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Case report

Scedosporium apiospermum invasive sinusitis presenting as extradural abscess

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ABSTRACT

Introduction: Chronic invasive fungal rhinosinusitis (CIFR) is a rare entity generally observed in immunodepressed subjects. The pathogen most frequently identified is *Aspergillus* spp. Imaging generally reveals invasive pseudoneoplastic features. We report a case of *Scedosporium apiospermum* (*S. apiospermum*) CIFR with an atypical clinical and radiological presentation.

Case report: A 72-year-old immunocompetent man presented with chronic headache, neck pain and bilateral limitation of lateral gaze. Imaging revealed an isolated left sphenoidal lesion with marked bone changes and an extradural abscess over the clivus. Large endoscopic sphenoidotomy with type II rhinopharyngectomy was performed and the diagnosis of *S. apiospermum* CIFR was based on histological examination and fungal culture. The patient refused all medical treatment and did not present any signs of recurrence after 1 year of follow-up.

Discussion: S. apiospermum is a fungal species rarely isolated in CIFR. The present case was revealed by an atypical clinical presentation including isolated sphenoidal infection complicated by bilateral abducens nerve paralysis and extradural abscess. Imaging was also unusual, revealing features of fibrous dysplasia or bacterial osteomyelitis rather than the typical pseudoneoplastic appearance. The patient was successfully treated by surgery alone, which may therefore be sufficient treatment in immunocompetent subjects.

1. Introduction

Two forms of fungal rhinosinusitis are classically distinguished: noninvasive and invasive. DeShazo et al. described three forms of invasive fungal rhinosinusitis: acute fulminant, granulomatous and chronic forms [1]. Chronic invasive fungal rhinosinusitis (CIFR) is defined by fungal invasion of the submucosa associated with chronic inflammation and fibrosis, present for more than 12 weeks. This disease is usually observed in immunocompetent patients, but patients with relative immunosuppression, such as diabetes, can also be affected [2]. The pathogens most frequently isolated are *Aspergillus* spp., *Dematiaceae* spp. and mucomycoses [3]. *Scedosporium apiospermum* (*S. apiospermum*) is a rare pathogen, present in polluted water, wastewater and soil [4]. Although it rarely induces significant infections in immunocompetent patients, it can be associated with serious invasive infections in immunodepressed hosts [4].

We report a case of *S. apiospermum* CIFR in an immunocompetent patient with an atypical clinical presentation and discuss the differential diagnoses and the treatment.

2. Case report

A 72-year old immunocompetent man was referred to our department with a 4-month history of worsening headache, associated with bilateral neck pain and bilateral progressive limitation of lateral gaze. He had received several cycles of antibiotics with no improvement. He reported paranasal sinus symptoms of congestion and post-nasal drip. Physical examination revealed bilateral paralysis of lateral gaze with normal pupillary reflexes. Nasal endoscopy showed slight mucosal congestion. CT scan visualized an asymmetrical sphenoidal sinus (Fig. 1) with normal pneumatization of the right sinus with thickened bone in the left sphenoidal sinus, base of the pterygoid and greater wing of the sphenoid

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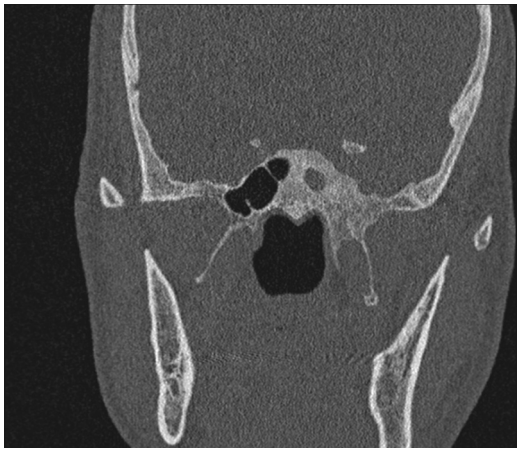

Fig. 1. Computed tomography, coronal section, showing osteosclerosis of the left sphenoid bone.

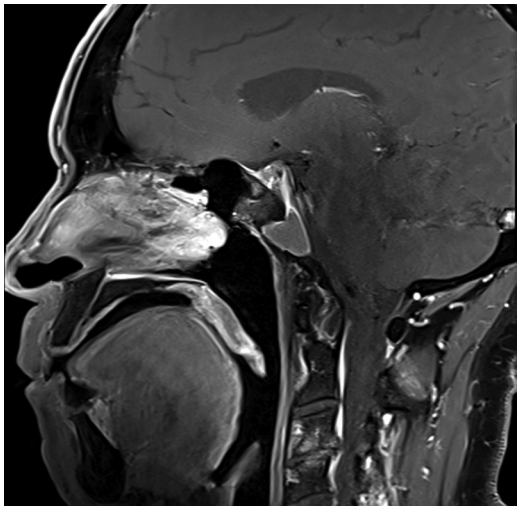

Fig. 2. MRI, gadolinium-enhanced T1-weighted sequence, sagittal section, showing an extradural collection over the clivus with peripheral contrast enhancement.

