The rare presentation of a frontal mucocele complicated by a Pott's puffy tumor and an epidural-cutaneous fistula: illustrative case

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BACKGROUND Frontal mucoceles develop due to accumulation of mucoid secretions within the frontal sinuses. They can lead to serious consequences with further expansion and destruction of the surrounding bones or infection that might spread intracranially.

OBSERVATIONS The authors present a case of a 37-year-old male with a frontal mucocele and the rare presentation of Pott’s puffy tumor and an epidural-cutaneous fistula, as well as a literature review of previously reported cases of epidural cutaneous fistula and sinocutaneous fistula, their predisposing factors, and their management.

LESSONS A mucocele is a benign entity that can rarely present with potentially significant complications. Open surgery is required in patients who have frontal sinus posterior wall involvement, osteomyelitis, or intracranial involvement.

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KEYWORDS frontal mucocele; Pott’s puffy tumor; epidural cutaneous fistula; frontal sinusitis; frontal osteomyelitis

Frontal mucocele is a benign entity that results from mucoid secretion accumulation. However, it can present with serious consequences, such as Pott’s puffy tumor (PPT), that need prompt medical attention and management. Another rarely reported complication is epidural-cutaneous fistula (ECF), which needs a complex surgical approach to avoid drastic intracranial complications.

In this report, we describe a case of frontal mucocele complicated by PPT and ECF, imaging features, and management, as well as a literature review of ECF and sinocutaneous fistula (SCF) and a proposed algorithm that aims to aid in the management of these complications in the setting of frontal sinus disease.

Illustrative Case

A 37-year-old male patient initially presented to the hospital with only the complaint of a painless “bump on his forehead.” On physical examination, he had a noticeable swelling eccentric to the right lower forehead. Computed tomography (CT) showed an infected left frontal mucocele and an overlying scalp abscess (Fig. 1). The patient was discharged with an outpatient appointment to follow up with magnetic resonance imaging (MRI) and further planning. MRI showed an infected frontal mucocele with an infected frontal soft tissue abscess with a connecting sinus tract (Fig. 2).

The patient presented for outpatient consultation 1 week later with worsened frontal swelling and exudative drainage, as well as headache and pain around the mass. He was immediately admitted to the hospital and started on ceftriaxone, metronidazole, and vancomycin for empirical coverage. Compared with the previous CT scan, the CT scan performed during this admission was significant for enlargement of the anterior frontal abscess (Fig. 3).

A decision was made to proceed with external sinus surgery and cranietomy rather than endoscopically due to the sinus wall defects, osteomyelitis, and presence of ECF1 (Fig. 4). A standard bifrontal cranietomy was scheduled with intention to cranialize the frontal sinus and drain the abscess. Intraoperatively, a large, well-encapsulated right frontal abscess was drained, and cultures were obtained. After thorough irrigation and debridement, a small tract

ABBREVIATIONS CT = computed tomography; ECF = epidural-cutaneous fistula; IV = intravenous; MRI = magnetic resonance imaging; PPT = Pott’s puffy tumor; SCF = sinocutaneous fistula.

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was noted that extended into the bone and to the left, where another separate large cystic lesion (mucocele) was identified. Subsequently, the mucocele was isolated, which appeared to have eroded the anterior skull base and the left medial orbital wall. It also appeared to have tracked down into the left ethmoidal air cells. The mucocele was then resected, and the underlying mucosa was removed. A large defect was noted in the posterior wall of the expanded left frontal sinus with exposed dura.

The infected posterior wall of the frontal sinus was removed, all of the frontal mucosa was resected, and the cavity was irrigated with antibiotic solution. A bifrontal craniectomy was performed in standard fashion, and the bone flap was elevated and discarded, ensuring clean bone margins without discoloration or evidence of infection. After ensuring complete resection of the mucocele and infected bone, we began closure. The entire surgical bed was thoroughly irrigated with antibiotic-impregnated saline solution. The previously harvested vascularized pericranial flap was then brought over and attached to the dura and folded over itself to line the cranialized sinuses. The surgical bed was irrigated thoroughly, and meticulous hemostasis was achieved. With the pericranial flap secured in place, two flat Jackson Pratt (JP) drains were placed overlying it into the anterior-most margins and tunneled posterior to the incision. A separate small, round drain was placed in the superficial abscess cavity.

