



Case Report

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## Fungal rhinosinusitis with atypical presentation – a report of two cases

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### PEER REVIEW

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### ABSTRACT

Rhinosinusitis affects approximately 20% of the population, and the chronic rhinosinusitis represents over 90% of all cases of rhinosinusitis. The correct diagnosis is important for proper treatment and to predict its evolution. This study presents two cases of atypical frontal sinus disease, which the follow-up revealed a diagnosis of fungal rhinosinusitis. The present study aims to describe the cases of two patients with atypical lesions on the left frontal sinus; the treatment options, surgical approach, results, diagnosis and follow-up are further discussed. A significant increase in the reported cases of fungal rhinosinusitis has been seen in the last two decades, justified by the use of broad-spectrum antibiotics and steroids, as well as the increased number of immunocompromised individuals. This study reports the cases of two patients with a type of fungal rhinosinusitis named "fungal ball", characterized by a tangle of hyphae in the sinuses without tissue invasion. The treatment included surgical removal of the fungal infectious process with aeration of the affected sinus, and the procedure was successfully performed in our patients.

## 1. Introduction

Rhinosinusitis affects approximately 20% of the population, and the chronic rhinosinusitis represents over 90% of all cases of rhinosinusitis[1,2]. Fungal rhinosinusitis represents 7% of all cases of rhinosinusitis. Isolated fungal sinusitis of frontal sinus is a rare condition, accounting for only 5% of all fungal rhinosinusitis[1,2]. The most commonly affected sinus is the maxillary, followed by the sphenoid, frontal and ethmoidal sinuses[1,2].

While the acute rhinosinusitis is well-categorized[1], the chronic rhinosinusitis criteria, as well as the role of the fungus in this condition are still in further research[1]. The first attempt to classify the chronic rhinosinusitis was in 1965, when Hora *et al.*[3] divided the cases in two categories: (1) invasive, in which the infection invades adjacent tissue, and (2) non-invasive, in which the disease has the same features and a chronic bacterial infection of the sinuses[3]. Today, the classification is mostly based on the immune interaction of the fungus/host and the degree of tissue

invasion[1-3]. The invasive forms could be chronic or acute and the non-invasive ones are (1) allergic fungal sinusitis; (2) fungal ball; and (3) saprophytic infection[4,5]. The objective is to present two cases of atypical frontal sinus diseases that the follow-up revealed a diagnosis of fungal rhinosinusitis, the focuses of the cases are on the diagnostic tools, treatment and follow-up.

## 2. Case report

A "case report" study was performed. The project was evaluated and approved by the Banco de Olhos de Sorocaba Hospital ethics committee. An informed consent form was given to the two patients enrolled in this study, and with it they received an explanation about the study and the objective. Both patients agreed with the terms of the study and signed the informed consent form. The charts of the patients were then collected and data regarding to the objective of the study were gathered.

### 2.1. Patient 1

A 46-year-old male patient with no previous medical history complained of the frontal and periorbital headache on the left that began 1 year before the consult. Moreover, it did not respond to over the counter painkillers. He denied nasal obstruction, as well as

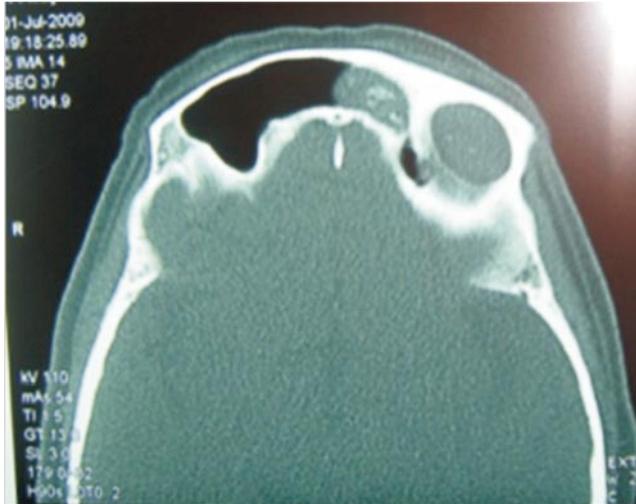
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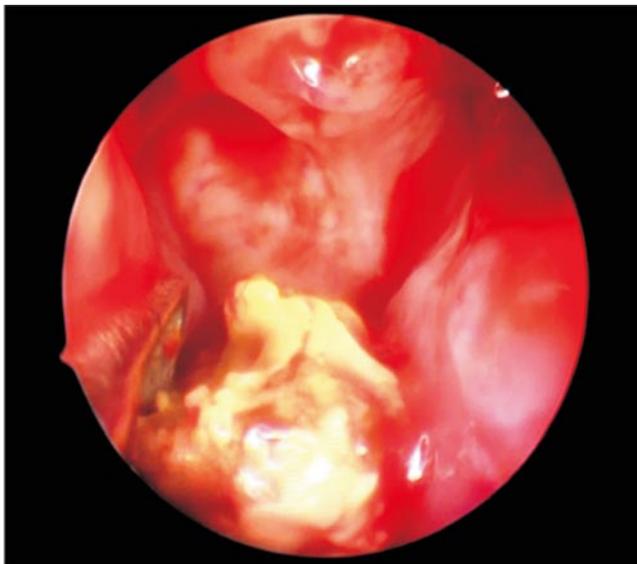
as rhinorrhea or olfaction disturbances. He was brought to consult a computerized tomography of the paranasal sinuses 9 m before, and it showed that opacification was on the left frontal sinus.

