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# Mucocele of the Paranasal Sinuses – Retrospective Analysis of a Series of Seven Cases

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**Aim:** The present study aimed at identifying the risk factors, typical clinical symptoms and applied treatment in seven cases with mucocele of the paranasal sinuses.

**Materials and methods**: Seven patients suffering from mucocele of the paranasal sinuses were admitted to the Clinic of Neurosurgery and the Clinic of Ear, Nose and Throat Diseases between 2014 and 2016. There were 4 females and 3 males aged between 22 and 78 (95% CI [31.44, 70.23]). Initial symptoms, their duration, clinical presentation upon admission, localization of the mucocele, type of surgical intervention and outcome have all been studied.

**Results:** The localization of the mucocele was frontal (2 cases), fronto-ethmoidal (2 cases), ethmoidal (1 case) and spheno-ethmoidal (2 cases). Risk factors were identified in 4 cases. Endoscopic marsupialization of the mucocele was performed in 5 cases. One patient with intracranial extension of frontal mucocele was treated via right frontobasal craniotomy. One of the patients refused surgery.

**Conclusion:** Endoscopic marsupialization should be considered as a method of choice in cases with mucoceles without extensive intracranial invasion. This approach offers adequate drainage, balloon dilatation of the natural sinus openings that prevents future recurrence.

## BACKGROUND

Mucocele of the paranasal sinuses (MPS) is a benign, cystic and destructive formation lined by respiratory epithelium that is filled with mucous collection.[1] The disease was first described by Langenbeck in 1820 and named 'hydatid'. Rollet was the first who used the term mucocele in 1909.[2] It causes accumulation of mucous discharge and desquamated epithelium in the sinus cavity that erodes its bony walls. The close anatomical relations between the paranasal sinuses, the orbit and the brain predisposes to mucocele invasion to the orbit and intracranial space. This can result in severe neurological damage and even death if treatment is delayed.[3]

The mucocele most commonly affects the frontal sinuses and the ethmoidal air cells (55%-65%), maxillary sinus (16-24%), ethmoidal cells alone (11-14%) and the sphenoid sinus (1-7%).[1,4,5]

The etiology of MPS is not entirely understood but some factors such as inflammation, allergy, trauma, previous surgery, anatomical anomalies, fibrous dysplasia, osteomas and fibromas can predispose to mucocele development.[2] These conditions may result in obstruction of the sinus openings, thus, impeding the normal drainage of the mucous discharge.

## AIM

The present study aimed at identifying the risk factors, typical clinical symptoms and applied treatment in seven cases with mucocele of the paranasal sinuses.

## MATERIALS AND METHODS

Seven patients with MPS were admitted to the Clinic of Neurosurgery and the Clinic of Ear, Nose and Throat Diseases between 2014 and 2016. There were 4 females and 3 males aged between 22 and 78 years (mean 49.2819.52; 95% CI [31.22, 67.34]; median – 53; P > 0.10). Medical files, clinical presentation, imaging studies and operative protocols have all been studied. Data was collected regarding the initial symptoms, their duration, clinical presentation upon admission, localization of the mucocele, type of surgical intervention and outcome.

## RESULTS

St George University Hospital in Plovdiv, Bulgaria is accredited with third level of competence and harbors 1500 beds. The total number of hospital admissions for the period between 2014 and 2016 was 228 864 while the number of patients identified with MPS were only seven which constitutes regional morbidity of 0.3 per 10 000 hospitalizations.

Clinical symptoms, their duration and suggested etiology have been summarized in **Table 1**.

In all patients, headache of varying intensity was observed. Consequently, damage to the cranial nerves and orbital content developed in 3 cases over a period of time. The time from disease onset to hospital admission varied between 1 and 96 months (mean 31.42 33.25; 95% CI [0.68, 62.14]; median – 24; P > 0.10).

Regular inflammatory parameters such as white blood count, C-reactive protein and erythrocyte sedimentation rate did not indicate existent infection.

