What’s in a fungus ball? Report of a case with submucosal invasion and tissue eosinophilia

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Abstract
Fungus balls are tangled mats of hyphae that are often found in the maxillary sinus. In approximately half of affected patients, radiologic evaluation will reveal areas of hyperdensity within soft-tissue masses. Histopathologic examination will reveal no invasion of the mucosa and no granulomatous reactions. Surgical removal is sufficient because fungus balls are not known to recur. We describe an interesting case of a sinonasal fungus ball that resembled dental filling material on radiologic imaging because of its extraordinary radiopacity. Histopathologic examination detected eosinophilic infiltration, hyphae in the submucosal tissues, and tissue necrosis.

Introduction
The introduction of the concept of allergic fungal rhinosinusitis and the publication of recent studies showing a high incidence of fungal matter in histologic specimens taken from patients with chronic rhinosinusitis have led to a good deal of debate lately. Clinically, a patient’s immune status determines the degree of fungal infection of the paranasal sinuses. Patients with invasive fungal rhinosinusitis are usually immunocompromised, and therefore they are likely to experience potentially fatal complications. On the other hand, allergic fungal sinusitis is thought to be the result of an atopic reaction to the causative fungus, and most patients with fungus balls are immunocompetent. Growth of saprophytic fungi has also been seen on the mucosa and crusts in the sinonasal cavity of immunocompetent patients following sinus surgery.

Fungus balls are tangled mats of hyphae that are frequently found in one sinus only, most often the maxillary sinus. In the rhinologic literature, they are often referred to as mycetomas. In approximately half of affected patients, radiologic evaluation will reveal areas of hyperdensity within soft-tissue masses. These areas are believed to represent dense hyphae or metallic depositions. Histopathologic examination will reveal no invasion of the mucosa and no granulomatous reactions. There has been 1 report of a fungus ball that became invasive after the patient became immunosuppressed as a result of a kidney transplant. Surgical removal is sufficient because fungus balls are not known to recur.

In this article, we describe an interesting case of a sinonasal fungus ball that resembled dental filling material on radiologic imaging because of its extraordinary radiopacity. Histopathologic examination detected eosinophilic infiltration, hyphae in the submucosal tissues, and tissue necrosis. These findings perhaps represented the initial phase of an invasive progression.

Case report
A dentist had treated the upper left canines of a 64-year-old woman. Shortly thereafter, the woman began to experience left facial pain. The dentist obtained a panoramic x-ray, which demonstrated a bright lesion in the left maxillary sinus (figure 1). Believing that this lesion probably represented dental paste in the sinus cavity, the dentist referred the patient to our facility for sinus exploration. The referral was made 3 months following the original dental procedure.

Findings on physical examination were normal except for the presence of left infraorbital pain on pressure. Computed tomography (CT) of the paranasal sinuses revealed the presence of a large concha bullosa on the left, mucosal thickening on the left, and a very bright 3 × 5-mm mass in the left maxillary sinus (figure 2). The brightness of the lesion was almost identical to that of the patient’s dental fillings, suggesting that they might very well have been made of the same material.

We advised the patient that because the lesion was large enough to eventually obstruct the sinus ostium, surgery was justified, and the patient consented. We approached the left maxillary sinus endoscopically through the enlarged...
natural ostium. The lower third of the maxillary sinus was filled with polypoid mucosa that peeled off easily from the underlying mucoperiosteum. In the center of the polypoid tissue was a brown, crusty lesion that was macroscopically identified as an *Aspergillus* mycetoma. The polypoid tissue also featured an area of pigmentation (figure 3). Histopathologic examination of the tissues revealed extension of hyphae into the submucosa and the presence of submucosal necrosis, findings that are consistent with aspergillosis (figure 4). A complete blood analysis failed to reveal any immune system failure.

Discussion
There are a few interesting aspects of this rather simple and straightforward case that make its publication worthwhile. The brightness of the maxillary sinus lesion and the patient’s recent history of dental work suggested that the mass might very well have been a foreign body. If that had been the case, would surgical intervention have been necessary to remove it? We based our surgical indications on the size of the lesion (which was large enough to obstruct the sinus ostium) and the mucosal reaction around it (a potential cause of chronic sinusitis). If the patient had been unfit for surgery or if she had refused it, we probably would have opted for a wait-and-see approach. However, because the patient was in the initial phase of an invasive process, this would have proved to be the wrong decision.

The literature contains several reports on the CT diagnosis of fungal sinusitis and the effects of zinc oxide-eugenol paste. Some authors strongly believe that CT studies—especially CT densitometry—are important tools in making the differential diagnosis of fungal sinusitis and foreign bodies of dental origin in the sinus; others claim that there are no valid predictive criteria for CT diagnosis. However, none of these authors has commented on the necessity of CT to validate surgery. An effective method of differentiating fungal growth from a foreign body preoperatively would be very useful, especially in cases of unsuspected submucosal invasion, such as occurred in our patient.

Articles on the effects of zinc oxide-eugenol paste in the sinus are more interesting from a rhinologist’s point of view because some authors suggest that it is a cause of fungal growth. This notion is not commonly recognized by otolaryngologists, and perhaps it deserves to be better researched by sinus surgeons. Although some authors believe that zinc is both the cause of aspergillus growth in the sinus and the reason for radiopaque foci, one controlled study on the effects of zinc oxide-eugenol paste failed to show that it acts as a growth factor for *Aspergillus*. In our literature review, we did not find any studies on what constitutes the radiopacity of fungal masses in the sinuses. Sulfur granules or calcifications have been suggested as possible causes.

The mucosa surrounding the fungus ball in our patient was sent for histopathologic evaluation. Areas of fungal growth in the mucosa could be seen macroscopically. Microscopic examination revealed that what had appeared to be submucosal invasion was mostly hyphae and spores covered by necrotic epithelium and eosinophils; there were also areas of submucosal hyphae extension and spores.
This case represents a rather unusual expression of all three possible types of fungal sinusitis in 1 patient (a fungus ball extending submucosally with a strong eosinophilic reaction around it). To our knowledge, no such case has been published before. The reason might be that most fungus balls are easily diagnosed macroscopically, so histopathologic examination is not usually performed. Whatever the case may be, histopathologic examination of every specimen removed from the sinuses appears to be necessary if we are to understand the involvement of fungal pathogens in rhinosinusitis. This single case, with all its complexities, provides a good lesson in fungal rhinosinusitis, and it illustrates how much more we have to learn about it.

References