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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 craniectomy rather than endoscopically due to the sinus wall defects, osteomyelitis, and presence of ECF[1] (Fig. 4). A standard bifrontal craniectomy 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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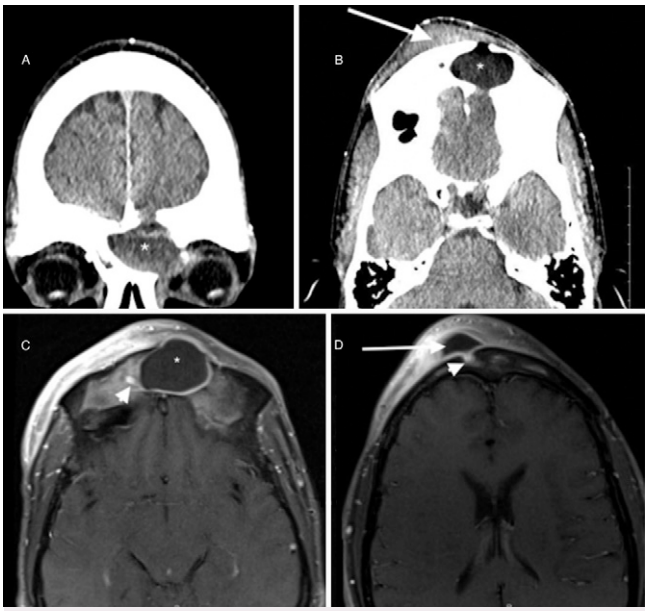

FIG. 1. Imaging performed at initial presentation. **A:** CT showing a cystic mass expanding the left frontal sinus (*asterisk*). **B:** CT showing right frontal scalp fluid collection (*arrow*) with defects in the anterior and posterior wall of the left frontal sinus. **C and D:** MRI showing 2.7 × 3.0 × 4.0–cm cystic lesion expanding the left frontal sinus (*asterisk*) with diffusion restriction (not shown) and a sinus tract (*arrowheads*) connecting to a 1.8 × 1.2 × 1.1–cm frontal soft tissue fluid collection (*arrow*). There is also mild bone marrow enhancement. These findings indicate an infected mucocele, osteomyelitis, and frontal soft tissue abscess with a connecting sinus tract. In constellation with findings of bony defects on CT, this raises concern about an ECF.

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

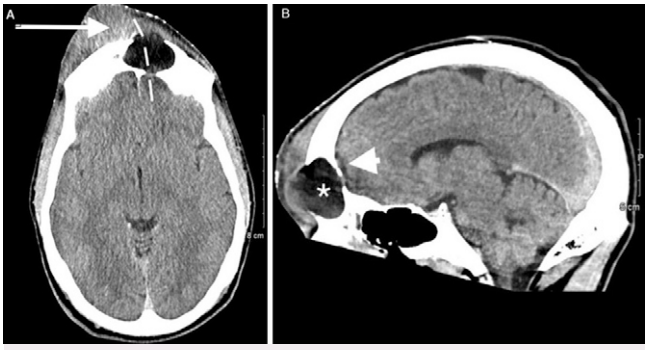

FIG. 2. CT scans performed preoperatively. **A:** Enlargement of the right frontal soft tissue swelling consistent with known PPT ruptured through the skin (*arrow*) with demonstration of the connection between the epidural space and ruptured abscess collection indicating an ECF (*dashed line*). **B:** Frontal mucocele appears more enlarged and bulging through the anterior wall defect (*asterisk*), and a small epidural fluid collection posterior to the frontal sinus is seen (*arrowhead*).