lateral to the foramen rotundum. Irregular osteolysis was observed in the clivus, occipital condyle, second part of the left internal carotid artery and the floor of the sella turcica. MRI showed that this osteolysis communicated with a T1 and T2 isointense extradural collection with peripheral gadolinium enhancement (Fig. 2). Surgical exploration was performed under general anaesthesia with neuronavigation. Right transethmoidal sphenoidotomy with posterior septectomy was initially performed in order to identify anatomical landmarks in this patient with marked reorganization of the anatomy of the left sphenoidal sinus. The left sphenoidal cavity, the site of a purulent collection, was identified after prolonged drilling. A type II rhinopharyngectomy was then performed to drain the clival abscess in close contact with the dura mater (Fig. 3). The resection included the posterosuperior nasopharyngeal wall as far as the pharyngobasilar fascia, the rostrum sphenoidale and the intersphenoidal septum, as well as the anterior wall and floor of the sphenoidal sinus. The dura mater was lined by a right nasoseptal flap. Histological examination of biopsies revealed signs of chronic nonspecific mucosal inflammation with fibrosis and mild bone changes. Fungal mycelia were demonstrated in the submucosa. Fungal culture was positive for *S. apiospermum*, sensitive to a large number of antifungal agents including amphotericin B and voriconazole. Bacteriological culture was negative in this patient who had not received any antibiotics preoperatively. The

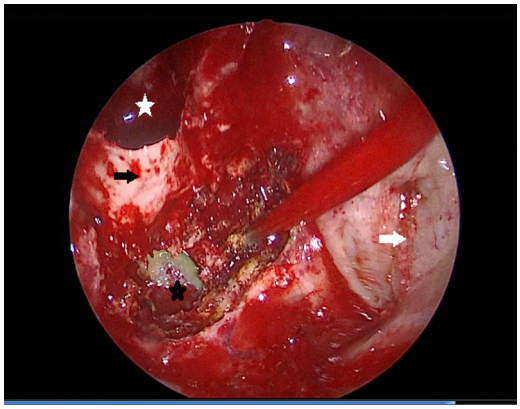

Fig. 3. Drainage of the abscess over the clivus with exposure of the dura mater (black asterisk). Note the appearance of the left sphenoidal sinus after drilling (white asterisk), and the remaining rostrum sphenoidale (black arrow).

final diagnosis was that of CIFR complicated by extradural abscess. Treatment with oral voriconazole was proposed, but was refused by the patient. At one-year follow-up, the patient was asymptomatic with no signs of recurrence on either clinical examination or MRI.

3. Discussion

In the case reported here, the diagnosis of CIFR was based on the prolonged duration of the symptoms, the histological findings and fungal culture results. *S. apiospermum* sinusitis is a rare disease. In their series of 400 cases of fungal sinusitis, Montone et al. isolated *S. apiospermum* in 5% of cases of aspergilloma and in only one case of CIFR [2].

Apart from the rarity of CIFR and the pathogen isolated, this case was remarkable in terms of its unusual clinical presentation: isolated left sphenoidal sinusitis with marked bone changes, extradural abscess over the clivus and bilateral paralysis of the 6th cranial nerve in an immunocompetent patient. The possible differential diagnoses included chondrosarcoma, chordoma, fibrous dysplasia, carcinoma and infection [5]. Fibrous dysplasia affects younger patients and rarely involves the sphenoidal sinus alone. However, CT scan in the present case revealed a “frosted glass” appearance of the bone associated with an intraosseous aneurysmal cyst [6]. Erosion of the clivus could be suggestive of chordoma. However, chordoma does not usually present these features of bone thickening with preserved bone margins, and gadolinium enhancement tends to be heterogeneous with a “beehive” appearance [7]. Extradural abscess is a classical complication of frontal sinusitis, but is exceptional in ethmoidal and sphenoidal sinusitis [5,8,9], and occurs in a context of bacterial sinusitis and no associated cranial nerve lesions have been reported. The radiological appearance of CIFR is often nonspecific and can consist of simple mucosal thickening, although the most common presentation is that of tissue opacification in one or more paranasal sinuses with bone erosion and invasion of adjacent spaces [10]. This appearance can also be suggestive of malignant tumour and it is often difficult to differentiate these entities exclusively on the basis of imaging findings. The case reported here was atypical in that imaging did not show an invasive pseudoneoplastic appearance, possibly related to the fact that the pathogen isolated, *S. apiospermum*, may present a different invasive behaviour to that of the pathogens more frequently isolated: *Aspergillus*, *Dematiaceae* spp. or mucomycoses [3].

No consensus has been reached concerning optimal management of CIFR, but most authors propose a combination of surgical debridement, systemic antifungal therapy adapted to antifungal susceptibility test results, and, when necessary, optimal correction

of immunosuppression [3]. In the case reported here, the patient refused voriconazole therapy, and, with a follow-up of one year, surgery alone appeared to have controlled the disease in this immunocompetent patient.

4. Conclusion

In conclusion, we report a rare case of *S. apiospermum* CIFR in an immunocompetent patient with an atypical clinical and radiological presentation. Surgery allowed biopsies that are essential for the diagnosis, and could constitute sufficient treatment in immunocompetent subjects.

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Disclosure of interest

The authors declare that they have no competing interest.

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