Skull x-rays detected abnormality in only one patient that included osteosclerotic reaction and bulging of the posterior wall of the frontal sinus. **(Fig. 1A)**

All patients were examined by computed tomography (CT) and/or magnetic resonance imaging (MRI). These two modalities play additive role in imaging diagnostics of the mucocele. The CT is superior with regard to detection of bone erosions, osteosclerotic changes and calcification **(Fig. 1B, Fig. 1C, Fig. 2 and Fig. 3A)**.

MRI is used to determine the duration of the mucocele and to distinguish it from neoplastic lesions that can obstruct natural sinus openings and impede drainage. MRI gives greater details regarding the expansion of the mucocele and its relation to adjacent soft tissues **(Fig. 1D, Fig. 3B,** and **Fig. 4A-C)**.

The etiology was unclear in 3 of the cases. The rest of cases had suspected etiology such as

**Table 1.** Gender, age, clinical symptoms and duration, suggested etiology

| Patient number | Gender/ age | Localization of mucocele | Clinical symptoms and duration | Suggested etiology |
|---|---|---|---|---|
| 1 | **F/60** | Frontal | Mild headache for 3 years | N/A |
| 2 | **F/53** | Ethmoidal | Intermittent headache for 2 years | Sustained skull base fracture in childhood |
| 3 | **M/29** | Frontal | Headache for 4 years | Two nasal surgeries for poliposis. Osteoma of the frontal sinus |
| 4 | **M/64** | Frontal and ethmoidal | Mild headache and left eye swelling for 8 years. Diplopia for 2 months | Surgery for mucocele before 10 years |
| 5 | **F/78** | Sphenoid and ethmoidal | Headache for 1 year proptosis, palpebral ptosis and diplopia for 7 days | N/A |
| 6 | **M/31** | Frontal and ethmoidal | Exophtalmus and diplopia for 30 days | Surgery for nasal poliposis. |
| 7 | **F/30** | Sphenoidal and ethmoidal | Intensive, drug-resistant headache for 3 months | N/A |

F-female; M-male; N/A – not available

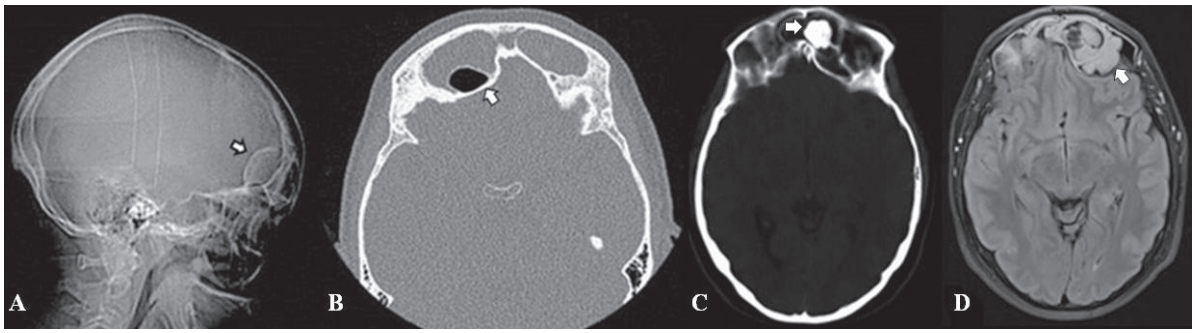


**Figure 1. (A)** and **(B)**: Frontal mucocele in a patient suffering from headache for 3 years - deformation and bulging of the posterior wall of the frontal sinus detected on plain x-ray and CT scan (white arrows); frontal mucocele in a patient suffering from headache for 4 years - **(C)** presence of osteoma obstructing the opening of the frontal sinus; **(D)** T2 FLAIR MRI: high signal intensity suggestive of long duration of the frontal mucocele.

