Case Report

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Invasive Sino-orbital-skull Base Aspergillosis Progressing from Aspergillus Ball of Maxillary Sinus: A Case Report

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Keywords

invasive sino-orbital-skull base aspergillosis, aspergillus ball, orbital apex syndrome

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Introduction

Aspergillus diseases of the sinuses are a diverse group of diseases with subtle presentations such as allergic fungal sinusitis and aspergillus ball (AB) to localized invasive aspergillosis and fulminant form.[1] Usually, localized disease first occurs in the sinuses and spreads to adjacent structures, such as orbital and cranial. The specific diagnosis of invasive sino-orbital-skull base aspergillosis (ISOSA) is often delayed, as it may mimic a number of inflammatory conditions and malignancies.[2] Effective treatment requires early diagnosis because immediate surgical debridement and antifungal therapy are highly recommended.[2,3] However, complete cure is rarely achieved. Here we present a rare and typical case of ISOSA demonstrating orbital apex syndrome arising from the AB of maxillary. This case was approved by the Ningbo Medical Center Lihuili Eastern Hospital institutional review board and ethics committee.

Case Presentation

A 62-year-old man complaining of a 4-week history of a typical right orbital apex syndrome including slight exophthalmos, dropping of the lid, total ophthalmoplegia, loss of vision, forehead pain, and temporal pain was sent to our department. Treatment of antibiotics and glucocorticoid guided by primary ophthalmologists and neurologists was invalid. In addition, the patient suffered from radical excision of gastric carcinoma 4 months prior without chemical therapy. Nasal endoscopic diagnostics brought unremarkable results. Computed tomography (CT) and magnetic resonance imaging showed an invasive inflammatory or malignancy process of the sino-orbital-skull base and pterygopalatinal fossa (**Figure 1A, B**). The patient underwent an endonasal sinus debridement with decompression of the orbit. Repeated postoperative debridements were also carried out. Histology revealed an invasive aspergillosis. *Aspergillus fumigatus* was found in sinus mucosa, orbital fascia, and pus from pterygopalatinal fossa and was the same as AB in maxillary sinuses. We then reviewed his total medical history and found AB in the right maxillary sinus by cranial CT before surgery of gastric carcinoma (**Figure 2**). Beyond that, during the postoperative period, long-term large-dose strong broad-spectrum antibiotics were directed due to persistent high fever. The patient was treated with intravenous voriconazole and finally released into outpatient care. Oral voriconazole was prolonged until galactomannan turned normal. Six months postoperatively, the patient is free of all symptoms except blindness.

Discussion

ISOSA is a well-documented cause of morbidity and mortality in immunocompromised hosts, as well as in immunocompetent ones. On the basis of the literature, *Aspergillus fumigatus* causes it in about 90%.[3] Risk factors include neutrophil defection, glucocorticoid use, HIV infection, diabetes mellitus, prosthetic devices, trauma, excessive environmental exposure, and possibly advanced age.[1]

The clinical manifestations, radiography, and laboratory tests of ISOSA are nonspecific and could mimic infectious, neoplastic, vascular, and neuro-ophthalmic diseases.[2] Endonasal scope may demonstrate almost normal findings. New diagnostic markers, such as serum aspergillus galactomannan and β-D-glucan, are reported to be useful for early diagnosis.[4] However, early diagnosis of ISOSA still remains

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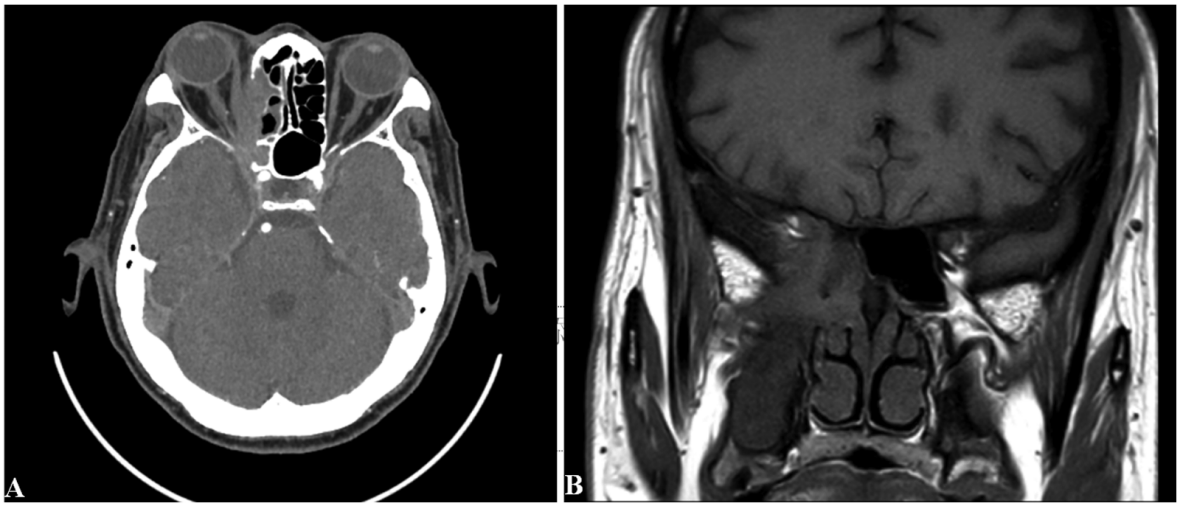

