Frontal Sinus Fungus Ball: a rare case and literature review

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Abstract

Aims: This article describes the report of a rare case of fungal ball in the frontal sinus in an adult male and discuss about the variables of this condition related to the patient. Additionally, this study aims to identify the state of the art of fungal ball in paranasal sinuses, especially in the frontal sinus. Method: We performed a literature review on fungal ball in the paranasal sinuses and described a rare case of fungal ball in the frontal sinus, which is listed as the paranasal sinus most rarely affected by fungal ball. Result: All reported cases of fungus ball in the frontal sinus affected male patients, contrary to the common prevalence of females in fungal ball of the other paranasal sinuses. Of the total of 8 patients with fungal ball in the frontal sinus reported, 40% had unilateral disease and 60% bilateral, also contrary to the incidence data of the other paranasal sinuses, with unilateral prevalence. However, with the present study, this index changes, with 50% unilateral and 50% bilateral in frontal sinus involvement. The average age of cases reported in the frontal sinus is 65.29 years, with a minimum age of 61.16 and a maximum of 69 years. As for the etiologic agent, Aspergillus sp., the endonasal endoscopic therapeutic approach corresponded to 80% of cases, while frontal osteoplasty accounted for 20% of cases, reaffirming this prevalence data from other studies. Conclusion: Despite being a low incidence entity, frontal sinus fungal ball should be considered in patients with pain in the frontal region refractory to the usual clinical treatments. We hope to contribute to the knowledge of this presentation as a differential diagnosis and reinforce the importance of thorough clinical investigation.

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Structured Abstract

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Keywords: Aspergillus fumigatus; Infectious Diseases;Rhinosinusitis and Complications; Paranasal Sinus Diseases; Transanal Endoscopic Surgery .

Introduction

The sinus fungal ball is defined as an agglomeration of debris and hyphae within the paranasal sinus, commonly affecting a single sinus[1]. It most often affects the maxillary sinus, followed by the sphenoid and ethmoid sinus, and frontal sinus affection is extremely rare implicated in only about 2% of all involved sinus[2, 3, 4]. There is a slight female predominance[5] and, in most cases, it manifests itself during the fourth and fifth decade of life, as a result of respiratory infection by *Aspergillus sp* [6].

Aspergillus fumigatus and *Aspergillus flavus* [6] are the main species involved in fungal rhinosinusitis. *Aspergillus* is a genus of anamorphic fungi, which is characterized by having filamentous fungi with hyaline hyphae, septate and branched at an acute angle[7]. They dominate warmer areas, being more common in tropical countries. Several species of *Aspergillus* have importance for humans due the ability to produce toxic metabolites, mainly *A. fumigatus* , which has a fast-growing, bluish-green, and powdery colony. *A. flavus* , on the other hand, is a filamentous fungus that can produce aflatoxins and cyclopiazonic acid that results in atoxic effect on the hosts[8,9].

Fungal rhinosinusitis comprises between 4% and 10% of surgical interventions in the paranasal sinuses region and can be subdivided into invasive and non-invasive[10, 11]. Non-invasive fungal rhinosinusitis is typical of immunocompetent patients[12], classified as a fungal ball or allergic fungal rhinosinusitis[13].

The slow and oligosymptomatic development of fungal ball in the frontal sinus often leads to late diagnosis, with orbital or intracranial complications as the first clinical signs[14]. The diagnosis of fungal ball is difficult, occasionally identified by Computed Tomography (CT) or Magnetic Resonance Imaging (MRI)[5, 15] and surgery with an endonasal endoscopic approach is the treatment of choice[2, 16].

Methods

It is a literature review and case report, written based on the PRISMA-E 2012 reporting guide (Preferred Reporting Items for Systematic Reviews and Meta-Analyzes)[17]. The PICO system (Population, Intervention, Comparison and Outcome) was followed and no specific protocol was used for its elaboration.

The PubMed, ScienceDirect, BIREME (Lilacs and Medline) and Scielo databases were used. The keyword "fungus ball" combined with "paranasal sinuses", "fungal rhinosinusitis", "frontal sinus" was used.

The inclusion criteria were prospective and retrospective studies on fungal rhinosinusitis and fungal ball in human paranasal sinuses, these being case reports and review articles, respectively. The exclusion criteria were articles in vitro, in animal models, or that did not cover the central theme of the research.The articles were extracted in duplicate and these were stored in EndNote - Clarivate Analytics.

Results

203 articles were found in PubMed, 889 articles in ScienceDirect, 172 articles in BIREME and 3 articles in Scielo based on the research strategy used. Inclusion and exclusion criteria were applied, removing articles that did not meet the needs of the research, being selected 1 article from the Scielo database, 2 from BIREME and 18 articles from PubMed, totaling 16 articles on fungal ball in maxillary paranasal sinuses, sphenoid and ethmoid and 5 articles in frontal sinus. According to the PRISMA-E 2012 reporting guide, the information on stages' review process can be found in Figure 1.

Literature review

The state of the art on fungal ball in paranasal sinuses shows the prevalence of fungal ball in paranasal sinuses in the age group of 50 to 60 years. Prospective studies reaffirm the prevalence of fungal ball in the maxillary sinus, followed by the sphenoid, ethmoid and, finally, the frontal sinus, recommended by retrospective studies, as well as the higher incidence being unilateral and in the female sex, with the most prevalent etiological

agent being *Aspergillus spp.* The therapeutic approach of choice, unanimously, is the endonasal endoscopy in any affected paranasal sinuses.