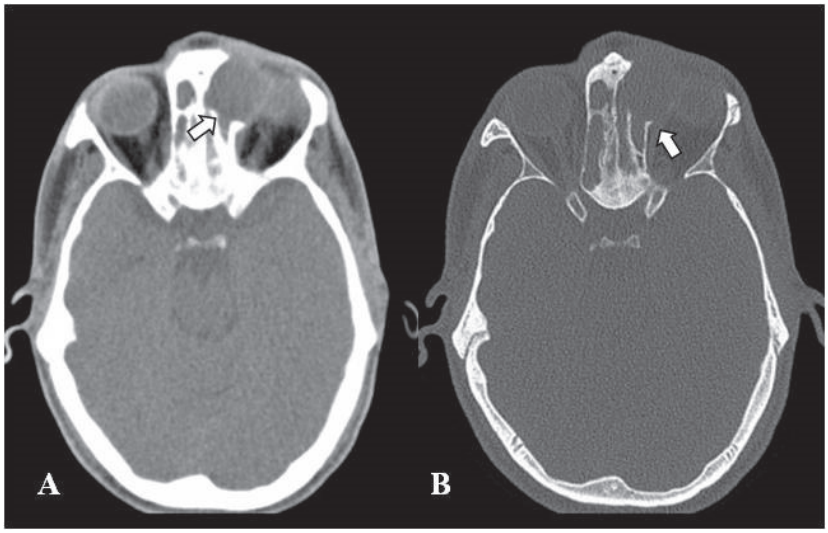


**Figure 2.** Fronto-ethmoidal mucocele in a patient with proptosis and diplopia. (**A)** CT – heterodense lesion eroding the anterior ethmoidal cells and the floor of the frontal sinus entering the orbit from medio-cranial direction (white arrows).

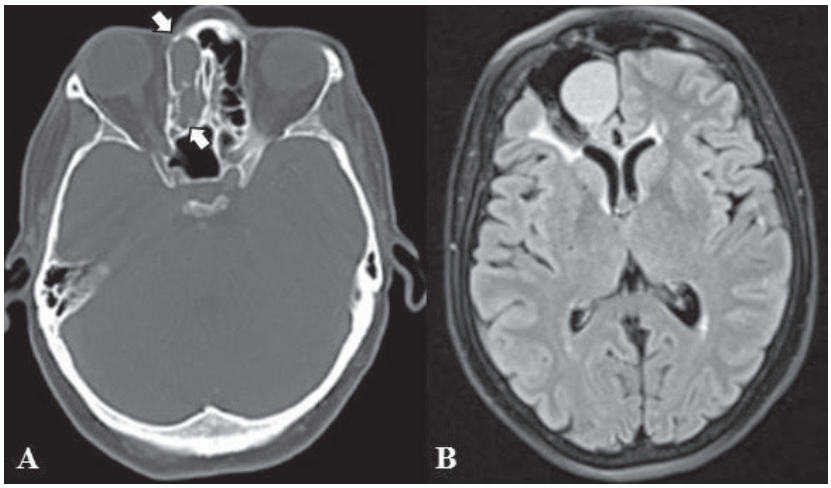


**Figure 3.** Mucocele affecting the anterior and posterior ethmoidal cells in a patient suffering from headache for 2 years. **(A)** CT (bone window) – obscurity of the right ethmoidal cells (white arrow); **(B)** T2 FLAIR MRI indicating high signal intensity lesion that invades the intracranial space and compresses the right frontal lobe.



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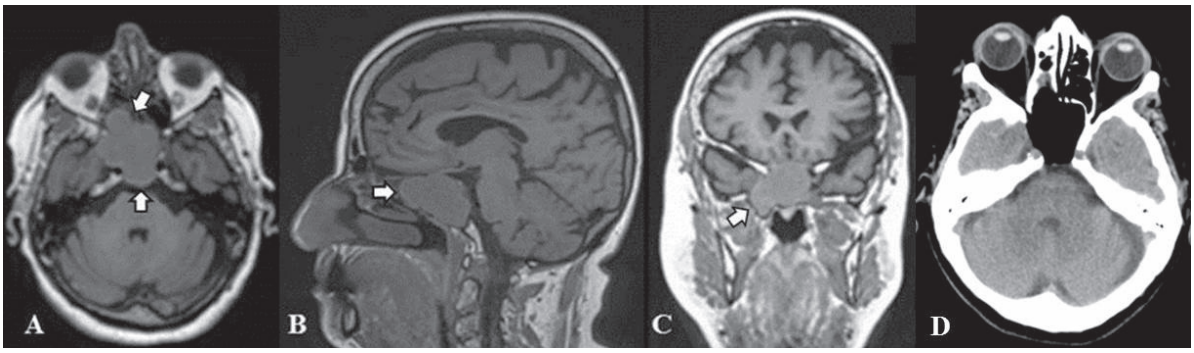


