Frontal Sinus Mucopyocele Presenting as a Painful Subcutaneous Forehead Mass in a Patient with Chronic Sinusitis

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ABSTRACT

Mucoceles of the paranasal sinuses are benign, expansile lesions that develop secondary to sinus ostia obstruction. Presenting signs and symptoms vary widely but frequently include frontal headache and swelling, as well as visual changes and globe displacement depending on orbital involvement in the case of frontal sinus mucoceles. Given the potential for orbital or intracranial involvement, urgent imaging with computed tomography (CT) is important for patients with symptoms concerning for a frontal sinus mucocele. Definitive treatment is surgical. In this article, we report a case of a 50-year-old male who presented to a primary care clinic with a painful forehead mass, found to have a frontal sinus mucocele with erosion through the frontal bone that was eventually treated surgically. We also summarize presenting signs and symptoms of frontal mucoceles reported in the literature as these are important for clinicians to be familiar with, considering the potential emergent complications.

KEYWORDS: Mucocele, frontal sinusitis, endoscopic sinus surgery, Pott Puffy Tumor

BACKGROUND

Mucoceles are accumulations of mucous and epithelial cells that expand over time as a result of sinus drainage obstruction.[1] These have been recognized from as early as 1524 and first documented medically by Dezeimeris in 1725 after traumatic injury.[2] There are multiple causes of the sinus ostium obstruction that ultimately leads to mucocele formation including trauma, prior surgery, chronic sinusitis, and idiopathic causes.[1,3] Mucoceles form most commonly in the frontal sinus but can still occur in the ethmoid and maxillary sinuses.[3,4] Presentation can vary depending on the presence of extension from the normal sinus boundaries. These can occur anteriorly presenting with forehead swelling, anterolaterally into the orbit, or posteriorly into the intracranial space. These can present with proptosis, vision changes, periorbital swelling, headache, ulceration with drainage, and more. If the mucocele becomes infected, it is termed a mucopyocele. In all directions mucoceles have the potential to erode through bony structures which is likely related to the pressure combined with release of cytokines and other remodeling factors.[5,6] It is reported that orbital involvement is more common than forehead involvement due to the thinner bone surrounding the orbits.[1]

Mucoceles are important to recognize and diagnose due to the potential dangers related to vision and intracranial involvement. Intracranial extension can cause epidural abscesses and meningitis which can become neurosurgical emergencies if not appropriately treated.[7,8]

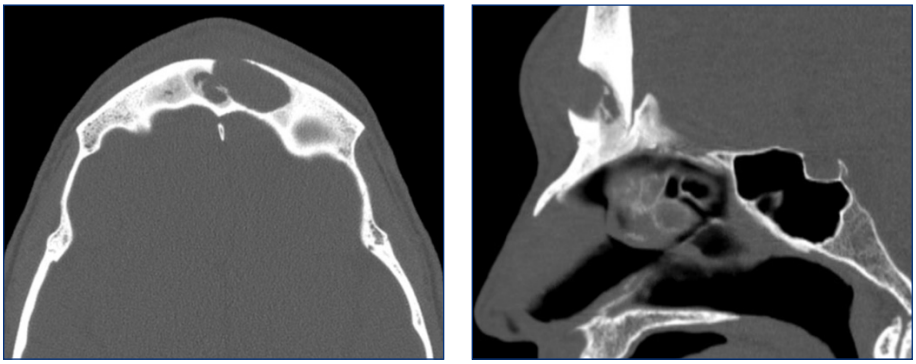
There are reports of both frontal sinus mucoceles presenting with forehead swelling as well as giant frontal sinus mucoceles presenting this way. The non-giant frontal sinus mucoceles are less common in our literature review with 19 cases[3,7,9-13] but the definition for 'giant mucocele' is not well defined.[14] These cases come from broad literature in otolaryngology, oral and maxillofacial surgery, dermatology, primary care, neurosurgery, and more, making it important to be aware of in diverse clinical settings. We describe a case of a 50-year-old male who presented with frontal swelling and headache, initially concerning for Pott's puffy tumor but later confirmed to be a frontal sinus mucocele eroding through the anterior table of frontal sinus.

