



ELSEVIER



www.elsevierhealth.com/journals/jinf

CASE REPORT

A pseudo tumoral sinusitis caused by *Bipolaris* sp.

P. Toul^{a,*}, L. Castillo^b, V. Hofman^c, J.-P. Bouchara^d, S. Chanalet^e,
M. Gari-Toussaint^a

^a Laboratoire de Parasitologie—Mycologie, Hôpital de l'Archet, Nice, France

^b Service d'oto-rhino-laryngologie, Hôpital Pasteur, Nice, France

^c Service d'anatomopathologie, Hôpital Pasteur, Nice, France

^d Laboratoire de Parasitologie—Mycologie, CHU d'Angers, France

^e Service d'Imagerie diagnostique, Hôpital Pasteur, Nice, France

Accepted 10 February 2006

KEYWORDS

Sinusitis;
Bipolaris;
Phaeohyphomycosis

Summary This is the first published French case of a rhinosinusitis caused by *Bipolaris* sp. in an immunocompetent patient and suggests that this organism must be considered as an opportunistic human pathogen in warm and damp climate like the south-east of France. *Bipolaris* sp. is the most isolated fungi in allergic fungal sinusitis.

© 2006 The British Infection Society. Published by Elsevier Ltd. All rights reserved.

Case report

A 50-year-old immunocompetent French man had presented a left nasal obstruction for more than 5 years. Furthermore, he was atopic, healthy, and took regular treatment composed of corticosteroids and antihistaminics. He had no history of travel abroad. The nasal endoscopy examination revealed polyps in the left sinuses and an anatomical mass obstruction.

The computed tomography scan (e.g. Fig. 1) showed important hyper-attenuating masses on the left maxillar frontal ethmoidal sphenoidal

sinuses with an extension in the homolateral orbit. On the sagittal view an erosion of the posterior wall of the sphenoid sinus was observed.

On magnetic resonance imaging the coronal view showed mucus retention in the sphenoid sinus that is directing towards a fungal lesion.

Endoscopic sinus surgery of the left side was performed. It revealed an anatomic sinus obstruction, polyp formation, gross mucus and the totality of the masses were extracted for both histological and mycological examinations.

Histology

Histological examination of the totality of the tissue from an endoscopic examination was performed.

* Corresponding author. Service de Parasitologie Mycologie, Hôpital de l'Archet II route de Saint Antoine de Ginestière 06200 Nice, France. Tel.: +33 493044095; fax: +33 492036258.
E-mail address: patoutchi@caramail.com (P. Toul).



Figure 1 CT Scan coronal view: opacification of the left sinuses by *Bipolaris*.

Hematoxylin and eosin (H&E) staining revealed varying degrees of inflammatory infiltrate with lymphocytes and plasma cells and a little contingent of eosinophils and neutrophils. In addition, it showed compacted mucin and no granulomatous or criteria of tumor. On the Gomori-Grocott stain rare fungal hyphae were found (e.g. Fig. 2).

Mycology

Fragments of the masses were cultured on Sabouraud-chloramphenicol agar medium at 28 °C. A pure culture of *Bipolaris* was grown in two days in the form of black velvety colonies in picture 1. Microscopic examination (e.g. Fig. 3) showed septate pigmented hyphae, geniculated conidiophores with thick-walled, brown cylindrical or fusoid conidia, with predominantly four cells and three

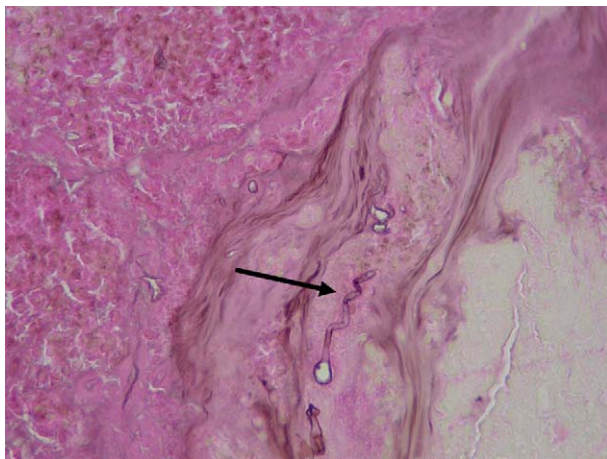


Figure 2 Microscopy × 630: Gomori-Grocott: a few twisted mycotic elements with bizarre forms are scattered in necrotic tissue.

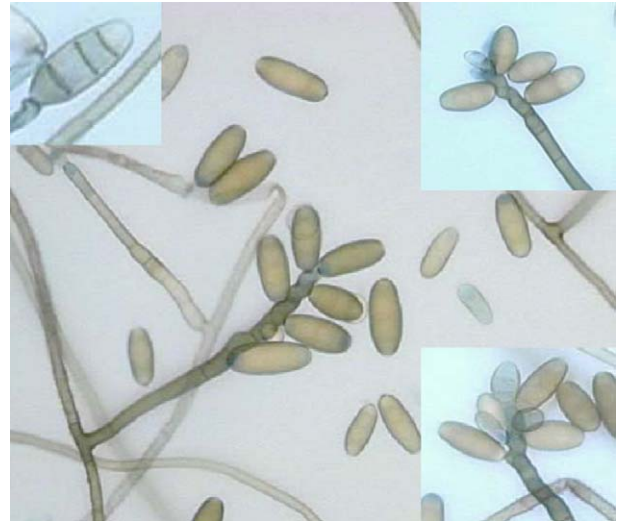


Figure 3 Microscopy × 400: lacto phenol cotton blue stain: sympodial geniculate conidiophore producing four celled conidia having truncated hilum.

septates. These conidia have sympodial presentation and dark truncated hilum as in *Bipolaris spicifera*.^{6,8}

To confirm the species,² PCR of ITS-5,8S region of the ribosomal gene was extracted from the culture because of the similarity between the different moulds.

