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Large Mycetoma of the Maxillary Sinus

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Mycetoma of the paranasal sinus is defined as noninvasive fungal sinusitis and frequently occurs in immunocompromised hosts. Diagnosis of mycetoma is mainly based on histopathology because fungal culture tests are frequently negative. The maxillary and sphenoid sinuses are most often involved. We studied a large mycetoma of the maxillary sinus in a patient who was not immunocompromised but had previously undergone functional endoscopic sinus surgery. A large calcified black-brown spore about 4 cm × 4 cm × 3 cm in size was detected over the right maxillary sinus. The calcified spore was cut into small pieces and removed endoscopically. Examination of the spore revealed many fungal hyphi with acute angles, which was compatible with aspergillosis. The condition did not recur during the two-year follow-up period.
Key words: mycetoma, fungus sinusitis, maxillary sinus

INTRODUCTION

Fungus ball, also known as mycetoma, of the paranasal sinuses is a noninvasive chronic fungal rhinosinusitis that usually occurs in immunocompromised patients[1]. Fungus balls are usually less than 2 cm in diameter. Fungal rhinosinusitis is classified into invasive and noninvasive forms depending on the presence or absence of a fungal agent in the mucous membrane, bone, or vesse[1]. Mycetoma of the paranasal sinus is noninvasive[2]. Several clinical presentations, which vary from acute infection of the sinus cavities to an asymptomatic form, have been reported[1-3]. Nevertheless, the most frequent clinical presentation is nonspecific: symptoms include nasal obstruction, purulent nasal discharge, facial pain, and chronic coughing. In such cases, only unilaterality may alert the clinician to the possibility of a mycetoma.

Endoscopic findings are usually normal, but edema or purulent secretions may be observed[1-6]. The characteristic computed tomography scan presentation includes typical heterogenous opacities associated with mottled hyperdense foci in the sinus cavity[7]. The maxillary and sphenoid sinuses are the most commonly infected, but mycetoma also occurs in frontal, ethomoid, and pansinus locations.

The aspergillus fungus is the most frequent cause of mycetoma[8]. This report concerns an unusual case of a large mycetoma that was identified by histopathology, treated by functional sinus surgery (FESS), and did not recur during the two-year follow-up.

CASE REPORT

A male patient aged 56 years presented with a main complaint of intermittent nasal obstruction and headache, which he had experienced for three years. The patient had suffered chronic right paranasal sinusitis for several years. He underwent FESS in a teaching hospital during October 2003. However, nasal obstruction, purulent discharge, postnasal drip, and headache developed in the months following surgery. He was admitted for further investigation and treatment in May 2004. There was no evidence of hepatitis, hypertension, hyperlipidemia, or other systemic disease. Physical examination revealed mucopus and crusts in the right nasal cavity. Multiple antigen simultaneous tests (MAST) revealed no specific allergy. Computed tomography scanning of the sinus revealed a large calcified mass with thickened mucosa over the right maxillary sinus (Fig. 1). Recurrent chronic paranasal sinusitis of the right side was observed. The patient underwent revisionary FESS. A large calcified black-brown spore about 4 cm × 4 cm × 3 cm in size was detected over the right maxillary sinus. After it was cut into pieces, the calcified spore was removed endoscopically. Examination of the spore revealed many fungal hyphi with acute angles (Fig. 2), which were compatible with aspergillosis.

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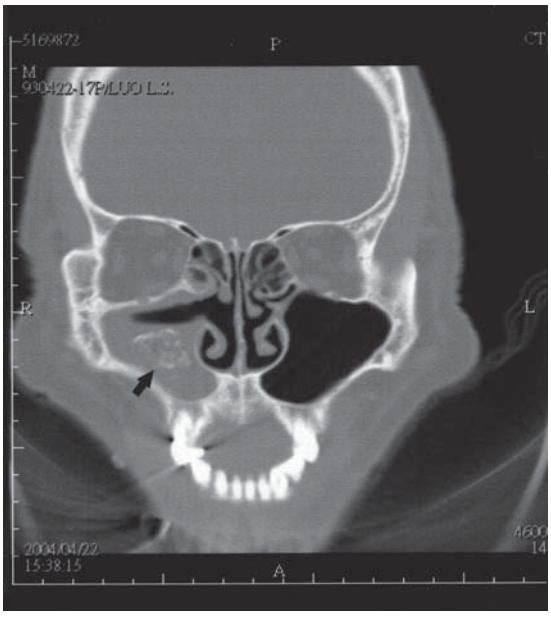

Fig. 1 Computer tomography scanning (CT) of the sinus showed a large calcified mass (arrow) with thickened mucosa over the right maxillary sinus.