CASE PRESENTATION

A 50-year-old male patient with chronic sinusitis initially presented to an outpatient primary care clinic with fluctuant central forehead swelling, headache, bilateral periorbital ecchymoses, bilateral periorbital swelling, and four days of intermittent, mild epistaxis. He had no proptosis, diplopia, or vision changes. The patient reported several years of sinus congestion and intermittent forehead pain. For four months, he had noticed pea-sized frontal swelling and chronic epistaxis that suddenly worsened. He was started on a 10-day course of twice daily oral amoxicillin-clavulanate and an urgent CT scan was obtained to rule out intracranial or intraorbital extension.

Non-contrast CT showed bilateral frontal sinusitis with erosion through the anterior frontal sinus wall without posterior frontal sinus wall erosion or evidence of epidural disease, as well as chronic sinusitis of the anterior ethmoid air cells and left maxillary sinus with left maxillary endodontal disease (**Figure 1A,B**). Otolaryngology was consulted and a needle aspiration of the frontal sinus was performed and

Figure 1A,B. CT scan from initial presentation
[**A**] Frontal sinusitis with erosion through anterior frontal sinus wall; axial view, bone window
[**B**] Frontal sinusitis with erosion through anterior frontal sinus wall; sagittal view, bone window



3cc of old blood and purulence were drained. Culture results at two days showed moderate mixed bacterial morphotypes suggestive of normal upper respiratory flora. At this visit, which was one day after his initial presentation, the patient noted increased frontal swelling and left periorbital swelling compared to the prior day. A working diagnosis of mucopyocele was made.

At the initial visit with otolaryngology, intranasal oxymetazoline was added to his regimen and left frontal sinus drill-out and drainage was tentatively scheduled to take place within one week. Three days after this visit, the patient reported marked improvement in the frontal swelling, erythema, and periorbital edema with antibiotics. A plan was made to delay surgery for one month to avoid operating during an active infection.

The patient canceled surgery and did not follow up for six months, by which time his frontal swelling and headache had gradually returned. Another 10-day course of twice daily amoxicillin-clavulanate was started but discontinued after eight days due to a delayed hypersensitivity reaction, although his frontal swelling once again improved. Surgery was scheduled for two months following this visit and repeat non-contrast CT sinus showed that the left frontal sinus mucocele had eroded past midline into the right frontal sinus through a stable defect in the left paramedian frontal sinus wall. There was decreased extension of inflammation to the prefrontal soft tissues compared to prior. It also showed increased frontal sinus hyperostosis, near complete opacification of the frontal sinuses, and stable chronic left maxillary sinusitis (**Figure 2A,B,C**).

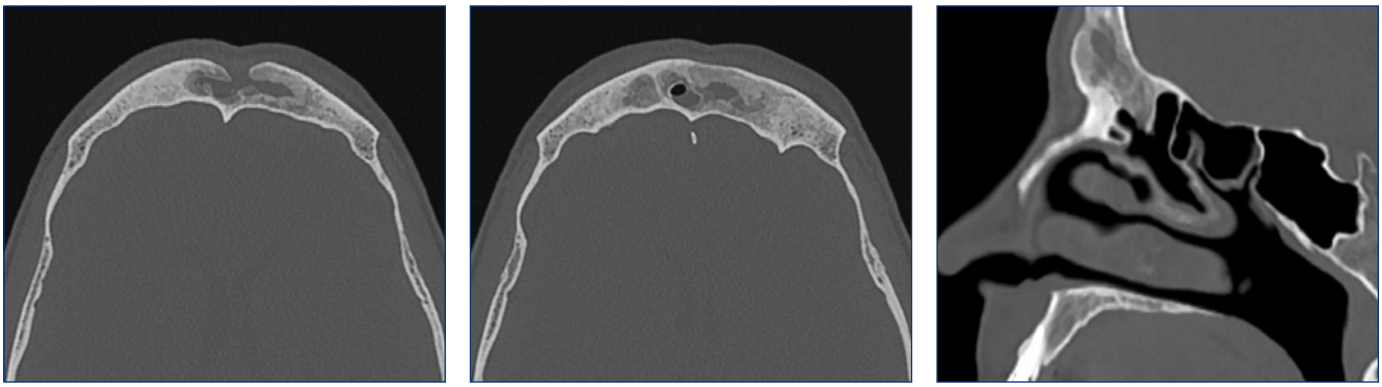
Endoscopic sinus surgery with left frontal sinus trephination was completed 10 months after the initial presentation. Image-guided access was obtained to the area of disease originating in the left frontal sinus noted on CT. There was periosteal scarring in the anterior table defect made by the mucocele. The defect was found to be 2mm in diameter with diseased mucosa. There was no frank purulence. The surgeon confirmed that the mucocele in the left frontal sinus was connected to the right frontal recess.