Physical exam was positive for retro-nasal discharge, and rhinoscopy showed hypertrophy of nasal turbinates bilaterally. A treatment with levofloxacin orally for 14 days, nasal irrigation and nasal mometasone 200 µg twice a day was prescribed, but the patient did not report improvements on the headache. A new CT (Figure 1) showed an opacification on the left frontal sinus with hyperdense images in the center, and a nasal endoscopy did not show any noteworthy findings.



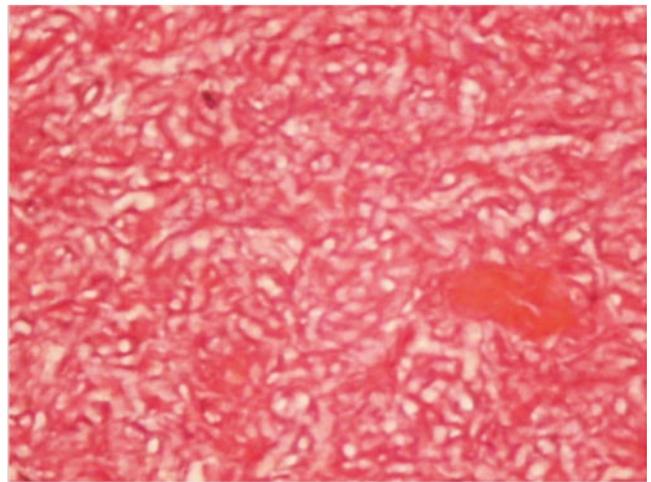
**Figure 1.** Bone-window of an axial CT of the paranasal sinuses showing and opacification on the left frontal sinuses with a typical “metallic density” area.

The patient underwent a functional endoscopic sinus surgery, and the left frontal, anterior ethmoid and maxillary sinuses were approached bilaterally. A thick material drained from the left frontal recess during the surgery (Figure 2), and was completely removed. Then, the left frontal sinus was cleaned with saline solution and surgical liquid chlorhexidine soap using a syringe.



**Figure 2.** Image obtained by a nasal endoscope during the surgery of the patient, showing the drainage of a thick material to the middle meatus.

The patient referred that the headache disappeared as soon as he woke up from the surgery. The outpatient follow-up was uneventful, and no signs of recidivation had been observed for one year after the surgical procedure. Histopathology material collected in the left frontal sinus was compatible with *Aspergillus* species (Figure 3).

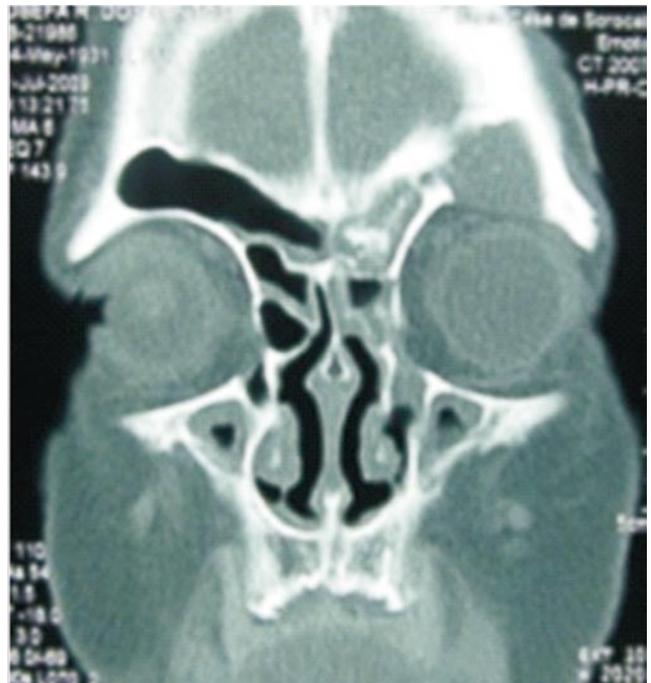


**Figure 3.** Analysis of the material obtained in surgery showing an mild inflammatory infiltrate with hyphae.  
Stain: Haematoxylin & Eosin; 400 ×

## 2.2. Patient 2

A 77-year-old female presented with pain and left peri-orbital edema that began 20 day before the consult, with associated cacosmia, nasal obstruction and thick rhinorrhea. When asked, she reported frontal pressure-like headache, which was partially improved with over the counter painkillers. She denied smoking or alcohol abuse. On physical exam, an evident eyelid ptosis was seen, but visual acuity testing was unremarkable. Rhinoscopy was positive for pus in the left nasal cavity, and nasal endoscopy showed pus drainage from the middle meatus and sphenoid-ethmoid recess.