The abscess culture grew *Staphylococcus haemolyticus*, sensitive to oxacillin, and the patient was started on intravenous (IV) nafcillin for a 6-week course of antibiotics. The pathology result of the excised frontal bone showed chronic osteomyelitis. There was also evidence of acute and chronic inflammation of the pericranium and the left frontal sinus content.

The findings of the patient’s neurological examinations remained intact in the preoperative and postoperative period. Although the abscess was close to the left orbit, the preoperative and postoperative ophthalmological examinations were intact. All drains were removed within 3 days of the patient’s postoperative stay. The patient was discharged in a stable condition with arrangements for administration of IV antibiotics at home, and he is planned to undergo a synthetic cranioplasty after completion of IV antibiotics.
Discussion

Observations

PPTs and SCFs/ECFs are rare complications of frontal sinusitis in the recent era of advanced antibiotics. In 2008, Wu et al. reported 13 cases from the English literature published since 1950 with SCF as a complication of frontal sinusitis, 4 of which also had associated PPT (Table 1). Simonin et al.’s literature review after 2008 described 5 more cases of SCF and PPT as complications of chronic sinusitis and 1 case with post-traumatic right frontal craniotomy and cranial osteoplastic reconstructive surgery. There were only 3 reported cases of ECF associated with PPT as a complication of frontal sinusitis. There were only 2 cases in the English literature of PPT as a complication of frontal sinus mucocele. Another case reported mucocele associated with frontal lacrimal fistula. We report the only case of ECF complicating frontal sinus mucocele.

The management of the majority of the previously reported cases with SCFs/ECFs with or without PPT involved, besides broad-spectrum antibiotics, either external open sinus surgery or combined external and endoscopic sinus surgery for adequate drainage (Table 1). One case with SCF and PPT underwent endoscopic sinus surgery; however, this treatment was unsuccessful and was followed by external sinus surgery. There are 3 cases with SCF who underwent successful drainage with an endoscopic sinus approach only. The 3 reported cases with ECF underwent open surgery or a combined approach.

Sinus mucocele is a benign expansile cystic mass that can be asymptomatic or associated with nonspecific symptoms. However, some patients may present with serious intracranial or ophthalmic complications. In this case report, we present an unusual complication of frontal sinus mucocele presenting with ECF and PPT.
| Authors & Year          | Predisposing Condition | Type of Fistula | PPT | Surgical Approach                                                                 |
|------------------------|------------------------|-----------------|-----|------------------------------------------------------------------------------------|
| Reinecki & Montgomery, 1969 | Frontal sinus mucocele | Frontal lacrimal fistula | No  | Open surgery: frontal mucocele resection w/ frontal sinus obliteration               |
| Simonsz et al., 1982   | Chronic frontal sinusitis | SCF             | No  | Open surgery: surgical incision & drainage of frontal sinus                         |
| Marfatia et al., 1997  | Chronic frontal sinusitis | SCF             | Yes | FESS (unsuccessful), open surgery: incision of diseased frontal periosseum w/ exenteration of disease mucosal lining & obliteration of frontal sinus |
| Marshall & Jones, 2000  | Chronic frontal sinusitis | SCF             | Yes | 4 patients—open surgery: 1 underwent drilling of myelitic bone, 1 had craniotomy & lytic bone drilled, 2 others underwent Riedel's procedure |
| Goldfarb et al., 2004  | Chronic frontal sinusitis | SCF             | Yes | Open surgery: incision & drainage of frontal scalp abscess that led to chronic draining SCF, w/ continuous irrigation of wound & planned osteoplasty surgery |
| Seyhan & Ozerdem, 2005 | Chronic frontal sinusitis | SCF             | No  | Open surgery: incision & drainage of frontal sinus, debridement of necrotic tissue, & intracp injection of red rifampicin into sinus |
| Davidson & McComb, 2006 | Chronic frontal sinusitis | ECF             | Yes | Open surgery: bicornoral incision made, followed by frontal craniectomy w/ resection of osteomyelitic bone & evacuation of epidural abscess |
| Wu et al., 2008        | Chronic frontal sinusitis | SCF             | No  | FESS: 3 patients underwent endoscopic sinus surgery w/ complex closure of SCF        |
| Minutilli et al., 2008 | Post-traumatic rt frontal craniotomy & cranial osteoplastic reconstructive surgery by acrylic resins 8 yrs earlier | SCF             | Yes | Open surgery: radical surgical removal of all prosthesis, infected collection & bone & secondary cranial reconstructive surgery through mandatory open access |
| Masterson & Leong, 2009 | Frontal sinusitis      | SCF             | Yes | Combined approach: frontal sinus surgery using combined endonasal & percutaneous approach w/ placement of frontonasal drain |
| Shin et al., 2012      | Frontal sinus mucocele | SCF             | Yes | Combined approach: frontal sinus surgery using combined endonasal & percutaneous approach & frontal bone reconstruction w/ resorbable mesh plate & bone cement |
| Perić et al., 2017     | Frontal sinusitis      | ECF             | Yes | Combined approach: endoscopic bilat anterior ethmoidectomy, open surgical drainage of epidural abscess & debridement of osteomyelitic focus, followed by removal of fistula & inflamed frontal sinus mucosa |
PPT typically presents with other infectious symptoms besides swelling, such as fever and leukocytosis. The patient in our case study, however, presented initially with only mild frontal scalp swelling and a frontal sinus mucocele on CT. The patient was discharged from the emergency department and presented later to our outpatient clinic with worsening symptoms and a ruptured abscess. In this case, it is difficult to determine whether the progressive mucocele expansion led to the bony erosions, dehiscence, and osteomyelitis or whether the acute or chronic sinusitis led to the infection of the mucocele and creation of an ECF and subsequent rupture of the frontal abscess. Regardless of the sequence of events, it is pivotal to recognize this potential and significant complication of a benign mucocele, and this presentation emphasizes the importance of having high clinical suspicion for PPT, regardless of the presence or absence of infectious markers to avoid the dreaded intracranial complications.

