Case Report

Frontal Sinus Fungus Ball Operated on Endoscopically and Endonasally

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Abstract

Fungus ball is defined as the non-invasive accumulation of dense fungal concretions in a solitary sinus. Bone, blood vessels, and sinus mucosa are free of fungal elements. It is usually encountered in immunocompetent individuals with a female preponderance. It can involve the maxillary or the sphenoid sinuses.

The authors aim to report the clinical presentation, imaging and surgical treatment of 2 patients presenting with a fungus ball developed exclusively in the frontal sinus and operated on successfully endoscopically and endonasally.

They review the literature about this very uncommon entity.

INTRODUCTION

Aspergillus is an ubiquitous microorganism that is frequently found in the soil and in decomposing organic material (food, fruit, grain and plants). It produces volatile spores. Their concentration depends on environmental conditions. Humans are in permanent contact with fungal elements but most healthy people do not react to the presence of fungus due to a functioning immune system. So fungal diseases are fairly rare. The entire respiratory system (nose, paranasal sinus cavities, bronchi and lungs) can be involved by fungal diseases. Of the numerous species of Aspergillus, Aspergillus fumigatus is by far the fungal species that causes fungal rhinosinusitis in man in Europe followed by dematiaceous fungi [1-3].

The classification of fungal rhinosinusitis (FRS) has been proposed by Hora in 1965 [4] and updated by Chakrabarti A [5] in 2009. FRS is divided into two categories: the “invasive” and “non-invasive” forms.

Invasive FRS (IFRS) is characterized by the infiltration of the tissue by the fungus. It typically occurs in immune compromised patients. Non-invasive FRS (NFRS) is also called extra-mucosal FRS. The fungus elements are implanted on the sinus mucosa into an otherwise healthy sinus. The patient is usually immunocompetent.

The authors report two cases of a fungus ball developed exclusively in the frontal sinus operated on successfully endoscopically and endonasally.

CASE PRESENTATION

Case 1

A.B. is a 70-year-old man. He was complaining of headache. The medical history was uneventful. The patient had no history of trauma, surgery or medical problems. CT scanning (Figure 1 A,B) demonstrated a partial heterogeneous opacity of the left frontal sinus with micro calcifications and an eroded posterior frontal sinus wall. MR images confirmed the suspicion of a fungus ball in the left frontal sinus by displaying an hypo intense signal on T2-weighted sequences and enhancement of the surrounding mucosa (Figure 1 C,D). Moreover it excluded a CSF leak or a meningocele in front of the erosion of the posterior sinus wall. The patient was then operated on under general anaesthesia endonasally and endoscopically. The procedure consisted of a type II frontal sinusotomy (“uncapping the egg”) and opening of the bulla ethmoidalis. During surgery, black and greyish clay-like concretions were found. These concretions were completely removed using curved instruments and regular lavages of the frontal cavity with saline under moderate pressure. The histological examination of the surgical specimen was consistent with the diagnosis of a fungus ball (Figure 2). Culture did not yield fungal elements. The postoperative period was uneventful. Postoperative care was done to clean the crusts. The headache disappeared. The patient remains disease and symptom free one year later. The post-operative CT scan performed 1 year after the surgery was normal (Figure 1 E,F).
Case 2

O.P is a 63-year-old patient. He was complaining with moderate facial pain on the left side.

The sinus CT scan demonstrated a nasal septal deformity to the left and a complete opacity of the left frontal sinus with microcalcifications suggesting the diagnosis of a fungus ball (Figure 3: A,B,C). The patient has a history of multiple sclerosis that is stable and not requiring any immune suppressive medication. He is otherwise well with no history of surgery, trauma or other causes of immunodeficiency. The patient was operated on endonasally. The procedure consisted of a septoplasty, anterior ethmoidectomy (opening of the bulla ethmoidalis) and complete resection of the uncinate process (Type II frontal sinusotomy). Black and compact concretions were aspirated and removed from the sinus cavity confirming the diagnosis of a fungus ball. The sinus mucosa looked almost normal. The frontal cavity was irrigated several times with saline and freed of any concretions. The histological examination of the concretions confirmed the diagnosis of a fungal disease. The biopsy of the sinus mucosa surrounding the fungus ball did not show any sign of tissue invasion. One year after the procedure the patient remains free of disease. The postoperative CT scan is entirely normal (Figure 3: D,E,F).

DISCUSSION

At microscopy, a fungus ball (FB) is described as a matted, dense conglomerate of hyphae separated from but adjacent...
to the respiratory mucosa of the sinus without invasion or granulomatous reaction. The haematoxylin-eosin staining confirms the absence of an allergic mucin. The sinus mucosa is free of hyphae even at the Gomori-methenamine-silver stain [6].

FB is the most prevalent presentation of NIFRS [1-5] in Europe. The disease is typically encountered in immunocompetent individuals (average age: 64 years) with a considerable female preponderance [1-7]. It is defined as the sequestration of a fungus within a solitary paranasal cavity [6]. The most common site is the maxillary sinus (94 per cent) followed by the sphenoid sinus, ethmoid sinuses (3 per cent) [1, 3, 6, 7]. The frontal sinus is commonly involved secondarily in association with other sinuses. Isolated frontal sinus fungus ball is extremely rare with only 29 reported cases confirming the low prevalence of the disease [1, 3, 7-21]. According to Dufour et al., frontal sinus fungus ball represents only 1.1 per cent of sinus fungus ball cases [1] Kloss et al., found approximately the same percentage with a value of 2 per cent of all cases of paranasal sinus fungus ball [7].