Caldwell-waters radiograph showed a blurred image on the left frontal sinus. With a hypothesis of acute rhinosinusitis, the patient was treated with oral amoxicilin for 14 days, nasal irrigation four times a day and mometasone, 200 µg twice a day, but the patient did not report any improvement on the symptoms. A CT scan and an MRI of the paranasal sinuses were then performed (Figures 4 and 5).

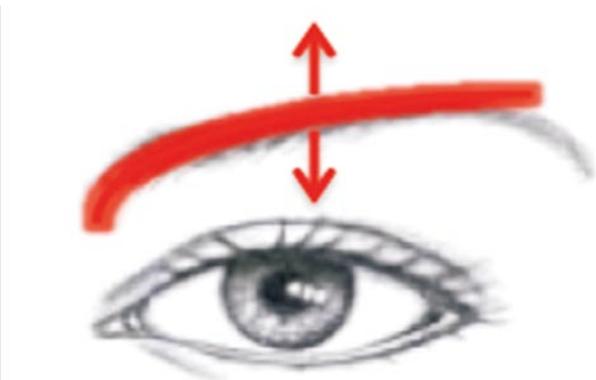


**Figure 4.** CT scan of the paranasal sinuses, showing bone erosion and opacification of the left frontal sinus with an adjacent image containing hyperdense material in the central area.



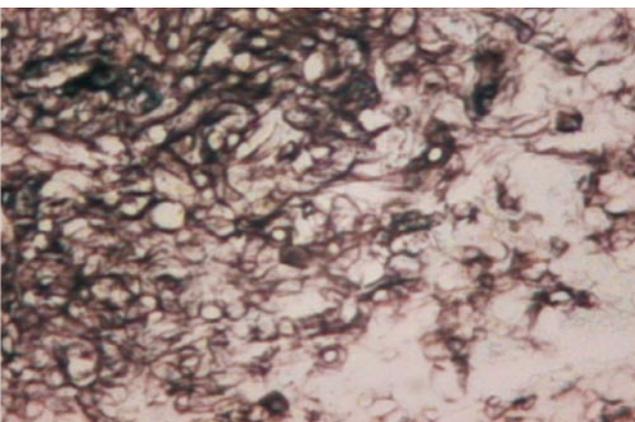
**Figure 5.** MRI of the patient showing a mass with regular contours in the left frontal sinus, measuring  $2.0 \times 1.2 \times 1.5$  cm, along with a second image of similar appearance in the left supra-orbital region.

Surgical approach was suggested, and it was performed under general anesthesia. The surgery was performed with combined modified Lynch-Howarth (Figure 6) and endonasal approaches, and the removal of the entire lesion of the frontal sinus was achieved. The outpatient follow-up was uneventful, and the histopathological exam and culture of the material revealed *Aspergillus* species (Figure 7).



**Figure 6.** Drawing of how the surgical incision was performed.

An eyebrow incision was made, but it was not extended to the medial canthus of the eye (like performed in the classical Lynch-Howarth approach). The underlying bone was removed with a proper drill, and the lesion was removed. Then an endonasal approach was performed, and the frontal recess was opened using a guide inserted from the external incision.



**Figure 7.** Analysis of the material obtained in surgery showing a mild inflammatory infiltrate with hyphae by Grocott stain (400  $\times$ ).

### 3. Discussion

A significant increase in the reported cases of fungal rhinosinusitis has been seen in the last two decades, justified by the use of broad-spectrum antibiotics and steroids, as well as the increased number of immunocompromised individuals. The patients described in this study had lesions in the frontal sinuses that could be either caused by fungus/bacteria or by a tumor, so the surgical approach was suggested to (1) improve the frontal sinus aeration and (2) collect material for histopathological study.

The first patient had complaints that suggested a chronic infection of the sinuses. Since the treatment with oral antibiotics and intranasal steroids did not improve the symptoms, thus an endonasal approach for a functional surgery of the paranasal sinuses was suggested. The second patient had a lesion on the left frontal sinus that seemed to be a solid mass, so an open surgery was suggested to attempt to remove the whole material for study, associated to an endonasal approach. Both procedures were uneventful and a full removal of material was achieved on both cases.

The resulting material was positive for fungal infection named “fungal ball”, characterized by a tangle of hyphae in the sinuses that do not invade adjacent tissue. The treatment included surgical removal of the fungal infectious process with aeration of the affected sinus, and the procedure was performed successfully in patients. Also, a cautious outpatient follow-up is important to prevent the disease to recur.

### Conflict of interest statement

We declare that we have no conflict of interest.

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