The patient was discharged same day and began nasal saline rinses. At one week follow-up, the patient was doing well without swelling or pain, but with some numbness at the top of his forehead. There was minimal bleeding and drainage from the nose.

DISCUSSION

We present a patient with forehead swelling due to a frontal sinus mucopyocele who was initially treated with oral antibiotics and later underwent endoscopic sinus surgery with frontal sinus trephination. While the patient's initial presentation could have represented a broad array of underlying

Figure 2. Follow-up non-contrast CT 7.5 months after initial imaging showing left frontal sinus mucocele erosion past midline into right frontal sinus through defect in left paramedian frontal sinus wall as well as increased frontal hyperostosis and near complete opacification of the frontal sinuses.
[**A**] Axial view, bone window
[**B**] Axial view, bone window
[**C**] Sagittal view, bone window



diseases including mucus retention cyst, epidermal inclusion cyst, arachnoid cyst, lipoma, fungal sinus infection, and neoplasm, it appears that our patient had a mucopyocele.[15] This patient's history of chronic frontal sinusitis, presenting symptoms, physical examination significant for fluctuant forehead swelling, radiopaque appearance of the frontal sinus on CT, erosion through bone, culture with mixed upper respiratory flora, and initial improvement with antibiotics then later with definitive surgical treatment, are all consistent with diagnosis of mucopyocele.

Mucopyoceles develop from secondary bacterial infection of mucoceles, and have been shown to involve predominantly oropharyngeal anaerobes including *Peptostreptococcus* sp., *Prevotella* sp., *Porphyromonas* sp., and *Fusobacterium* sp. Impaired drainage likely reduces oxygen tension and pH of the obstructed sinus creating a favorable environment for these microbes. Aerobic bacteria, most commonly *S. Aureus* and alpha-hemolytic streptococci have also been isolated in mucopyocele specimens. B-lactamase producing bacteria are common amongst both the anaerobes and aerobes isolated, which is important in guiding antibiotic selection. Reasonable options include coverage for *S. aureus* and anaerobes with an agent such as a penicillin with a B-lactamase inhibitor or clindamycin plus coverage for aerobic gram-negative rods with an agent such as an aminoglycoside, ceftazidime, a quinolone, or a carbapenem.[16] Risk factors for development of mucopyoceles include trauma, prior sinus surgery, chronic sinusitis, neoplasms, allergic reactions, nasal polyps and skeletal abnormalities including osteomas, fibrous dysplasia, and ossifying fibromas. They occur most commonly between the fourth and seventh decades of life.[11,17]

Bosmans and Vanhoenacker proposed 5cm as a cutoff value for "giant" mucoceles, and we use that value here in characterizing this patient's lesion as non-giant, and in describing presenting signs and symptoms of other non-giant mucoceles in **Table 1**.[14] The most common features were forehead swelling, proptosis, and nasal obstruction. While our patient presented with frontal swelling, he also experienced headache, periorbital swelling, periorbital ecchymoses, and epistaxis, the former two of which have been reported accompanying both giant and non-giant mucoceles, as shown in **Table 1**.