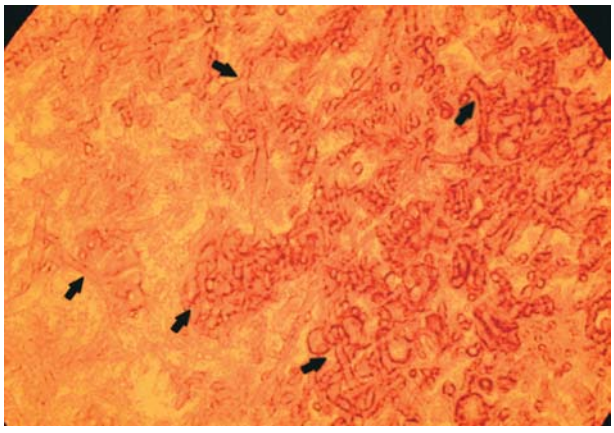

Fig. 2 Many fungus hyphi (arrows) with acute angles were noted. (H&E 200X).

DISCUSSION

Five diagnostic categories of fungal rhinosinusitis disorders are currently recognized[9], and can be differentiated using histopathology. Clinical presentations are usually characteristic. Three types of fungal rhinosinusitis cause true tissue-invasive infectious diseases: acute necrotizing fungal sinusitis, chronic invasive fungal sinusitis, and granulomatous invasive (indolent) fungal rhinosinusitis. Two forms of fungal rhinosinusitis are not tissue-invasive: fungus ball (sinus mycetoma) and allergic fungal sinusitis (AFS). There is no accurate epidemiological data for sinus mycetoma in Taiwan, including its incidence and fungal etiology. However, a local study revealed that aspergillus accounted for 90.7% of mycetomas and 38.4% of acute invasive fungal sinusitis cases[10]. The most commonly infected sinus is the maxillary sinus (86.2%), followed by the sphenoid sinus (7.3%)[11]. In our patient, there was no fungal invasion of the sinus mucosa, indicating that he did not have allergic fungal sinusitis. The MAST analysis revealed that he did not have allergic reactions to fungi, including aspergillus. If AFS were present, the MAST test would have been positive. Therefore, we made a diagnosis of sinus mycetoma. After discharge, the patient was referred to an immunospecialist who examined his immunological status and judged it to be uncompromised.

Mycetoma of the paranasal sinus is a noninvasive fungal sinusitis that frequently occurs in immunocompromised hosts. Sinus mycetoma is an extramucosal accumulation of the many degenerating fungal hyphae into a ball within the sinus cavity; usually only one sinus is involved. Low-grade chronic inflammatory infiltrate fibrosis of the sinus mucosa is often present. Diagnosis of mycetoma is mainly based on histopathology because fungal culture tests are frequently negative. The maxillary and sphenoid sinuses are most frequently involved. In our case, the maxillary sinus was involved.

Although previous sinus surgery, oral-sinus fistulation, and previous cancer chemotherapy have been identified as risk factors for this disease[1,6], risk factors are often absent[6,9]. If a risk factor such as an oral-sinus fistula is found, surgical repair may be required to prevent recurrence. Although most mycetomas occur in immunocompromised patients, our patient was not immunocompromised. In this case, the disease may have been caused by previous surgery, as he had undergone FESS in another clinic one year before presenting with sinus mycetoma.

Surgical removal of the fungal ball and adequate resection of the associated obstructive or diseased sinonasal mucosa seems to be curative[1,5]. Since 1980, FESS has become popular for treating surgical cases of sinus pathologies. In the hands of expert surgeons, morbidity associated with FESS is low and the rate of complications is similar to that of the Caldwell-Luc procedure. Our case of mycetoma was treated successfully without complications or recurrence using an endonasal approach, which supports previous findings on the reliability and safety of endoscopic procedures.

The degree of hydration of the fungus ball as well as the thickness of the sinus concretions may play a role in the ease of removal of the fungus ball. In this case, complete removal of the fungus ball was achieved after it was cut into small pieces. In sinus mycetoma, irrespective of the location, a single procedure for removing the fungus ball without any local or systemic antifungal treatment is sufficient to cure the patient. The patient attended regular follow-up consultations in our outpatient department and did not display any signs of recurrence.

Allergic fungal sinusitis is increasingly recognized as a chronic, recurring hypersensitivity disease. It is not a tissue-invasive fungal process, but consists of an allergic/hypersensitivity response to the presence of extramucosal fungi within the sinus cavity. Screening of AFS patients with quantitative immunoglobulins should reveal that they are clinically immunocompetent; delayed hypersensitivity skin tests for common T-cell recall antigens and antibody-response tests for polysaccharide and protein immunizations may be performed for further confirmation if required[12]. Total serum IgE levels are usually elevated but can be normal[12-14]. Therefore, a specific IgE assay is better than a total IgE assay for differential diagnosis between AFS and mycetoma. Although total serum IgE analysis was not performed on our patient, the specific IgE tests were negative.

In conclusion, this mycetoma was caused by previous sinus surgery. As there was no invasive lesion of the sinus mucosa, it was not an invasive type of fungal sinusitis. Because the patient was negative for specific IgEs, including an IgE that recognizes aspergillus, AFS could be ruled out. FESS was a satisfactory treatment for this case of mycetoma of the sinus.

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