Figure 1. Transversal computed tomography scan and coronal T1-weighted magnetic resonance imaging indicated invasive inflammatory or malignancy process of the sino-orbital-skull base (maxillary sinus, ethmoid sinus, sphenoid sinus, orbit/orbital apex, skull base) and pterygo-palatinal fossa; the boundaries between normal structures were destroyed.

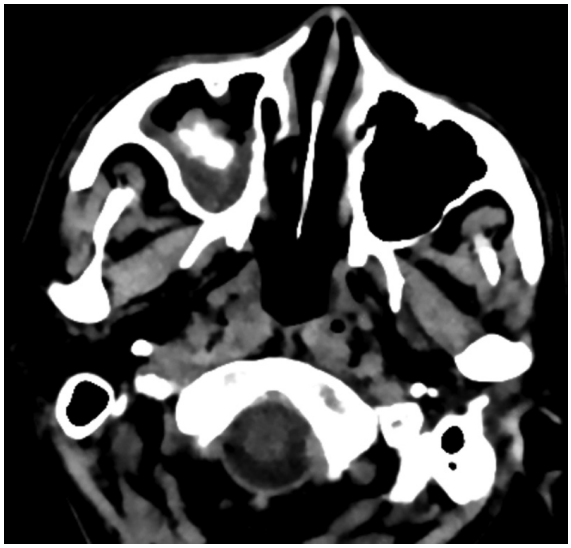

Figure 2. Transversal scan of cranial computed tomography demonstrated an aspergillus ball in the right maxillary sinus without any invasive feature.

a challenge. Isolation of fungus from pathological tissue is the gold standard for the diagnosis. Biopsy—even repeated biopsy—is recommended.[1]

According to the main clinical presentations of ISOSA, such as ocular abnormalities and persistent unilateral headache, ophthalmologists and neurologists have a greater chance to be the first to see patients.[4] Directions such as antibiotics and glucocorticoid may worsen infection, as our case presented. There is still no uniformly accepted, completely effective treatment. Some literature reports that the foremost treatment is surgical debridement without radical resection of functional structures followed by systemic antifungal therapy. Radical procedures must be considered in serious cases depending on the general prognosis of the patient.[2,3] However, biopsy accompanied by systemic antifungal therapy is suggested in other literature.[1] While voriconazole is recommended as the primary therapy of ISOSA according to the Infectious Diseases Society of America, amphotericin B or itraconazole is the first-line therapy in developing countries.[5] Local irrigation with antifungal agents or hyperbaric oxygen has not significantly changed the outcome of the disease.[2]

This is a rare and typical case showing progress from the AB of the maxillary sinus to ISOSA attributed to abnormal use of antibiotics and surgical stress. Therefore, once a patient has noninvasive sinus aspergillosis, any factor that may contribute to the progression of aspergillosis should be considered. In addition, if ISOSA is suspected, antibiotics and glucocorticoid should be suspended,[1] otolaryngologists should be invited onto the treatment team, and multidisciplinary discussion should also be conducted if necessary.

Author Contributions

Qi Huang, analysis and interpretation of data for the work, drafting the work, final approval of the version, agreement to be accountable for all aspects of the work; **Kan Zhao**, acquisition of data for the work, drafting the work, final approval of the version, agreement to be accountable for all aspects of the work; **Zhenhua Wu**, design of the work, revising the work, final approval of the version, agreement to be accountable for all aspects of the work.

Disclosures

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