Maxillary Sinus Mycetoma Associated With Hypersensitivity To Mucor Racemosus

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Citation

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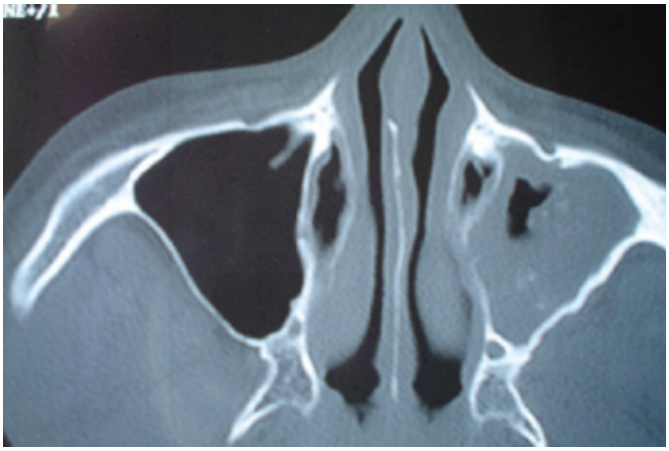
Abstract

Mold hypersensitivity may play a role in chronic sinusitis. Although mucor has been identified in allergic fungal sinusitis, to our knowledge this is the first reported case of mucor associated mycetoma in association with immediate hypersensitivity to mucor.

CASE REPORT

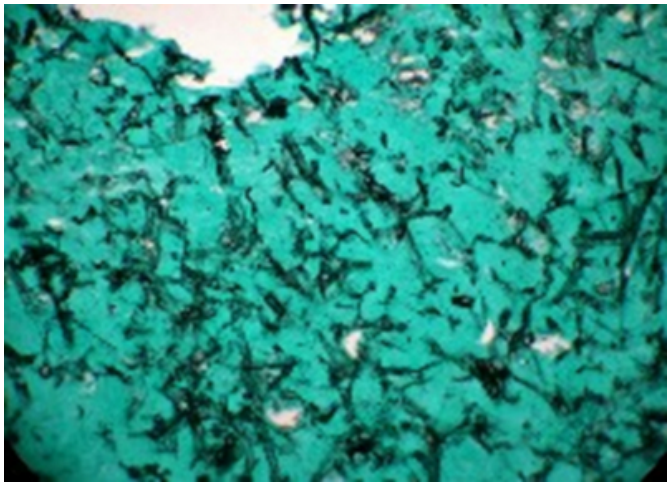
In the beginning of 2005, a 39 year old Chinese male presented to the St Vincents Hospital otolaryngology clinic with a 10 year history of nasal congestion with the left side of the nose being more affected. He had a nasal operation in 1996 in China, the country of his birth. His work experience included working in a bread factory in China. He immigrated to the United States in 2003. En route, he was allergy tested in Japan where he was told that he had many allergies. The patient also complained of a foul smell in his nose and occasional yellowish discharge from the left side of his nose. The patient denied other medical problems including diabetes, immunodeficiencies, and asthma. On endoscopy an enlarged left uncinate process, and polypoid tissue in the area of the left osteomeatal complex were noted. The patient was treated with mometasone nasal spray, oral loratadine, and amoxacillin/clavulinate without any improvement in symptoms. A maxillofacial CT scan showed extensive left maxillary sinus disease. Soft tissue density was noted in the left maxillary sinus with higher density areas within. There was widening of the left osteomeatal complex (Figure 1).

Figure 1



The other paranasal sinuses were normal. A complete blood count, basic blood chemistries, and liver panel were within normal limits. There was no increase in blood eosinophils. In March 2005, the patient underwent sinus surgery where a left antrostomy/uncinectomy and left maxillary sinus evacuation were performed. The contents in the maxillary sinus consisted of yellowish to greenish, inspissated mucoid material. Tissue specimens revealed inflammation with some eosinophil infiltration but no eosinophilic staining mucin. There was also some tissue necrosis, and presence of fungal hyphae collections(Figure 2 GMS fungal stain. Original magnification X40).

