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A case of invasive paranasal aspergillosis that developed from a non-invasive form during 5-year follow-up

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Abstract

Invasive paranasal aspergillosis has been previously reported, but there have been no case reports of non-invasive paranasal aspergillosis that progressed to the invasive form during follow-up. A case of non-invasive aspergillosis of the maxillary sinus that appeared to become invasive during 5-year follow-up is reported. The patient was diagnosed as having non-invasive aspergillosis at the first visit because CT images revealed only mucosal thickening and calcifications in the right maxillary sinus. Five years later, CT images showed an invasive mass lesion in the orbit and large bone destruction of the posterior wall of the maxillary sinus. The patient was rescued by total removal of the orbital contents with zygomatic ostectomy followed by appropriate antifungal chemotherapy irrespective of residual tissue invasion. We would propose that appropriate surgical treatment and antifungal agents are necessary to improve the prognosis of invasive aspergillosis.

Keywords: Invasive aspergillosis; Non-invasive aspergillosis; Maxillary sinus; Surgical treatment; Antifungal agent

1. Introduction

Paranasal aspergillosis is classified into invasive and non-invasive forms. Non-invasive aspergillosis is the most common and occurs as chronic sinusitis that is unresponsive to conservative medical treatment. A fungus ball of Aspergilli is present in a sinus, usually the maxillary sinus. On histology, tangled mycelia of Aspergilli with inflammation are seen [1]. Invasive aspergillosis is an aggressive infection that is commonly found in immunocompromised individuals and usually occurs by contiguous spread from the paranasal sinuses into the orbit [2]. Moreover, the superior orbital fissure and optic canal open directly into the middle cranial fossa and are easy pathways for further intracranial spread of infection. Thus, the clinical course of the invasive form is quite aggressive, and half of the patients die from disease due to treatment failure [3]. It has been postulated that the severity of paranasal aspergillosis is related to the duration of the disease, and the non-invasive form may become invasive over time [1,4–6]. However, there have been no prospective case reports of the non-invasive form progressing to the invasive form during follow-up. A case of invasive aspergillosis of the maxillary sinus that appeared to develop from the non-invasive from over 5 years of follow-up is reported.

2. Case report

In May 2000, a 74-year-old Japanese woman who had neither diabetes nor immunosuppressive diseases visited our clinic with symptoms of bilateral mild nasal obstruction. CT images revealed mucosal thickening and multiple mottled calcifications in the right maxillary sinus (Fig. 1). She was diagnosed as having non-invasive fungal maxillary sinusitis. She refused surgical treatment and decided to have conservative observation without medication. In October 2004, she complained of right cheek pain. CT images

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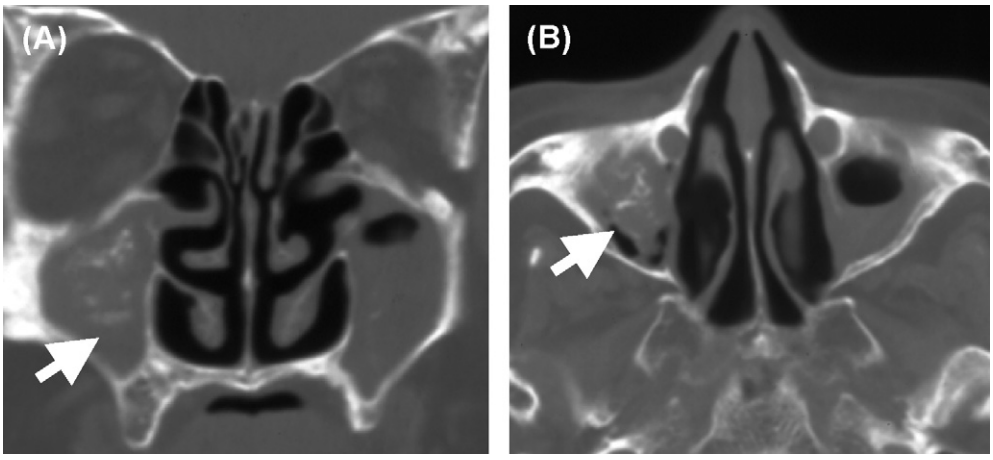