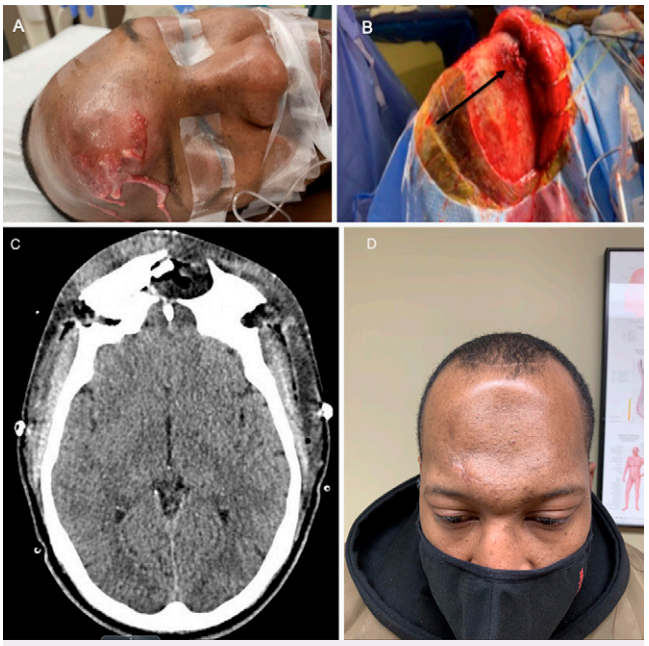

FIG. 3. A: Ruptured PPT with exudative drainage through the skin. **B:** Subperiosteal abscess with necrotic bone (*arrow*). **C:** Postoperative CT scan with changes of bilateral frontal craniotomy and drainage of right frontal scalp collection and excision of left frontal mucocele. **D:** Resolution of frontal swelling on follow-up at 3 weeks.

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.

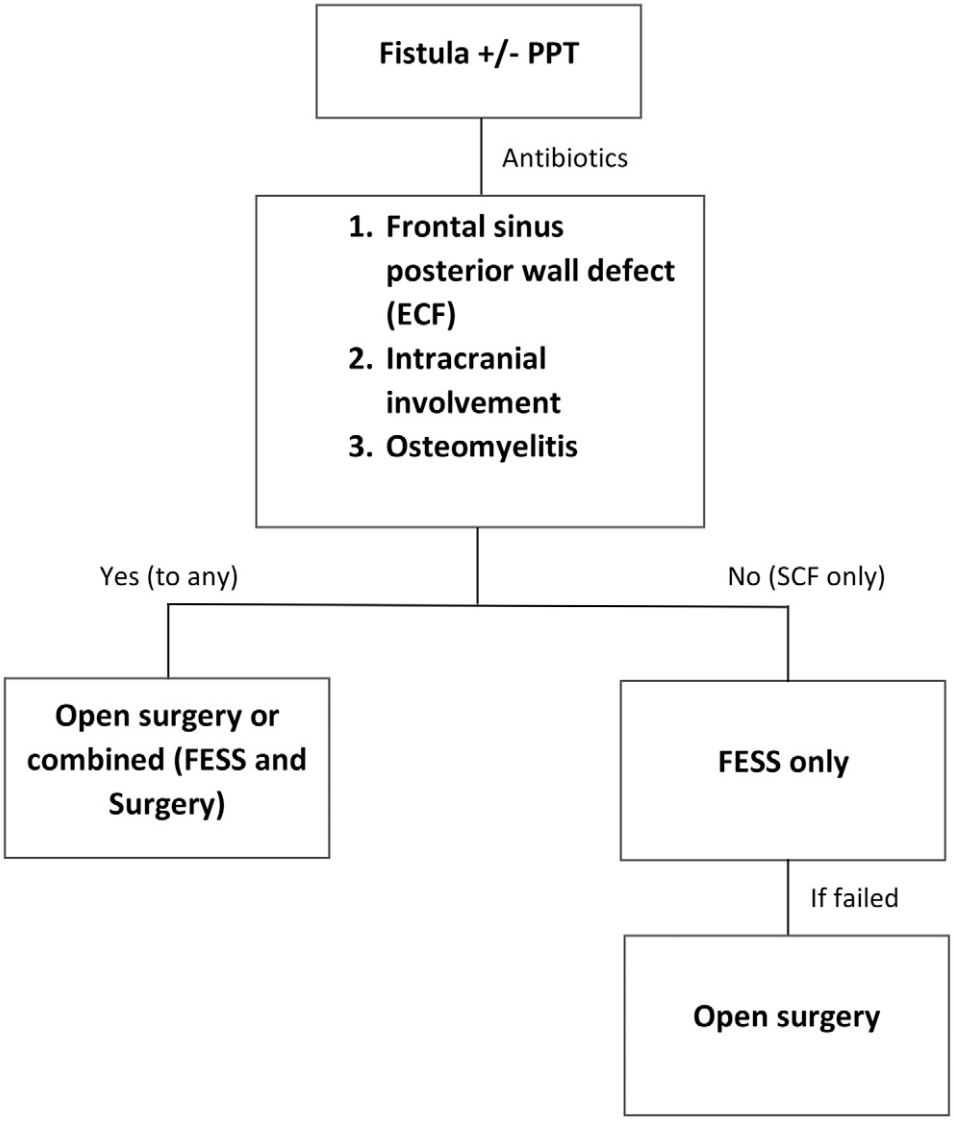

FIG. 4. Decision algorithm for the management of SCF/ECF, in addition to antibiotics. FESS = functional endoscopic sinus surgery.

