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Cerebral blastomycosis in a Tunisian woman

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ABSTRACT

Background: Blastomycosis is a non-opportunistic fungal infection caused by Blastomyces dermatitides. Infection of the central nervous system is an uncommon localization of this disease occurring only in 5–10% of cases.

Case report: We report a new case of a 57-year-old woman, who was admitted in the neurosurgical department for severe headache, vomiting, and a visual disturbance. Physical and biological investigations were normal. Magnetic resonance imaging led to find an intracranial and extracranial expansive processes, measuring 42 mm in length. The microscopic examination revealed a chronic granulomatous inflammation, associating epithelioid and multinucleated giant cells containing thick-walled budding yeast-like forms. Grocott-Gomori methenamine–silver staining led to confirm its fungal nature, and it was precisely Blastomyces dermatitidis. The postoperative course was complicated by cerebral edema associated with an important cerebral hemorrhage. The patient died 11 days after the surgery.

Conclusion: Cerebral involvement remains a serious but uncommon manifestation of blastomycosis. It should be suspected in front of a granulomatous inflammation with the presence of spore on the pathological examination. The delay in diagnosis has been reported to play a significant role in the high mortality rate.

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KEYWORDS

Blastomycosis; fungus infection; central nervous system

Introduction

Fungal infections of the brain are rare, usually occurring as opportunistic disease in immune suppressed patients. Blastomycosis is different by its eventual occurrence in a normal immune state patient. Originally described by Gilchristl as a cutaneous infection, it is now accepted as a systemic disease with many classic and unusual locations [1–3].

Infection of the central nervous system is a secondary localization of the systemic disease. It is uncommon occurring in only 5–10% of patients and it is potentially a fatal complication of this infection [2,4]. This is possibly due to hematogenous spread of the infection to the brain. Chronic meningitis is the more frequent form of presentation, but cerebral and extra dural abscesses, intraparenchymal granulomas, and cerebritis have also been reported [5–7].

Although effective antifungal therapy is available, the difficulty of the establishment of the disease in early stages hampers management of the infection.

Case Report

We report the case of a 57-year-old woman, who was admitted to the hospital for severe headache, vomiting, and a visual disturbance.

Tomodensitometry and magnetic resonance imaging (MRI) revealed an intracranial and extracranial expansive process, measuring 42 mm in length, located in the left frontal lobe (Fig. 1(a)). This process was located in the anterior floor on the base of the skull, reposing on the sphenoid bone and the orbit roof. It crosses the perpendicular plate (Fig. 1(b)) and comes in contact of the orbital nerve.

MRI showed that the lesion was isointense in T1; hypointense in T2, and was enhanced after injection

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of contrast. It was surrounded by edema, and exercised a mass effect on the frontal horn of the left lateral ventricle.

The intra-operative appearance of the process suggested an aggressive left spheno-orbitar meningioma.

The patient underwent surgery; the mass was resected and totally removed.

In gross, we received a well-demarcated mass, gray-whitish, measuring 4 cm in length, homogenous in the cut surface.

The microscopic examination of the specimen showed a chronic granulomatous inflammation (Fig. 1(c)), including epithelioid and multinucleated giant cells containing foreign body presented as thick-walled budding yeast-like forms (Fig. 1(d)). Grocott-Gomori methenamine–silver and Periodic acid-Schiff staining confirmed that it was fungus: Blastomyces dermatitidis (Fig. 2).

The postoperative course was complicated by cerebral edema associated with an important cerebral hemorrhage. The patient died 11 days after the surgery.

Discussion

Blastomycosis is caused by the thermally dimorphic fungus Blastomyces dermatitides. It is generally thought to be soil-borne, but the isolation of this organism from nature is difficult.

North-American blastomycosis is endemic in the central and the south eastern of the United States. Recently, other cases have been also reported in Canada, Europe, Mexico, Central America, and Africa [1–3].

It occurs more commonly in males, with reported 4:1 to 15:1 male-to-female ratios [7].

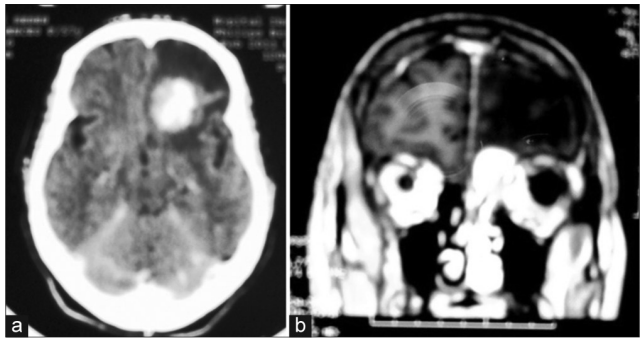

Figure 1. (a) Tomodensitometry shows intracranial and extracranial expansive processes, localized in the left frontal lobe; (b) MRI shows a lesion localized in the anterior floor on the base of the skull, reposing on the sphenoid bone and the orbit roof, and crosses the perpendicular plate.

