



## Frontocutaneous Fistula Secondary to Frontal Sinusitis

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### Authors' contributions

This work was carried out in collaboration with all authors. Author PAT operated the patient on both occasions, designed the study and wrote the first draft of the manuscript. Authors EKN and AK managed the literature searches and contributed to developing a management plan for the patient and author BAT managed the postoperative eye problems of the patient. All authors read and approved the final manuscript.

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Case Study

### ABSTRACT

Frontocutaneous fistula is a rare complication of frontal sinusitis. This mostly occurs as a result of osteomyelitis of the frontal bone on account of sinusitis. These are managed by endoscopic closure or by external approach depending on the cause and extent of sinusitis. We report on the case of a 57 year old man who presented to our ENT Unit with a defect on the face medial to the right eye.

**Keywords:** Frontocutaneous fistula; frontal sinusitis; pott's puffy tumour; osteomyelitis.

## 1. INTRODUCTION

Frontocutaneous fistulae have been reported in many publications but the incidence is rare at present. Frontal sinusitis today rarely results in frontal abscess leading to fistula formation [1]. This is due to a reduction in the number of cases of osteomyelitis as antibiotic use has greatly increased [2-4]. In countries with large populations of low socio-economic status however the incidence may still be high. In a study carried out in Ibadan, Nigeria, out of 228 patients with chronic sinusitis only 2 i.e. 0.88% had frontocutaneous fistulae [5]. These lesions mostly result from osteomyelitis of the frontal bone i.e. 'Pott's puffy tumour'. The anterior, posterior or inferior walls are mostly affected. This only occurs in diploic bone thus only noted in the maxillary sinus in children and frontal sinus in adults [6]. This is a complication of sinusitis which tends to occur as a result of the obstruction of the frontal sinus ostia by a tumour, trauma or due to defective mucociliary clearance e.g. cystic fibrosis. Frontal sinusitis commonly presents with headache, facial pain, nasal obstruction and then frontal swelling due to subperiosteal abscess formation with eventual rupture of the swelling. This leaves a defect on the face as reported in this case report.

## 2. CASE HISTORY

A 57 year old farmer with a history of asthma presented to the ENT Unit of our hospital with a one year history of a right facial defect and recurrent nasal obstruction. The patient had been seen a year and half earlier with a right frontoethmoidal mass and had been asked to do a CT scan of the brain and paranasal sinuses. Patient however got lost to follow up and had some herbal medications due to financial difficulties. The mass then ruptured leaving a defect on the face with a foul smelling discharge. There was no nasal discharge, epistaxis or postnasal drip.

On examination, the significant findings were a fistula on the right frontoethmoidal region (as shown in Fig. 1) with hypertrophied nasal turbinates. Impression formed was Frontocutaneous Fistula secondary to Chronic Sinusitis.

Investigations done were reported as follows: Hb13.8 g/dL, WBC  $5.32 \times 10^9/\mu\text{L}$ , Platelet count  $91 \times 10^3/\mu\text{L}$ , and Sickling negative. CT scan of the paranasal sinuses showed heterogeneous soft

tissue densities in the right frontal, ethmoidal and maxillary sinuses. The floor of the right frontal sinus and the lateral wall of the right ethmoidal sinuses were also defective. These are shown in Fig. 2. Impression formed by the radiologist was Chronic Invasive Fungal Sinusitis.