Definitive treatment for frontal sinus mucocele is surgical.[1] There are endoscopic, combined, and open options. Some authors suggest that endoscopic approaches be avoided in cases with intracranial involvement to avoid leaving behind residual mucous secreting tissue that will cause recurrence.[18] In all cases removal of the secretory tissue and separation from the intracranial space is key. The nasofrontal duct should either be preserved sometimes with stenting or completely closed with prior removal of all upstream secretory tissue. In a retrospective study of mucocele treatment results, recurrence after surgical intervention was 23.4%, occurring after a mean interval of four years, with a follow-up range from four months to 16 years.[19] Consistent follow-up with repeat imaging should be conducted if possible. Some authors recommend surveillance with MRI at one, two, and five years postoperatively, or immediately if symptoms were to recur.[20]

Table 1. Presenting Symptoms and Signs in Frontal Sinus and Frontoethmoidal Mucoceles

Presenting Symptom/Sign	Total N (%); N =379	Giant Mucoceles N (%); N =360	Non-Giant Mucoceles N (%); N =19
Frontal pain/ headache	204 (52.8)	202 (56.1)	2 (10.5)
Forehead swelling	132 (34.8)	123 (34.2)	9 (47.4)
Globe Displacement	101 (26.6)	100 (27.8)	1 (5.26)
Diplopia	80 (21.1)	80 (22.2)	0 (0.000)
Proptosis	44 (11.6)	36 (10.0)	8 (42.1)
Nasal obstruction	36 (9.50)	31 (8.61)	5 (26.3)
Vision loss	34 (8.97)	33 (9.17)	1 (5.26)
Nasal discharge	20 (5.28)	20 (5.56)	0 (0.000)
Eyelid swelling	20 (5.28)	18 (5.00)	2 (10.5)
Orbital/periorbital swelling	12 (3.17)	11 (3.06)	1 (5.26)
Post-nasal drip	10 (2.64)	10 (2.78)	0 (0.000)
Ptosis	6 (1.58)	6 (1.67)	0 (0.000)
Anosmia	4 (1.06)	4 (1.11)	0 (0.000)
Orbital cellulitis	3 (0.792)	1 (0.278)	2 (10.5)
Ophthalmoplegia	3 (0.792)	3 (0.833)	0 (0.000)
Epiphora	3 (0.792)	3 (0.833)	0 (0.000)
Frontal drainage	3 (0.792)	2 (0.556)	1 (5.26)
Ophthalmalgia	3 (0.792)	3 (0.833)	0 (0.000)

379 cases were reviewed from 39 studies. Presenting signs and symptoms were tabulated. Other signs and symptoms occurring less frequently included acute-onset paresis and urinary incontinence, fever, nausea, vomiting, dizziness, fistula formation, ulceration, subcutaneous air, conjunctival injection, and perioral numbness.[1-4,6-7,10-14,18-45]

Urgent evaluation of new or worsening forehead swelling is warranted given the possibility of intracranial or orbital complications. Imaging is needed prior to incision and drainage of the forehead mass as extension intracranially to the frontal lobe is possible. Sustained elevated pressure from mucus secretion and the presence of osteolytic mediators can cause erosion of the posterior table of the frontal sinus, allowing for extension into the anterior cranial fossa.[20] This case supports surgical intervention as definitive management, as this patient's mucopyocele that initially responded well to oral antibiotics alone did recur after six months. As this case highlights, urgent imaging is crucial when frontal swelling is encountered in the primary care setting, as intracranial complications could be avoided or mitigated.