Most patients were aged between 20 and 70 years, and the diagnosis was most often established in the fourth decade. Rare cases were reported in patients younger than 2 year old [7].

There are no well-documented cases of man-to-man, man-to-animal, or animal-to-animal transmissions [7].

Airborne spores of blastomyces are first inhaled by lungs. At body temperature, the spores are converted into budding yeasts. The initial response is a pneumonic inflammation consisting of polymorphonuclear leukocytic suppuration. The findings on chest roentgenograms are not distinctive, with changes varying from a consolidated lobar pneumonia to multiple diffuse infiltrations. Hilar adenopathy is common but cavitation is rare [5].

As the inflammatory process continues, a foreign body giant cell reaction is developed, associated to frank epitheloid granulomas.

The pneumonic infection may heal spontaneously, or infection may spread locally to mediastinal lymph nodes or disseminate hematogenously, involving skin, bones, kidney, liver, brain, or prostate. Disseminated lesions may occur at the time of primary infection or may be synchronous to the pneumonic illness [5].

Central nervous system involvement by blastomyces is estimated to occur in 5–10% of case [6]. It presents, commonly, as a chronic meningitis, subdural focal leptomeningeal abscess, or intraparenchymal abscess. The latter two pathological entities are

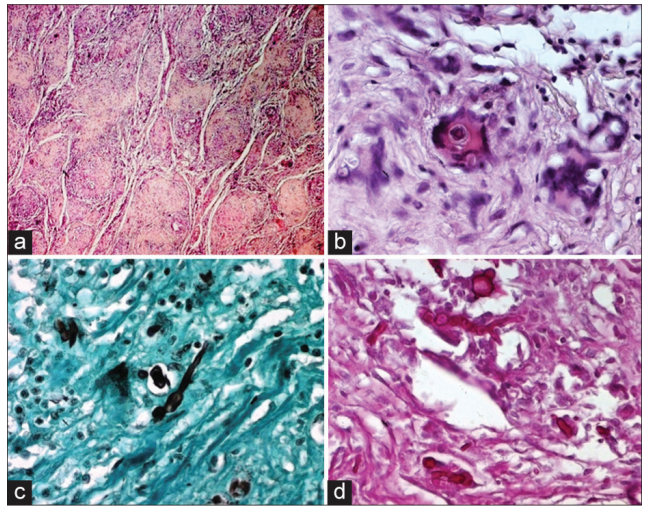

Figure 2. (a) Chronic granulomatous inflammation at microscopic examination (×40); (b) Multinucleated giant cells containing foreign body at microscopic examination (×400); (c) Grocott-Gomori methenamine–silver staining confirmed that it was Blastomyces dermatitidis (×400); (d) Periodic acid-Schiff staining confirmed that it was Blastomyces dermatitidis (×400).

encompassed under the rubric of cerebral blastomycoma [1]. In patients with intracranial locations, may be single and multiple lesions can be seen, and the cerebellum is frequently involved [2].

Microscopically, the fungus can be suspected, on standard Hematoxylin and eosin (HE) stain, by its dimorphism, and visualization of the small spores born on hyphae in the mold form. Its cell's membrane is doubly refractile. In tissue, it grows as large, spherical, thick-walled yeast, measuring 7 to 20 µm in diameter (7), with polar budding evocating a draft filament [3].

The treatment of choice of blastomycosis is Amphotericin B [2]. It is relatively toxic, in fact, a high incidence of nephrotoxicity and bone marrow suppression were reported. The 2-hydroxystilbamidine had an acceptable cure rates in patients in whom the blastomycosis has been slowly progressive and of limited extent, confined to the lung and the skin. Ketoconazole was also recently used in few selected patients with some cures [1,5–7].

Conclusion

Cerebral involvement remains a serious but uncommon manifestation of blastomycosis. It should be suspected in front of a granulomatous inflammation with the presence of spore on the pathological examination. The delay in diagnosis has been reported to play a significant role in the high mortality rate.

Conflict of interest

None

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