Figure 2



Fungal cultures however revealed no growth. Post-operatively the patient was evaluated for aero-allergies. The total IgE was 83 IU/mL. Specific IgE was present for several common pollens including orchard grass, common ragweed, birch, maple, pecan tree, white ash, and oak. All allergen specific IgE levels were greater than 1 kIU/mL as determined by the Immunocap ™ method. A lower amount of specific IgE(0.46 kIU/mL) to Dermatofagoides farinae(dust mite) was also noted. Because of the fungal elements noted on histopathology, extensive fungal aeroallergens were studied. No specific IgE was noted to Alternaria tenius, Aspergillus fumigatus, Candida albicans, Cladosporium herbarum, Echinococcus purpurascens, Erythronum purpurascens, Fusarium moniliforme, Helminthosporum halodes, or Penicillium notatum. However, the specific IgE to Mucor racemosus was increased at 2.14 kIU/mL. A complete blood count, erythrocyte sedimentation rate, and c-reactive protein were within normal limits at that time. The CD4 and CD8 T-lymphocyte counts were also within normal limits. A culture of nasal secretions showed no fungal growth. No nasal eosinophils were observed. The patient continued to have intermittent nasal symptoms during the year of follow-up treatment which included topical nasal corticosteroids and 2 further courses of oral antibiotics.

DISCUSSION

Sinus mycetomas are considered a non-invasive form of fungal sinusitis([1]). They are usually not associated with immunosuppressed hosts. The most common causes are Aspergillus fumigatus and dematiaceous fungi. Unlike allergic fungal sinusitis, immediate hypersensitivity and other atopic manifestations are less common. Furthermore mycetomas are usually not bilateral. Non-invasive mucormycosis of the sinuses is uncommon([2]). At least one non-invasive case has shown calcification([3]). To our knowledge the presence of specific IgE to Zygomycetes in fungal mycetoma has not been reported previously. As inhalation is the usual route of infection for Zygomycetes, it is of interest that the patient previously worked in a bread factory, where fungal species may have been present in high concentrations. Despite the fact that the patient had no fungal culture growth of the sinus tissues, the histopathology and serological findings support a mycetoma due to mucor. Difficulties in culturing tissue specimens for Zygomycetes are well known([2]). The patient had obvious proclivity towards aeroallergy as evidenced by the increase in multiple pollen specific IgE levels. This atopic diathesis likely resulted in elevated specific IgE to mucor due to continuous exposure from the mycetoma. It is notable that hypersensitivity to other commonly inhaled fungi such as Alternaria, Cladosporium, and Aspergillus were not present in this patient. Hypersensitivity to mucor is decidedly less common than hypersensitivity to these 3 molds, even in an atopic population([3]).This suggests that the patient did not have a predilection towards fungal allergy. Mold-allergic individuals are often sensitive to several mold species([5]) and there appears to be cross-hypersensitivity to some of the major molds([6]). This patient did not have any mold specific IgE except to mucor.

Although mold allergy studies has not been extensively studied in China, at least one report([7]) finds that prevalence of similar mold spores such as those that are prevalent in other Western countries including Alternaria, Cladosporium, and Aspergillus. A search of index medicus revealed no published reports of mucor being a prevalent mold spore in China.

Allergic mucin in response to fungi are felt to be an essential diagnostic feature in allergic fungal sinusitis([2]). Allergic mucin was not observed in the herein described patient. It is conceivable that this patient had an incomplete form of allergic fungal sinusitis, despite not having allergic mucin, or sheets of eosinophils which are commonly described in allergic fungal sinusitis. The diagnostic criteria for allergic fungal sinusitis may be evolving([8]).

Mold hypersensitivity may also play a role in chronic sinusitis([9]). Although mucor has been identified in allergic fungal sinusitis([10]), to our knowledge this is the first reported case of mucor associated mycetoma in association with immediate hypersensitivity to mucor.

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