**Figure 4.** Female patient suffering from mucocele affecting the sphenoid sinus and posterior ethmoidal cells who presented with proptosis and right oculomotor nerve palsy. T1 MRI images in axial, sagittal and coronal planes **(A-C)** – isointense lesion eroding the bone of the sphenoid sinus and posterior ethmoidal cells that propagates to the intracranial space and compresses the right cavernous sinus; **(D)** Postop CT demonstrating efficient drainage of the mucocele following endoscopic marsupialization.

previous skull base fracture, massive polyposis, osteoma, preceding surgery for polyposis.

All patient were offered to sign an informed consent prior to surgery but one of them refused and was not operated. Endoscopic marsupialization and drainage was applied in 5 patients (Patients No 3 to No 7). The extensive intracranial propagation in patient No 2 necessitated frontobasal craniotomy and navigated total resection of the mucocele followed by plastic closure of the ethmoidal bone defect.

The postoperative period was uneventful in all operated cases. The neurological deficit, where present, resolved completely in the follow-up period. Postop imaging was used to confirm mucocele resections **(Fig. 4D)**.

## DISCUSSION

The incidence of MPS varies from 1.5 to 4 per year dependent on the level of competence of the medical institution.[4,6,7] This is in contrast to the incidence reported in our study.

The mucocele has similar distribution between sexes and occurs most often between 20 and 40 years with peak incidence in the 4$^{th}$ decade that coincides with our observations.[3,4,8] According to Devars du Mayne et al., there is slight preponderance to the male gender (3:2) but here we report the opposite.[7]

The disease etiology is not entirely clarified. There are various theories that attempt on explaining the origin of MPS. However, recently, there has been a consensus that obstruction of bone openings of sinuses caused by trauma, neoplasms, surgery, allergy, fibrous dysplasia, etc. can lead to the development of mucocele.[2,4] We found similar etiology in 4 (67%) of our patients that included previous surgery, inflammation, skull base fracture and presence of osteoma. The rest 3 cases (48.8%) had unclear etiology.

The clinical presentation of MPS is not specific and depends on its localization. Clinical symptoms have insidious onset and tend to exacerbate over time. This is the reason why the disease can be accidentally diagnosed after imaging for other conditions. The duration between the occurrence of the debut symptoms and the final diagnosis varies between 3 days to 38 years, averaging 4 years.[9]

Patients suffering from frontal and fronto-ethmoidal mucocele initially complain of frontal headache that may be complicated by facial asymmetry and ophthalmic symptoms such as decreased visual acuity, proptosis (83%), diplopia (45%).[2,10]

The clinical presentation of the sphenoidal mucocele depends on the direction of its propagation and the relations with the surrounding anatomical structures. The most common symptom is retrobulbar headache (70%-80%) which results from the mechanical compression on the dura mater that covers the region of planum sphenoidale, cavernous sinus and the skull base as well as some inflammatory factors such as cytokines (TNF alpha, IL-6, IL-1, PGE2) that modulate the pain threshold and stimulate trigeminal nerve receptors.[11,12]

Ophthalmic involvement constitutes the second most common group of symptoms. Decrease in visual acuity is common.[13] Ophthalmic involvement is present in 30-50% of the cases. The third nerve is affected in 70% of the cases in contrast to the fourth and sixth cranial nerves.[11] The clinical presentation in our series confirm the literature data.

