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ORIGINAL ARTICLE

Clinical features and outcome of sphenoid sinus aspergillosis: A retrospective series of 15 cases

A. Thery[a], F. Espitalier[a], E. Cassagnau[b], N. Durand[a], O. Malard[a,*]

[a] Service d'ORL et de chirurgie cervicofaciale, Hôtel-Dieu, CHU de Nantes, 1, place A.-Ricordeau, BP 1005, 44093 Nantes cedex 01, France
[b] Service d'anatomopathologie, Hôtel-Dieu, CHU de Nantes, 1, place A.-Ricordeau, BP 1005, 44093 Nantes cedex 01, France

KEYWORDS
Sphenoid sinusitis;
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Headache;
Nasal discharge;
Immunodepression

Summary
Introduction: Sphenoid sinusitis is uncommon, and aspergillus infections represent a minority of these cases. This study was designed to describe the characteristics of this disease and present a review of the literature.
Patients and method: Retrospective study from 2004 to 2010 based on 15 patients managed at Nantes University Hospital. Risk factors and history, symptoms, intranasal examination, imaging, histological and mycological results as well as analysis of the response to treatment and outcome were analysed.
Results and discussions: Patients were aged between 14 to 78 years, almost 75% of patients were older than 50 and 73% of patients were women. The most common symptoms were headache [80%], nasal blockage or discharge [33%], and recurrent mild epistaxis [20%]. Predisposing factors were immunodepression in three patients, with no cases of diabetes. Two patients had a history of intranasal surgery and one had a history of facial trauma. Nasal endoscopy was normal in 40% of cases. CT was suggestive of sphenoid sinus aspergillosis in more than one half of cases [8/15] and demonstrated osteolysis in four patients. An incorrect preoperative diagnosis of mucocele was proposed in three patients. Histological examination demonstrated spore-forming structures in every case, but culture was positive in only four cases. Only two patients required antifungal therapy, including one patient with invasive aspergillosis.
Conclusion: Chronic noninvasive sphenoid sinus aspergillosis appears to be a benign disease, essentially affecting women and patients over the age of 50 years. Symptoms are fairly non-specific. Imaging and histological examination are essential for management. The invasive form is a serious disease requiring rapid, multidisciplinary management.

Introduction

The estimated incidence of sphenoid sinusitis is 2.7% of all nasal sinus infections [1]. Isolated sphenoid sinusitis can be bacterial or fungal. Fungal sinusitis represents 15 to 20% of

* Corresponding author.
E-mail addresses: omalard@chu-nantes.fr, olivier.malard@chu-nantes.fr (O. Malard).

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all cases and is classified as noninvasive, invasive indolent and fulminant [2].

The diagnosis of sphenoid sinus aspergillosis is sometimes difficult [3]. Clinical signs are often nonspecific and nasal endoscopy can be strictly normal [4]. Early diagnosis is therefore difficult and diagnosis is often delayed with headache that may sometimes persists for several years before diagnosis of the disease [5].

The aetiopathogenesis is poorly elucidated, but decreased aeration of the sinus appears to play a major role in the development of the main pathogen, *Aspergillus fumigatus*. Surgical resection of the aspergilloma by endoscopic sphenoidotomy appears to be the only treatment required in this case. However, this is not the case for invasive aspergillosis, in which systemic antifungal therapy must be administered [6].

Although several reviews of the literature have been devoted to maxillary aspergillosis or sphenoid sinusitis, only a few cases of sphenoid sinus aspergillosis have been documented. A recent study [1] published in 2009 reported 50 cases published in the literature since 1950 and provided limited epidemiological data.

The present study concerns 15 cases, which, to our knowledge, is one of the largest series yet to be published in the literature. This study was designed to describe the epidemiological, clinical and radiological characteristics of these patients. Data concerning treatment and outcome of sphenoid sinus aspergillosis are discussed and compared to the data of the literature.

Patients and methods

This 6-year retrospective study, from January 2004 to July 2010, was conducted in the ENT department of Nantes University Hospital. All cases of sphenoid sinusitis were retrieved from the *Classification Commune des Actes Médicaux* (CCAM) [Common Classification of Medical Procedures] database. To avoid missing any cases, data obtained from the International Classification of Diseases (ICD 10) and CCAM procedure codes were cross-matched. Only fungal forms of sinusitis were included

Inclusion criteria were:

- patients who had undergone an intranasal surgical procedure, consisting of sphenoidotomy or posterior ethmoidectomy with sphenoidotomy,
- patients with histological results in favour of aspergillosis.

The following variables were analysed: demographic, epidemiological, clinical, CT data, treatment, as well as outcome and prognosis. Epidemiological data such as age of onset and gender were analysed. The time to management following onset of symptoms was evaluated when available from retrospective review of the medical charts. The patient's history was investigated, especially a history of trauma or intranasal surgery and the presence of immunodepression or diabetes.

Clinical symptoms were evaluated (headache, nasal discharge, epistaxis, diplopia, anaesthesia of the maxillary division of the trigeminal nerve). Clinical signs and nasal endoscopy data were also studied.

Computed tomography (CT) examinations were analysed, looking for calcifications, osteolysis or osteosclerosis and signs of extension to the cavernous sinus. Diagnoses proposed at the time of the initial assessment were taken into account. The histological and mycological results were recorded.

A telephone survey of all surviving patients was performed to collect data concerning the post-treatment course, including all postoperative symptoms and residual symptoms after the operation.

Two clinical cases presenting severe forms are reported.

Case 1: Sphenoid sinus aspergillosis in an immunodepressed patient

This 53-year-old woman was treated with azathioprine (Imurel®) for ulcerative colitis and primary biliary cirrhosis. She described initially isolated headache, then associated with facial sensory loss. Magnetic resonance imaging (MRI) was performed urgently following the appearance of diplopia and revealed an opacity of the right sphenoid sinus associated with a solid mass in the nasopharynx and right cavernous sinus. Clinical examination revealed palsy of the V1, V2 and VI cranial nerves. Imaging was completed by CT of the facial bones to evaluate bone structures. This examination demonstrated intrasphenoidal calcifications in favour of the diagnosis of aspergillosis, and signs of cavernous sinus thrombosis.

Sphenoidotomy was performed. Histological examination demonstrated spore-forming structures confirming the presence of fungal infection. The course was very slowly favourable despite systemic antifungal and anticoagulant therapy with persistence of the cavernous sinus thrombosis postoperatively. The staging and complementary investigations performed during assessment of the thrombosis demonstrated abdominal lymphadenopathy related to concomitant non-Hodgkin's lymphoma, for which the patient is currently receiving treatment.

Case 2: Invasive sphenoid sinus aspergillosis

This non-immunodepressed 74-year-old man with no particular history reported a 2-month history of occipital headache associated with persistent nasal blockage despite several topical treatments. Nasal endoscopy revealed a suspicious discharge from the ostium of the left sphenoid sinus. CT examination demonstrated complete filling of the left sphenoid sinus for which the radiologist proposed a diagnosis of mucocele or tumour associated with hyperostosis.

Sphenoidotomy and biopsies were performed. The histological results were in favour of sphenoid sinus aspergillosis with acute necrotic and ulcerated changes of the mucosa. Mycological examination was negative on direct examination and culture revealed *Aspergillus fumigatus* after 48 hours of culture. The patient received postoperative antibiotic therapy. No systemic antifungal therapy was proposed in the absence of osteolysis.

One month postoperatively, the patient described persistent discomfort. Clinical examination showed a polypoid appearance of the edges of the sphenoidotomy. Treatment