The standard of care for PPT and SCF as a complication of frontal sinusitis without intracranial complications is endoscopic sinus surgery with the possibility of performing external sinus surgery in case of failure. However, in the case of the presence of a defect in the posterior wall of the frontal sinus (ECF), some intracranial complications (such as epidural and subdural empyema and frontal abscess), and/or osteomyelitis, the mainstay of treatment is an open surgical approach with craniectomy to remove infected part of frontal bone, evacuation of epidermal collection, and endoscopic sinus surgery. In the case of the presence of a defect in the posterior wall of the frontal sinus (ECF), some intracranial complications (such as epidural and subdural empyema and frontal abscess), and/or osteomyelitis, the mainstay of treatment is an endoscopic sinus surgery with or without an endoscopic sinus procedure. The patient in our case study, however, presented initially with only mild frontal scalp swelling and a frontal sinus mucocele on CT. The patient was discharged from the emergency department and presented later to our outpatient clinic with worsening symptoms and a ruptured abscess. In this case, it is difficult to determine whether the progressive mucocele expansion led to the bony erosions, dehiscence, and osteomyelitis or whether the acute or chronic sinusitis led to the infection of the mucocele and creation of an ECF and subsequent rupture of the frontal abscess. Regardless of the sequence of events, it is pivotal to recognize this potential and significant complication of a benign mucocele, and this presentation emphasizes the importance of having high clinical suspicion for PPT, regardless of the presence or absence of infectious markers to avoid the dreaded intracranial complications.

Lessons
A mucocele is a benign entity that can rarely present with potentially significant complications such as PPT and ECF. Early recognition and proper management are important to avoid dreaded complications. According to prior literature, open surgery with or without an endoscopic sinus procedure is the best approach in patients with osteomyelitis, intracranial involvement, or frontal sinus posterior wall defect.

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Conception and design: Abbas, Al-Smadi, Luqman. Acquisition of data: Abbas, Al-Smadi, Smitt, Luqman. Analysis and interpretation of data: Abbas, Al-Smadi, Luqman. Critically revising the article: all authors. Reviewed submitted version of manuscript: Abbas, Al-Smadi, Luqman. Approved the final version of the manuscript on behalf of all authors: Abbas. Administrative/technical/material support: Smitt.

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