Fig. 1. CT images of the patient at the first visit to our clinic in May 2000. Mucosal thickening and multiple mottled calcifications without bone destruction are seen in the right maxillary sinus (arrows), diagnostic of non-invasive fungal maxillary sinusitis.

showed a soft tissue mass with mottled calcifications, and partial thinning of the bone of the posterior wall of the right maxillary sinus (Fig. 2) was suspected. Endoscopic sinus surgery was performed. A yellowish caseous material was present in the right maxillary sinus and was totally removed. Infiltration of the fungus to the maxillary sinus mucosa and bone destruction were not seen macroscopically. Histological examination revealed Aspergillus infection without tissue invasion. The final diagnosis was non-invasive aspergillosis. After the first operation, the patient did not perform nasal irrigation for local treatment. However, she had no complaints of nasal discharge, nasal blockage, or any other nasal complaints. On nasal endoscopy, the maxillary sinus and ethmoids on the right side were widely opened to the nasal cavity and were found to be normal.

In February 2005, severe cheek pain recurred. CT images revealed an invasive mass lesion in the right orbit and large bone destruction of the posterior wall of the right maxillary sinus (Fig. 3). β-D-glucan is an ingredient of the fungal body, and the serum β-D-glucan level (normal: 0–20.0 pg/ml) is strongly correlated with the clinical course of invasive fungal infections [7] and is assumed to be useful for monitoring the treatment effect. Intravenous micafungin (MCFG) was started at a dose of 300 mg daily. However, the serum β-D-glucan level increased, and the mass lesion in the orbital apex grew after 3 weeks of MCGF treatment. This patient's serum β-D-glucan level increased to 200 pg/ml. In March 2005, a right partial maxillectomy was performed, and the lesions of the orbit and maxillary sinus were removed as much as possible. Histological examination showed aspergillosis with tissue invasion. The serum β-D-glucan level decreased temporarily, but it increased again 3 weeks after surgery. The antifungal agent was switched to intravenous amphotericin B (AMPH) at a dose of 15 mg daily, and the serum β-D-glucan level decreased temporarily to 50 pg/ml. However, the orbital lesion grew (Fig. 4), and the serum β-D-glucan again increased to 230 pg/ml. In June 2005, total removal of the orbital contents with zygomatic ostectomy was performed. In the resected tissue, residual invasion of Aspergillus was found on the proximal side of the optic nerve. Intravenous voriconazole (VRCZ) at a dose of 140 mg/day was administered for 6 weeks, followed by

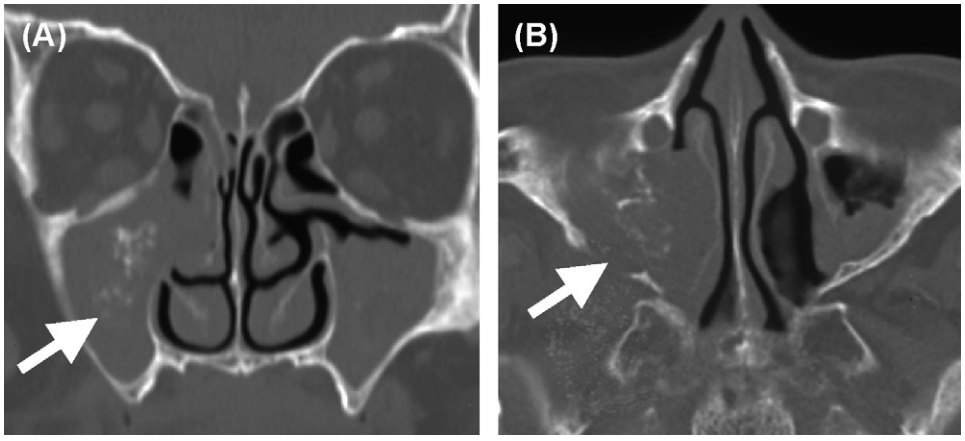

Fig. 2. At the second visit in October 2004 when the patient complained of right cheek pain, a soft tissue mass with mottled calcifications (arrow of the A) and partial bone thinning of the posterior wall (arrow of the B) are seen in the right maxillary sinus.