The reported cases of fungal ball in the frontal sinus affected male patients, contrary to the common prevalence of female sex in a fungal ball of the other paranasal sinuses. Applying statistical data, it appears that of the 8 patients with reported fungal ball in the frontal sinus, 40% had unilateral affection and 60% bilateral, also contrary to the incidence data of the other paranasal sinuses, with unilateral prevalence. However, with the present study, this index changes, with 50% unilateral and 50% bilateral in frontal sinus involvement.

As for the etiologic agent, *Aspergillus sp.* the endonasal endoscopic therapeutic approach corresponded to 80% of cases, while frontal osteoplasty accounted for 20% of cases, reaffirming this prevalence data from other studies. The average age of reported cases is 65.29 years, with a minimum age of 61.16 and a maximum of 69 years. The summary of data referring to reports of fungal ball in the frontal sinus are listed in Table 1.

Case report

63-year-old immunocompetent man, with no comorbidities, has as main complaint pain, in the forehead, predominantly left, with a stabbing character, starting two months ago, with no evolution. During this period, he used antibiotics, anti-inflammatories, and topical and systemic corticosteroids, with no improvement in his condition.

The initial hypotheses that guide the request for the first exams were nasal tumors, recurrent acute and chronic rhinosinusitis.No abnormalities in blood and serological tests. The endoscopic examination of the nasal cavity revealed mucosal edema with purulent secretion of the ostiomeatal complex. CT scan of the paranasal sinuses identified veiling of the frontal sinus with microcalcifications, hyper-attenuating areas, associated with bone wall sclerosis, suggestive of fungal bolus (Figures 2 and 3). As these CT findings are typical of a fungal ball, there were no differential diagnoses.

The patient underwent maxillary sinusectomy, wide ethmoidectomy and frontal sinusectomy like DrafIIa, with complete removal of the lesion (Figure 4). The microbiological examination showed sparse fragments of mucosa covered by respiratory epithelium, with edema of the lamina propria and a marked inflammatory infiltrate (lymphocytes, neutrophils, histiocytes and plasma cells). Abundant septate fungal hyphae were dichotomized at an acute angle, compatible with *Aspergillus sp* ., with no signs of malignancy in the present material and the search for fungi by Grocott stain was positive. Biopsy of the healthy frontal sinus mucosa confirmed negative non-invasive fungal ball.

There were no intraoperative and postoperative complications. Currently, the patient has an 8-month follow-up and complete improvement of the condition.

Discussion

Globally, the prevalence of unilateral involvement of paranasal sinuses is higher than bilaterally. However, with the present study, this index, related to the frontal sinus fungal ball, is equivalent, corresponding to 50% of unilateral involvement and 50% of bilateral involvement, perhaps due to some anatomical alteration of the frontal sinus.

All reported cases of fungal ball in the frontal sinus affected male patients, contrary to the common female prevalence in fungal ball of the other paranasal sinuses; a possibility for this gender difference may be hormonal. The average age of the reported cases is 65.29 years, with a minimum age of 61.16 and a maximum age of 69 years, exceeding the age range common to other fungal balls with paranasal sinuses; the fact that it occurs in older patients may be related to the delay in the proliferation of fungi, reaching more this age group[21].

The aerogenic hypothesis[22] suggests that fungal spores are deposited on the mucosa by inhalation and acquire pathogenic capacity when in anaerobic conditions within the sinus[23]. Other authors indicate osteomeatal complex obstruction or chronic rhinosinusitis as predisposing[24]. However, this theory does not explain the cases of fungal ball that affect the sphenoid or frontal sinus. Not all patients with occluded frontal sinuses

develop a fungal ball, which probably means that spores are not always able to reach the frontal sinus due to the complex anatomy of the frontal recess.

Of the several anatomical sinonasal variants, the presence of bullous shell was significantly associated with the development of fungal ball, as well as a narrow infundibulum and anatomical variations in the region of the ostiomeatal complex, known to cause sinus hypoventilation, may also be related to this pathogenesis[25]. Concomitant to this, our patient also had fronto-ethmoidal cells that obstructed the frontal recess, which may explain the pathophysiological mechanism.

The therapeutic approach of choice is the endonasal endoscopy in any paranasal sinuses, as the pharmacological treatment does not result in improvement of the condition. The endonasal endoscopic therapeutic approach corresponded to 80% of cases with frontal osteoplasty reserved only for those in whom the endonasal approach is not possible.

Many important neurovascular structures are adjacent to the frontal sinus, putting the patient at risk for orbital and intracranial complications. Thus, the presence of a fungal ball in the frontal sinus, although non-invasive, is potentially much more serious when compared to the involvement of the other sinuses. Thus, early diagnosis and surgical intervention are essential.

Conclusion

Despite being an entity with low incidence, the frontal sinus fungal ball should be considered in patients with pain in the frontal region without improvement with usual clinical treatments.

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Reference	Patients with fungal ball in frontal sinus	Mean age at diagnosis	Affected paranasal sinus	Affection (unilateral or bilateral)	Etiological agent
Seo et al. (2019)[18]	1	61,16 years	Front	Unilateral (left)	Aspergillus spp.
Bernardini et al. (2017)[2]	2	62,5 years	Front	Bilateral	Aspergillus fumigatus
Wei xin et al. (2016)[19]	2	-	Front	Bilateral	Aspergillus spp.
Popko, A Broglie e Holzmann (2009)[14]	2	68,5 years	Front	Bilateral	Aspergillus fumigatus
Stevens (1978)[20]	1	69 years	Front	Unilateral (right)	Aspergillus fumigatus