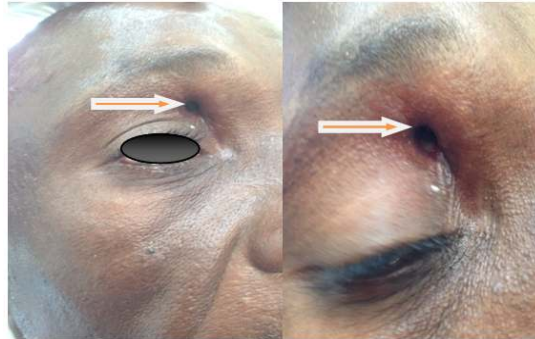


Fig. 1. Arrowed portions shows the right frontocutaneous fistula

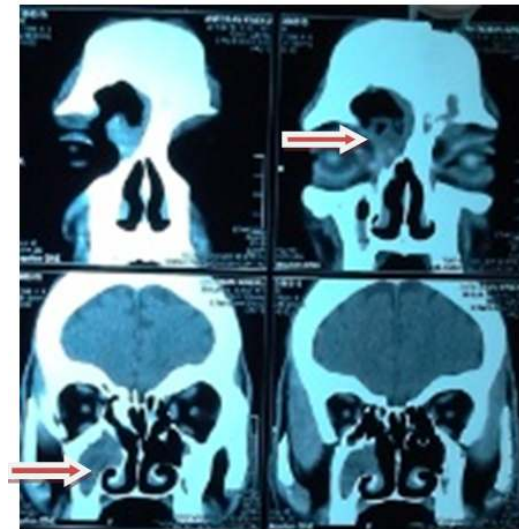


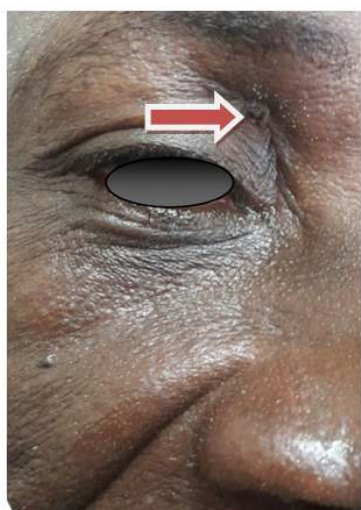
Fig. 2. CT scans showing the right frontal sinus wall defect and right frontal and maxillary sinus soft tissue densities (arrowed portions)

A two-staged surgery was then planned. First surgery was to establish the diagnosis of the cause of the fistula by histology and then to treat patient in the second stage based on the histology report. Thus patient had Right Inferior Meatal Antrostomy and Right Intranasal Frontoethmoidectomy and curettage of the right frontal sinus through the frontocutaneous fistula. A frontonasal tube was also inserted. The

intraoperative findings were hypertrophied right maxillary antral mucosa with a polyp in the right middle meatus and an enlarged right frontal sinus containing soft greenish tissues. The frontonasal tube was left in place for 8 weeks. Samples from both the right maxillary and frontal sinuses were then sent for histopathology.

Histology samples taken from the right maxillary and frontal sinuses and the middle meatal polyp were reported to show partly ulcerated, pseudostratified epithelia. The stromata were edematous and mixed with plasma cells, neutrophils and lymphocytes. Other parts of the samples showed hyperplastic glands, granulation tissue and numerous bacterial colonies. No evidence of malignancy was seen. Impression formed by the pathologist was Chronic Sinusitis.

Postoperatively patient was given fluticasone furoate nasal spray for 8 weeks and Ciprofloxacin for 2 weeks. At the 8th postoperative week, a right external frontoethmoidectomy was done and the fistulous tract excised. The wound was then closed in layers and the frontonasal tube removed. Subsequently patient had chemosis, edema of the upper eyelid and restrictive ophthalmopathy of the right eye. 2hourly Ciprofloxacin eye drops on the right eye was thus given. Patient was then discharged after the edema had receded. Initially at home patient had a right preseptal cellulitis which was treated with oral Clindamycin 300 mg qid for 7 days. Resolved defect after 6 months may be noted in Fig. 3.



**Fig. 3. Repaired frontocutaneous fistula (arrowed portion)**

### 3. DISCUSSION

The frontal sinus develops as an outpouching of the anterior ethmoidal air cells around the second year of life. The sinus is drained through the frontonasal duct via the frontal recess which is shaped as an hour-glass. The frontal recess can get blocked by mucosal edema. This leads to retention of secretions which then get infected. *Streptococcus milleri*, *Bacteroides spp*, *Staphylococcus aureus* and *Proteus species* have been implicated as the commonest causes [7,8]. The mucous thus becomes more viscid and results in frontal recess scarring.

Other causes of defective frontal sinus drainage may include a middle meatal polyp, a large concha bullosa, a laterally rotated uncinata process that contacts the middle turbinate, and, a medially convex middle turbinate that contacts the lateral nasal wall. Previous middle turbinectomy can also lead to stenosis of the frontal sinus ostium [9].