The pathogenesis of fungus ball remains unclear. Two ways of entry of the fungus have been suggested: 1. the odontogenic pathway and 2. the so-called "aerogenic" pathway.

The first author to point out the relationship between the teeth and fungal infection was Pr. Legent in France in 1989 [22]. Since then there has been an increasing amount of literature demonstrating that treated teeth with overextension of the root canal sealer or solid materials such as gutta-percha or silver cones into the sinus might be the main etiological factor for aspergillosis of the maxillary sinus in healthy patients [23]. Nevertheless this way of infestation is unlikely in case of isolated frontal sinus involvement. The second portal of entry (the aerogenic pathway) seems more relevant to explain the frontal sinus localization. Fungal spores are inhaled into the sinus through the natural ostium and find favourable condition (anaerobic condition) for their growth and subsequent sequestration [1, 3, 7, 12].

The frontal sinus has actually some anatomical particularities that can contribute to the development of a fungus within its cavity. It is the least accessible paranasal cavity. Its ostium is located in the anterosuperior part of the nasal cavity at the end of a long and funicular pathway [24]. When the sinus cavity is well-pneumatized, it can play the role of a reservoir in which the fungus can grow easily and slowly without any symptoms. Yet, the presence of fungi in the sinus cavity can stimulate an inflammatory reaction with secondarily hyperplasia of the mucosa, decrease of the mucociliary clearance and obstruction of the ostium. The symptomatology of frontal sinus fungus ball is commonly vague, subtle and non specific unless complications occur. The patient is asymptomatic for a long period of time or presents with nose bleeds [20] or mild to moderate facial pain [8-11,15,16]. In our 2 cases, the patients reported some fullness in the forehead and upper eyelid region. In some cases, the disease is associated with osteomyelitis of the frontal wall (Pott’s puffy tumor) [14] and gives the change for a mucocele or a malignant tumor [9,10,17]. The disease is potentially lethal: orbital [10,19-21] or intracranial [8-10] complications have been reported.

The diagnosis of the fungus disease is made with the combination of the physical examination (nasal endoscopy), the imaging, and the histological examination. Black concretions in the middle meatus are pathognomonic of a fungus sinus ball but this sign is often absent [7]. The sinus CT scan displays a partial to complete, heterogeneous, opacification of the frontal sinus. Micro calcifications and/or metallic dense spots can be observed associated with a sclerosis of the bony sinus walls. Bony erosion may be present as well [1, 3, 7, 25-27]. It is commonly due to pressure atrophy and/or dissolution by products of inflammation like proteolytic enzymes and cytokines. In such a case, it is not indicative of invasive fungal rhinosinusitis. MR images depict a nodular and heterogenous iso- or hypointensity on T1-weighted images and a marked hypo intensity on T2-weighted images that does not enhance after injection of gadolinium, in contrast to an enhancement of the inflammation adjacent to the sinus mucosa [27,28].

The definitive diagnosis of fungus ball is made by histological analysis of the surgical samples. Morphologically, Aspergillus sp. are described as thin, septated, acute-angled (45°) or dichotomous branching hyphae.

The sensitivity and specificity of the histological investigations are significantly higher than those of the mycological analysis performed on a routine basis on Sabouraud dextrose agar [29]. According to the literature, only 23-50 per cent of fungus ball cultures are positive. In our cases the culture was negative in both cases.

The treatment of a SFB is exclusively surgical. It consists of a complete debridement of the fungal mass, preservation of the healthy mucosa and re-establishing the drainage from the affected sinus. This can be performed by either an open [10-11, 19-21] (osteoplastic flap) or endonasal approach [12,13]. In our cases we used the endonasal endoscopic approach (complete uncinectomy and opening of the bulla ethmoidalis) associated to frequent intraoperative irrigations of the affected sinus with saline at moderate pressure. Gupta et al., used the same approach (type II frontal sinusotomy) for the primary surgery and recommended a type III frontal sinusotomy for the revision surgeries [12]. In the literature the first author who used the type III frontal sinusotomy for this indication was Kodama [13].

Antifungal therapy is not required as the patients are immunocompetent.

CONCLUSION

Frontal sinus fungus ball is a rare entity. It is encountered in immunocompetent. The symptomatology is extremely variable, subtle and non-specific. However the disease is potentially lethal. The definitive diagnosis is based upon the combination of nasal endoscopy, imaging and histological investigation. Surgery is the treatment of choice. It consists of a complete debridement and restoration of drainage pathway of the frontal sinus. The endonasal approach (type II frontal sinusotomy) is an elegant and successful alternative to an open approach with a lower morbidity and quicker rehabilitation. Frequent intraoperative irrigations of the operated cavity are very helpful to clean all the fungal debris and avoid any recurrence.

REFERENCES