References

1. Carmichael RA, Kang DR. Frontal sinus mucopyocele presenting as a subcutaneous forehead mass. J Oral Maxillofac Surg. 2015; 73:2155-2161.
2. Bordley JE, Bosley WR. Mucoceles of the frontal sinus: Causes and treatment. Ann Otol Rhinol Laryngol, 1973; 82:696-702.
3. Connolly M, Siddique I, Nusrath M. A frontal swelling with a cautionary tale. J Ir Dent Assoc. 2015; 61:138-140.
4. Natvig K, Larsen TE. Mucocele of the paranasal sinuses: A retrospective clinical and histological study. J Laryngol Otol. 1978; 92:1075-1082.
5. Fenton WH, Donald PJ, Carlton W. The pressure exerted by mucoceles in the frontal sinus: An experimental study in the cat. Arch Otolaryngol Head Neck Surg. 1990; 116:836-840.
6. Pemberton JD, Behrens A, Salter M. Frontal sinus mucocele manifests as persistent draining abscess of upper eyelid. J Ark Med Soc. 2015; 112:82-83.
7. Rosenbaum LE, Migden MR. A tender subcutaneous fluctuant forehead nodule: An unexpected foray into the cranium. Dermatol Surg. 2018; 44:1224-1227.
8. Younis RT, Lazar RH, Anand VK. Intracranial complications of sinusitis: A 15-year review of 39 cases. Ear Nose Throat J. 2002; 81:636-638, 640-642, 644.
9. Tan CSH, Yong VKY, Yip LW, Amrith S. An unusual presentation of a giant frontal sinus mucocele manifesting with a subcutaneous forehead mass. Ann Acad Med Singapore. 2005; 34: 397-398.
10. Akiyama M, Inamoto N, Hashigucci K. Frontal mucocele presenting as a subcutaneous tumor on the forehead. Dermatology. 1999; 199:263-264.
11. Herndon M, McMains KC, Kountakis SE. Presentation and management of extensive fronto-orbital-ethmoid mucoceles. Am J Otol. 2007; 28:145-147.
12. Altıntaş Kakşi S, Kakşi M, Balevi A, Özdemir M, Çakır A. Unusual case of frontal mucocele presenting with forehead ulcer. Dermatol Online J. 2014; 20.
13. Weiss JA. Mucocele of the frontal sinus. Arch Otolaryngol. 1940; 32:1090-1097.
14. Bosmans F, Vanhoenacker F. Giant frontal paranasal mucocele: Case report and review of the literature. J Belg Soc Radiol. 2020; 104.
15. Panda NK, Reddy CEE. Primary frontal sinus aspergillosis: an uncommon occurrence. Mycoses. 2005; 48:235-237.
16. Brook I, Frazier EH. The microbiology of mucopyocele. Laryngoscope. 2001; 111:1771-1773.
17. Bijith EV, Mathew S, Mahadevan K. Frontal Mucocele Mimicking a Frontal Subcutaneous Tumor. Asian J Neurosurg. 2017; 12:760-762.
18. Severino R, Severino P. Fronto-orbital mucocele with intracranial extension: a case report. J Surg Case Rep. 2017; 2017.
19. Devars du Mayne M, Moya-Plana A, Malinvaud D, Laccourreye O, Bonfils P. Sinus mucocele: Natural history and long-term recurrence rate. Eur Ann Otorhinolaryngol Head Neck Dis. 2012; 129:125-130.
20. Constantinidis J, Steinhart H, Schwerdtfeger K, Zenk J, Iro H. Therapy of invasive mucoceles of the frontal sinus. Rhinology. 2001; 39:33-38.
21. Tatsumi S, Ri M, Higashi N, Wakayama N, Matsune S, Tosa M. Pott's puffy tumor in an adult: A case report and review of literature. J Nippon Med Sch. 2016; 83:211-214.
22. Akiyama K, Karaki M, Mori N. Evaluation of adult pott's puffy tumor: Our five cases and 27 literature cases. Laryngoscope. 2012; 122:2382-2388.
23. Skomro R, McClean KL. Frontal osteomyelitis (Pott's puffy tumour) associated with Pasteurella multocida–A case report and review of the literature. Can J Infect Dis. 1998; 9:115-121.
24. Aggarwal SK, Keshri A, Rajkumar. Sinonasal undifferentiated carcinoma presenting as recurrent fronto-ethmoidal pyomucocele. Natl J Maxillofac Surg. 2012; 3:55-58.