Various complications can result from the defective frontal sinus drainage. These may be classified as either intracranial or extracranial complications. The intracranial complications include meningitis, intracerebral abscess, epidural and subdural abscesses and dural sinus thrombosis. The extracranial complications which are mainly orbital are classified by Chandler [10]. This is as shown in Table 1.

**Table 1. Chandler's classification of orbital complications of sinusitis**

Group	Chandler's classification
First	Inflammatory edema
Second	Orbital cellulitis
Third	Subperiosteal abscess
Fourth	Orbital abscess
Fifth	Cavernous sinus thrombosis

Frontocutaneous fistulae may also result from the obstruction of the ostia of the frontal sinus. The obstruction leads to a buildup of pressure within the sinus which then results in osteitis and subsequently osteomyelitis of the frontal sinus wall. A subperiosteal abscess then forms and this ruptures resulting in the fistula formation. The frontal sinus is commonly affected due to the tortuous course run by the frontonasal duct through the ethmoidal labyrinth [6]. The duct is thus susceptible to blockage. Mucocele formation is another extracranial complication of frontal sinusitis [1].

The diagnosis is made by clinical examination and by radiology, most preferably by CT scan. This defines the bone destruction and the soft tissue densities in the paranasal sinuses.

The treatment is conservative and surgical. The conservative treatment includes antibiotics and nasal decongestants. The surgical treatment is largely dependent on the extent and site of the disease [11]. The aim of frontal sinus surgery is to drain the empyema and reestablish frontoethmoidal drainage. This is achieved by decompression of the frontal recess by removing parts of the ethmoidal bulla and the uncinate process [1]. It may involve functional endoscopic sinus surgery or external frontoethmoidectomy with fistulectomy. Other cases in literature were managed by obliterating the frontal sinus with autologous fat, hydroxyapatite cement, muscle, bone, bioactive glass etc [1,6,12]. Obliteration of the frontal sinus however requires meticulous removal of the frontal sinus mucosa and permanent occlusion of the frontonasal duct.

Functional endoscopic sinus surgery (FESS) clears the sinuses of inflammatory tissue and bacteria and enhances mucociliary clearance by eliminating obstruction of outflow tracts while preserving normal sinus physiology and function. Endoscopic procedures for frontal sinusitis may be more technically difficult than open/external procedures. However, endoscopic approaches always achieve better cosmesis with no outwardly visible scars or deformities. Diseases in the other sinuses can also be treated concurrently [11].

Endoscopic surgery was not possible at our centre since we did not have the equipment. In other reports the defects in the frontal walls were sealed up with grafts via osteoplastic flaps [1]. This may not be possible if the frontal sinus is hypoplastic. Ballon sinuplasty has also been described though its effectiveness has not been strongly supported in literature [10].

It must be noted that these fistulae are mostly managed by multiple surgeries. A few reports have however recorded treatment of such cases endoscopically in one-staged surgeries. The patient thus needs to be consented on the need for multiple surgeries as was done in our case [1].

In our case, the unhealthy frontal sinus mucosa and contents were curetted through the fistula and intranasal frontoethmoidectomy also done

with excision of the middle meatal polyp. This enhanced aeration of the right frontal sinus. Right inferior meatal antrostomy was also done to remove the diseased maxillary antral mucosa and contents to enhance drainage of the rest of the paranasal sinuses. Enhancing drainage of the maxillary sinus ultimately enhances the function of the other paranasal sinuses. The maxillary sinus is thus described as the 'conductor of the orchestra' [13]. Functional endoscopic sinus surgery was not done in this case since the facility was not available at our centre.

#### 4. CONCLUSION

Functional endoscopic sinus surgery is the standard for managing frontal sinusitis today when conservative management has failed. However, frontocutaneous fistula can be still be managed in resource challenged facilities once the underlying pathology has been well treated.

#### CONSENT

Patient gave an informed consent for this case to be reported. This is available for review by the editor.

#### ETHICAL APPROVAL

It is not applicable.

#### COMPETING INTERESTS

Authors have declared that no competing interests exist.

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