All cases of MPS demonstrate similar findings at CT examination. The fluid collection of the mucocele is demonstrated as homogenous isodense substance that varies between 9HU and 41HU. Long-lasting mucoceles has higher density over 20 HU whereas acute disease has a density below 20 HU.[14] Contrast application can enhance the capsule of the lesion. CT bone window can show bone erosions in axial, sagittal and coronal planes. It is important to differentiate between mucocele and retentional mucous cyst which is a cystic formation with fluid collection that do not destruct surrounding bones and is often asymptomatic.[15] CT is extremely important for proper preoperative planning.

MRI demonstrates well delineated expansible lesion that differs in signal on T1 and T2 sequences. Acute mucocele demonstrates high signal intensity on T2 and medium to low signal intensity on T1 because they have high water content. Chronic lesions has high to medium signal intensity on T1 and T2 that is based on the higher protein content and dehydration of mucocele collection.[16] Contrast enhancement is usually observed at the periphery of the lesion. MRI is a valuable tool in presenting underlying etiology such as neoplastic formations. It provides superior details regarding the propagation of the mucocele and its relations with surrounding anatomical structures compared to the CT scanning.

The surgery for MPS aims at removal of the lesion and adequate sinus drainage that prevents from future recurrence.[2] The selection of appropriate surgical approach depends on the localization, size and direction of propagation of the mucocele. Cases with superimposed infection should be covered by antibiotics.

Currently, the endoscopic surgery is considered to be a method of choice. Conventional radical procedures are being replaced by minimally invasive functional procedures as a result of the recent development of endoscopic surgery of the paranasal sinuses that preserves anatomy and offer superior cosmetic results.[17]

There are relative contraindications to endoscopic surgery such as severely altered postoperative anatomy that impede sufficient visualization and extreme propagation of the mucocele to the intracranial space.[18] In such cases, various surgical approaches can be used. We performed craniotomy and total microsurgical removal of the mucocele with plastic restoration of the destructed bone in patient №2. Even in such cases, some authors advocate wide endoscopic marsupialization of the cyst.[17]

The endonasal endoscopic approach to the mucocele provides efficient drainage and marsupialization. It is a safe procedure that offers favorable clinical outcome with low rate of recurrence (0.9–9.7%).[6,8,11,19,20] According to Dhepnorrarat et al., the period of postoperative follow-up in patients who had endoscopic surgery should be long - at least 6 to 7 years.[6]

## CONCLUSIONS

The MPS can remain asymptomatic for a long period of time. Its propagation to the orbit and/or intracranial space can cause neurologic deficit. The disease is benign and treatable but the early diagnosis is of extreme importance. CT and MRI are the imaging methods of choice that determine the selection of appropriate surgical approach. Endonasal endoscopic surgery is a safe treatment method with excellent results. Continuous follow-up period is considered to be economic and preventive way to secure good quality of life to patients who suffer from MPS.

## REFERENCES


1. Adouly T, Adnane C, Loufad FZ, et al. A rare cause of diminished vision: large spheno-ethmoidal mucocele Otorhinolaryngol Head Neck Surg 2016;1(2):51-2.
2. Aggarwal SA, Bhavana K, Keshri A, et al. Frontal sinus mucocele with orbital complications: Management by varied surgical approaches. Asian J Neurosurg 2012;7(3):135-40.
3. Shah NL, Upasham PS. A pathologist's perspective on mucocele of frontal sinus – a case report. Int J Int Med Res 2015;2(4):40-2.
4. Essaid O. Mucoceles fronto-orbitaires. A propos de 12 cas et revue de la litterarure. These № 279, Rabat 2008; pp 250.
5. Martel-Martín M, Gras-Cabrerizo JR, Bothe-González C, et al. Clinical analysis and surgical results of 58 paranasal sinus mucoceles. Acta Otorrinolaringol Esp 2015;66(2):92-7.
6. Dhepnorrarat RC, Subramaniam S, Sethi DS. Endoscopic surgery for fronto-ethmoidal mucoceles: a 15-year experience. Otolaryngol Head Neck Surg 2012;147(2):345-50.
7. Devars du Mayne M, Moya-Plana A, Malinvaud D, et al. Sinus mucocele: Natural history and long-term recurrence rate. European Annals of Otorhinolaryngology, Head and Neck diseases 2012;129:125-30.
8. Obeso S, Llorente JL, Rodrigo JP, et al. Paranasal sinuses mucoceles: our experience in 72 patients. Acta Otorrinolaringol Esp 2009;60:332-9.