25. Alshoabi S, Gameraddin M. Giant frontal mucocele presenting with displacement of the eye globe. Radiol Case Rep. 2018; 13:627-630.
26. Borkar S, Tripathi AK, Satyarthee G, Sharma BS, Mahapatra AK. Frontal mucocele presenting with forehead subcutaneous mass: an unusual presentation. Turk Neurosurg, 2008; 18:200-203.
27. Smith GW, Chavez M. Giant mucocele of the frontal sinus. J Neurol Neurosurg Psychiatry. 1960; 23:179-181.
28. Galiè M, Mandrioli S, Tieghi R, Clauser L. Giant mucocele of the frontal sinus. J Craniofac Surg. 2005; 16:933-935.
29. Delfini R, Missori P, Iannetti G, Ciappetta P, Cantore G. Mucoceles of the paranasal sinuses with intracranial and intraorbital extension: Report of 28 cases. Neurosurgery. 1993; 32:901-906.
30. Arole FO, Olaitan AA, Fatusi OA. Giant frontal sinus mucocoele with intracranial extension and orbital displacement in an elderly Nigerian. West Afr J Med, 2002; 21:262-264.
31. Koifman ACB, Ferraz L de O, Blanco BT, et al. Frontal sinus pneumocele: case report and literature review. Radiol Bras. 2013; 46:259-260.
32. Cagigal BP, Lezcano JB, Blanco RF, et al. Frontal sinus mucocele with intracranial and intraorbital extension. Med Oral Patol Oral Cir Bucal. 2006; 4.
33. Ada S, Yalamanchili M, Kambhampati G. Frontal sinus mucopyocele. Arch Intern Med. 2002; 62:2487-8.
34. Koike Y, Tokoro K, Chiba Y, Suzuki SI, Murai M, Ito H. Intracranial extension of paranasal sinus mucocele: two case reports. Surg Neurol. 1996; 45:44-8.
35. Singh M, Sen K, Shankar G, Mohanty SS. Imaging of a Giant Frontal Sinus Mucocele with Orbital Displacement: A Case Report. Int J Contemp Med Surg Radiol. 2019; 4:117-120.
36. Kawaguchi S, Sakaki T, Okuno S, Ida Y, Nishi N. Giant frontal mucocele extending into the anterior cranial fossa. J Clin Neurosci. 2002; 9:86-89.
37. Fernandes R, Pirgousis P. Longstanding giant frontal sinus mucocele resulting in extra-axial proptosis orbital displacement and loss of vision. J Oral Maxillofac Surg. 2010; 68:3051-3053.
38. Russell JL. Massive frontal pyomucocele: a case report. Dentomaxillofac Radiol. 1989; 18:185-187.
39. Suri A, Mahapatra AK, Gaikwad S, Sarkar C. Giant mucoceles of the frontal sinus: a series and review. J Clin Neurosci. 2004; 11:214-218.
40. Aggarwal SK, Bhavana K, Keshri A, Kumar R, Srivastava A. Frontal sinus mucocele with orbital complications: Management by varied surgical approaches. Asian J Neurosurg. 2012; 7:135-140.
41. Stiernberg CM, Bailey BJ, Calhoun KH, Quinn FB. Management of invasive frontoethmoidal sinus mucoceles. Arch Otolaryngol Head Neck Surg. 1986; 112:1060-1063.
42. Khong JJ, Malhotra R, Selva D, Wormald PJ. Efficacy of endoscopic sinus surgery for paranasal sinus mucocele including modified endoscopic Lothrop procedure for frontal sinus mucocele. J Laryngol Otol. 2004; 118:352-356.
43. Yap SK, Aung T, Yap EY. Frontal sinus mucoceles causing proptosis--two case reports. Ann Acad Med Singapore. 1998; 27:744-747.
44. Misirovs R, Gohil R, Ross P, Manickavasagam J. Not quite Pott's puffy tumour. BMJ Case Rep. 2018; bcr-2018.
45. Saki N, Nikakhlagh S, Rasekhi R. Frontal mucocele presenting as a subcutaneous giant mass. Biomed Pharmacol J. 2015; 8:43-45.

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Disclaimer

The views expressed herein are those of the authors and do not necessarily reflect the views of Rhode Island Hospital, The Warren Alpert Medical School of Brown University, Geisel School of Medicine at Dartmouth, Dartmouth Hitchcock Clinics, or Dartmouth Hitchcock Medical Center.

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