Discussion

Observations

PPTs and SCFs/ECFs are rare complications of frontal sinusitis in the recent era of advanced antibiotics.[2] In 2008, Wu et al.[3] 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).[4–23] Simonin et al.'s[4] 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.[4–6] There were only 2 cases in the English literature of PPT as a complication of frontal sinus mucocele.[7] Another case reported mucocele associated with frontal lacrimal fistula.[8] 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.[9] In this case report, we present an unusual complication of frontal sinus mucocele presenting with ECF and PPT.

TABLE 1. Review of previously reported cases of SCF and ECF

Authors & Year	Predisposing Condition	Type of Fistula	PPT	Surgical Approach
Reinecki & Montgomery, 1969[15]	Frontal sinus mucocele	Frontal lacrimal fistula	No	Open surgery: frontal mucocele resection w/ frontal sinus obliteration
Simonsz et al., 1982 [16]	Chronic frontal sinusitis	SCF	No	Open surgery: surgical incision & drainage of frontal sinus
Marfatia et al., 1997[17]	Chronic frontal sinusitis	SCF	Yes	FESS (unsuccessful), open surgery: incision of diseased frontal periosteum w/ exenteration of disease mucosal lining & obliteration of frontal sinus
Marshall & Jones, 2000[18]	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[19]	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 osteoplastic surgery
Seyhan & Ozerdem, 2005[20]	Chronic frontal sinusitis	SCF	No	Open surgery: incision & drainage of frontal sinus, debridement of necrotic tissue, & intraop injection of red rifampicin into sinus
Davidson & McComb, 2006[6]	Chronic frontal sinusitis	ECF	Yes	Open surgery: bicoronal incision made, followed by frontal craniectomy w/ resection of osteomyelitic bone & evacuation of epidural abscess
Wu et al., 2008[3]	Chronic frontal sinusitis	SCF	No	FESS: 3 patients underwent endoscopic sinus surgery w/ complex closure of SCF
Minutilli et al., 2008[21]	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[12]	Frontal sinusitis	SCF	Yes	Combined approach: frontal sinus surgery using combined endonasal & percutaneous approach w/ placement of frontonasal drain
Shin et al., 2012[8]	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[5]	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

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TABLE 1. Review of previously reported cases of SCF and ECF

Authors & Year	Predisposing Condition	Type of Fistula	PPT	Surgical Approach
Simonin et al., 2018[4]	Frontal sinusitis	Epidural abscess, communicating through multiple frontal bone defects, to the skin; ECF	Yes	Combined approach: craniectomy to remove infected part of frontal bone, evacuation of epidural collection, endoscopic sinus surgery
Min & Kim, 2020[22]	Frontal sinusitis	SCF after incision & drainage of scalp collection under the impression that it is infected sebaceous cyst	Yes	FESS: endoscopic sinus surgery w/ removal of inflammatory lesions in frontal sinus; skin lesion healed spontaneously
Terui et al., 2021[23]	Chronic frontal sinusitis	SCF	Yes	Combined approach: endoscopic sinus surgery & Killian frontal sinus surgery silicone frontal sinus stent placement in nasofrontal duct

FESS = functional endoscopic sinus surgery.

PPT typically presents with other infectious symptoms besides swelling, such as fever and leukocytosis.[10] 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.[11]

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.[12] 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 or without an endoscopic sinus procedure.[1,13,14] Figure 4 shows an algorithm proposal for the management of SCF and ECF in the setting of frontal sinus disease.

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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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Abbas, Al-Smadi, Luqman. Acquisition of data: Abbas, Al-Smadi, Smitt, Luqman. Analysis and interpretation of data: Abbas, Al-Smadi, Luqman. Drafting the article: Abbas, Al-Smadi. 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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