There was the same percentage of homology for *Cochliobolus spicifera* and *Cochliobolus australiensis*, which are both teleomorphs of these two species.

The dosage of the total of immunoglobulin E was not searched in the patient's blood.

The increase of IgE is a criterion of clinical severity of AFS.¹¹

He did not receive any post-operative medication and was cured by surgical resection alone.

At present, he is still well and so far has had no recurrence of the disease.

Bipolaris sp. is a widespread worldwide dematiaceous fungi which generally occurs in immunocompetent individuals and causes phaeohyphomycosis^{1,2,4,5,7,8} and particularly nasal sinusitis.² *Bipolaris* is found in soil and plant materials and is known to be plant pathogens and was considered for a long time as a contaminant.^{1,4} However, the increase of its presence in culture suggests that it must be under diagnosed until now.⁴ Since the end of the 1980s it has been reported as a cause of fungal rhinosinusitis and to be the commonest agent of allergic fungal sinusitis (AFS),^{1,3,10} particularly caused by *B. spicifera*⁹ in the south-western United States,^{1,2,10} where it is habitually diagnosed. In France, no case was

described until now. We reported the first French case to our knowledge which occurs in a warm and damp climate of the south-east of France.

Fungal sinusitis is seen in five distinct clinico-pathological forms. Three of them are invasive: acute fulminant, chronic and granulomatous fungal sinusitis and the others are non-invasive: sinus mycetoma and AFS.⁹

This case does not correspond to a specific entity of fungal sinusitis. The differentiation between the different forms of rhinosinusitis is important for prognosis and treatment.¹⁰ Clinically, it presented as a sinus tumor, few aggressive with extension into the contralateral sinus and the left orbit whereas the investigations found no criteria of tumor but a pure culture of *Bipolaris* sp. It was considered as an invasive entity close to granuloma because of the clinical presentation included the risk factor and the gross examination but histology described no granuloma or an atypical chronic invasive form due to the absence of the necrotic spread of the infection.⁹

Although the non-invasive forms were also eliminated by histology in particular the allergic form (AFS), the criteria described by Mark S. Schubert were not all present.^{9,10}

Finally, the treatment approach consisted of a surgical resection without post-operative antifungal drugs like in the non-invasive mycetoma form which expected a good prognosis⁹ without taking account of the sphenoid because the bone erosion may be due to pressure atrophy than due to fungal invasion which must be histologically demonstrated. The diagnosis needs collaboration between surgeons and medical specialists, such as radiologist, mycologist and anatomic-pathologist, who will permit more diagnosis of these different entities of fungal sinusitis.

This case was diagnosed as a chronic invasive form in an immunocompetent patient and cured

by surgery and demonstrated that it is extremely difficult to conclude on a definite entity.

Normally, chronic invasive sinusitis caused by dematiaceous fungi requires both surgical and medical treatment by antifungal drugs and steroid therapy.⁹

It is also important to know that it is possible to have several entities in one case as was described by Brandt and Warnock.¹

References

1. Brandt ME, Warnock DW. Epidemiology, clinical manifestations, and therapy of infections caused by dematiaceous fungi. *J Chemother* 2003;15(Suppl. 2):36–47.
2. Buzina W, Braun H, Schimpl K, Stammberger H. *Bipolaris spicifera* causes fungus balls of the sinuses and triggers polypoid chronic rhino sinusitis in an immunocompetent patient. *J Clin Microbiol* 2003;41(10):4885–7.
3. Ferguson BJ. Definition of fungal rhinosinusitis. *Otolaryngol Clin North Am* 2000;33:227–36.
4. Karim M, Sheikh H, Alam M, Sheikh Y. Disseminated *Bipolaris* infection in an asthmatic patient: case report. *Clin Infect Dis* 1993;17(2):248–53.
5. McGinnis MR, Campbell G, Gourley WK, Lucia HL. Phaeohyphomycosis caused by *Bipolaris spicifera*: an informative case. *Eur J Epidemiol* 1992;8(3):383–6.
6. McGinnis MR, Rinaldi MG, Winn RE. Emerging agents of Phaeohyphomycosis: pathogenic species of *Bipolaris* and *Exserohilum*. *J Clin Microbiol* 1986;24:250–9.
7. Ogden PE, Hurley DL, Cain PT. Fatal fungal endarteritis caused by *Bipolaris spicifera* following replace of the aortic valve. *Clin Infect Dis* 1992;14(2):596–8.
8. Rao A, Forgan-Smith R, Miller S, Haswell H. Phaeohyphomycosis of the nasal sinuses caused by *Bipolaris* species. *Pathology* 1989;21(4):280–1.
9. Schubert MS. Fungal rhinosinusitis: diagnosis and therapy. *Curr Allergy Asthma Rep* 2001;1:268–76.
10. Schubert MS, Goetz DW. Evaluation and treatment of allergic fungal sinusitis. I. Demographics and diagnosis. *J Allergy Clin Immunol* 1998;102(3):387–94.
11. Schubert MS, Goetz DW. Evaluation and treatment of allergic fungal sinusitis. II. Treatment and follow up. *J Allergy Clin Immunol* 1998;102(3):395–402.