9. Arkha Y, Benazzou S, El Ouahabi A, et al. Mucocèle géante sphénoïdale: A propos d'un cas. Fr ORL 2007; 92:312-4.
10. Tan CS, Yong VK, Yip LW, et al. An unusual presentation of a giant frontal sinus mucocele manifesting with a subcutaneous forehead mass. Ann Acad Med Singapore 2005;34:397-8.
11. Lee JC, Park SK, Jang DK, et al. Isolated sphenoid sinus mucocele presenting as third nerve palsy. J Korean Neurosurg Soc 2010;48:360-2.
12. Kariya S, Okano M, Hattori H, et al. Expression of IL-12 and T helper cell 1 cytokines in the fluid of paranasal sinus mucoceles. Am J Otolaryngol 2007; 28:83-6.
13. Gupta AK, Menon V, Sharma P, et al. A sphenoid sinus mucocele simulating as retro bulbar optic neuritis. Indian Journal of Ophthalmology 2012;60:(3):216-8.
14. Tsitouridis I, Michaelides M, Bintoudi A, et al. Frontoethmoidal mucoceles: CT and MRI evaluation. Neuroradiol J 2007;20:586-96.
15. Galiè M, Mandrioli S, Tieghi R, et al. Giant mucocele of the frontal sinus. J Craniofac Surg 2005;16:933-5.
16. Nath HD, Barua KK. A frontal retrobulbar mucocele with lytic lesion at orbital plate and orbital ridge, rare case report. Bangladesh Journal of Neuroscience 2011;27 (2):105-7.
17. Chew YK, Noorizan Y, Khir A, et al. Frontal mucocoele secondary to nasal polyposis: an unusual complication. Singapore Med J 2009;50:374-5.
18. Bockmühl U, Kratzsch B, Benda K, et al. Surgery for paranasal sinus mucocoeles: Efficacy of endonasal micro-endoscopic management and long-term results of 185 patients. Rhinology 2006;44:62-7.
19. Har-EI G. Endoscopic management of 108 sinus mucoceles. Laryngoscope 2000;111:2131-4.
20. Facon F, Nicollas R, Paris J, et al. La chirurgie des mucocèles sinusiennes: notre expérience à propos de 52 cas suivis à moyen terme. Rev LaryngoL Otol Rhinol 2008;129 (3):167-73.

# Мукоцеле параназальных синусов - ретроспективный анализ серии из семи случаев

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**Цель:** Целью настоящего исследования является установление факторов риска, типичных клинических симптомов и назначенного лечения в семи случаях мукоцеле параназальных синусов.

**Материалы и методы:** Семеро больных, страдающих мукоцеле параназальных синусов, поступило на лечение в клинику оториноларингологии в период между 2014 и 2016 гг. Среди них четыре женщины и трое мужчин в возрасте между 22 и 78 годами. [CI95% - 31.44 – 70.23]. Были исследованы первоначальные симптомы, их продолжительность, клиническая картина при поступлении, расположение мукоцеле, вид хирургического вмешательства и исход.

**Результаты:** Были установлены следующие случаи локализации мукоцеле: лобная (2 случая), центрально-этмоидальная (2 случая), этмоидальная (1 случай) и сфено-этмоидальная (2 случая). Были установлены факторы риска в 4 случаях. Эндоскопическая марсупиализация мукоцеле была проведена в 5 из случаев. Больному с интракраниальным ростом лобного мукоцеле была проведена фронтальная базальная краниотомия. Один из больных не согласился на операцию.

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**Заключение:** Эндоскопическая марсупиализация должна стать основным методом при случаях мукоцеле без экстенсивной интракраниальной инвазивной процедуры. Данный подход предлагает адекватный дренаж, баллонное расширение естественный отверстий синусов, что в свою очередь предотвращает дальнейший рецидив.