to trauma, surgery, chronic sinusitis, and tumors^{5,7,8}.

Pediatric paranasal sinus mucocoele is rare. Di Cicco et al have reported a prevalence of 16.4% among children with symptomatic cystic fibrosis⁹. In this study, the pediatric patient didn't have any complaints of cystic fibrosis.

In this series, iatrogenic cause accounted for 43.75%. Obeso et al observed that 35% of patients had a prior history of nasal surgeries and 14% had a history of head injury¹⁰. Benkhatar et al in a observed that around 13.1% of patients who undergo FESS for chronic rhinosinusitis develop mucocoele¹¹. However, Lee T-J et al reported that there was incidence of mucocoele was 66-86% in their extensive review of 82 patients¹². Blaier et al observed that 3.6 % of the patients developed mucocoele after the reconstruction of skull base defects¹³.

Implantation of respiratory mucosa has been implicated as one of the causes for mucocoele formation after the skull base surgeries^{14,15}. Care should be taken to prevent the entrapment of the healthy mucosa near the skull base defect within the flap and also to place the flap in accurate position so as not to cause blockage of the sinus ostium. Violation of the normal mucosa can lead to adhesions and synechia formation which could obstruct the drainage of the sinus. Ablation of the 1 cm normal mucosa around the defect region before reconstruction can prevent the mucocoele formation^{13,16-18}.

Nazar et al have observed that frontal and ethmoid sinuses were involved in 63%, maxillary in 30.4%, and sphenoid sinus in 6.5%¹⁹. Frontal and ethmoid are commonly involved because the nasofrontal outflow pathway can be obstructed by edema, polyps, adhesions, and septal deviation.

Proptosis and visual disturbances were seen in frontoethmoidal mucocoele whereas patients with sphenoid mucocoele present with headache and diminished vision. In maxillary sinus mucocoele, facial swelling with pain was the main complaint. Nasal block, discharge, and bleeding can be associated symptoms. Proptosis (37.5%), headache (37.5%), diminished vision (12.5%) were the most common symptom. In one case, patient had an orbital extension and had no perception of light at presentation and diagnosed with optic neuropathy. Optic neuropathy might be due to mechanical compression, ischemia, or optic neuritis. Posterior ethmoid and Sphenoid sinus mucocoeles are commonly associated with poorer outcomes. Fasina et al reported a similar observation. Of the 12 patients with paranasal sinus mucocoele, 4 presented with diminished vision and no perception of light in two patients due to optic neuropathy⁵. Jaswal et al in their series observed that headache and facial swelling were the commonest symptoms in 81% and 75% respectively²⁰.

Carvalho et al have discussed the common and uncommon presentation of the mucocoele on a CT scan²¹. MRI PNS is done when intracranial/intraorbital extension is suspected²².

Endoscopic Marsupialization or excision with extravasation of the accumulated mucus and releasing the obstruction at the ostium is the treatment of choice. This will bring back the normal sinus mucosal clearance and prevent any further recurrence²³. In cases with far lateral extension or intracranial extension, an external approach or combined

approach is used. In our study, 14 patients underwent successful endoscopic surgery. One patient underwent excision through a craniotomy procedure. One patient with laterally extending frontal mucocoele had combined approach surgery where the osteoplastic flap was raised and combined with an endoscopic procedure to excise the mucocoele. Hence it is advised to denude the mucosa during the primary repair of the skull base defects and also while placing the graft²⁴.

The mainstay of treatment is surgery. Endoscopic marsupialization with stripping of mucosa/excision is the preferred technique. Initially, external approaches like Lynch Howarth and Caldwell-Luc procedures were used. The surgical technique is decided based on the location and involvement of surrounding structures. The endoscopic approach is preferred because it is associated with less morbidity and a low recurrence rate. With improved endoscopic surgical practices, it can be used even in complicated cases. The expansion of mucocoele can erode the bones like lamina papyracea, the floor of the frontal sinus, posterior table of the frontal sinus and can spread the infection from the sinuses into the orbit and intracranium causing serious consequences. Staphylococcus aureus, Streptococcus, and Haemophilus influenza are common organisms found associated with infection of mucocoele and its intraorbital and intracranial complications. Drainage of such abscess is done without damaging the vital soft tissue structure of the orbit. There were no complications noted in the patients treated postoperatively.

The recurrence rate is considered to be less than 10%²⁴. In our study, recurrence was seen in one case of frontal mucocoele for which the patient had undergone surgery 3 times previously. The patient underwent endoscopic excision and followed up for two years without recurrence.

CONCLUSION:

A robust approach is needed for early diagnosis and treatment thus preventing the incidence of complications of mucocoele. Iatrogenic cause of mucocoele accounts for 44% of cases in this study. A high index of suspicion for the occurrence of mucocoele in patients following endoscopic endonasal procedure with recurrent symptoms has to be considered for early diagnosis and intervention. Meticulous technique with the preservation of healthy mucosa is mandatory to prevent the formation of mucocoele. Care should be taken in the reconstruction of skull base defects to remove the surrounding mucosa before placement of the flap.

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