

## Aspergillus otomastoiditis: a case report and review of the literature

Verónica Martínez-García<sup>1\*</sup> and Leonardo D. De la Torre-Carmona<sup>2</sup>

<sup>1</sup>Departamento de Medicina Interna; <sup>2</sup>Departamento de Infectología. Instituto de Seguridad y Servicios Sociales de los Trabajadores del Estado, Hospital Regional "Gral. Ignacio Zaragoza", Mexico City, Mexico

### Abstract

Otomycosis is an infection in the external ear canal, which involves the inner ear. The genus *Aspergillus* is the most frequent causal agent, can present with otorrhea, otalgia, pruritus, and tinnitus and is more frequent in patients with predisposing factors such as immunocompromise or living in humid and/or warm climates. We present the case of a 61-year-old woman, a merchant, who was considered immune-competent on entry and had a history of storing brick and cement fragments in her home, in those who have been found to have fungal-induced otic and mastoid infection with adequate clinical response to treatment.

**Keywords:** Otomycosis. Aspergillosis. *Aspergillus*. Otitis. Voriconazole. Mycosis.

### Introduction

Otomycosis is an infection in the external ear canal and the inner ear. It can occur *de novo* in a patient with predisposing factors or appear as a complication of the use of broad-spectrum antibiotics. This is caused by different types of fungi, mainly *Candida* and *Aspergillus*, highlighting the species *Aspergillus niger* (the most common in this condition), *Aspergillus alliaceus*, *Aspergillus candidus*, *Aspergillus flavus*, *Aspergillus fumigatus*, *Aspergillus terreus*, and *Aspergillus versicolor*<sup>1</sup>. A rare and potentially fatal complication is otomastoiditis<sup>2</sup>.

This disease can present clinically with otorrhea, otalgia, pruritus, and tinnitus and is more common in patients with predisposing factors such as immunocompromise, living in tropical or subtropical climates, insulin resistance and a history of storing brick and cement fragments in the home; important data to support the suspicion of the final diagnosis<sup>3</sup>.

### Case report

A 61-year-old woman, resident of Mexico City, presented with left otalgia of 7 days of evolution, of insidious onset, associated with fetid seropurulent otorrhea and progressive hearing loss. Her personal history included prediabetes, systemic arterial hypertension, previous hospitalization for hypokalemic paralysis, storage of brick, and cement fragments in her home. On physical examination, bone fragments were observed in the left external auditory canal, foul-smelling purulent discharge, and loss of integrity of the tympanic membrane. Plain and contrast-enhanced computed tomography of the skull showed left mastoid destruction, with no evidence of neuroinfection (Fig. 1).

A left mastoidectomy with tissue debridement was performed. The histopathological study reported fungal structures compatible with *Aspergillus* spp. (Fig. 2), later identified as *Aspergillus terreus* by

**\*Correspondence:**

Verónica Martínez-García  
E-mail: mc01.veronicamartinez@gmail.com  
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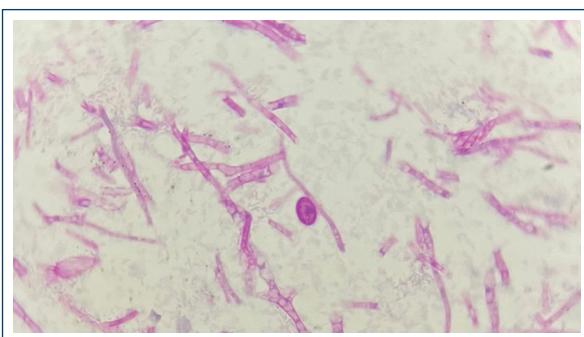
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**Figure 1.** Cranial CT in contrast-enhanced phase, axial section: loss of pneumatization of the left mastoid cells associated with bone destruction is observed.



**Figure 2.** Periodic acid-Schiff stain in mastoid bone biopsy sample: hyaline, septate, thin-walled hyphae are observed, with dichotomous branches at 45 degrees.

genetic sequencing. Treatment with oral voriconazole was started for 6 weeks, with adequate clinical and imaging response, without subsequent neurological alterations, and with complete resolution of the condition.

## Discussion

A case was presented with the diagnosis of *Aspergillus* otomastoiditis confirmed by histopathological report and genetic sequencing. This was treated satisfactorily with a medical and surgical approach, with favorable evolution. The diagnosis requires a high level of suspicion according to risk factors and epidemiological context since the prognosis worsens exponentially when these are delayed and the initial clinical course

can be insidious. In the case presented here, timely detection prevented additional complications.

The diagnosis can be made by identifying hyphae in a debridement sample of the lesions. During the approach, soft-tissue involvement, bone destruction, or intracranial extension must be evaluated. Skull tomography with simple and contrasted phases is the best study to evaluate bone tissue and data of possible infection in the central nervous system; among the findings, bone erosion, decreased density of the skull base, abscesses, and mastoid involvement should be looked for. To assess whether there is intracranial extension, magnetic resonance imaging is more appropriate. The definitive laboratory diagnosis is by mycological culture, direct microscopy, or histopathology with samples taken directly from the external auditory canal and secretions. In the case presented, the diagnosis could be established by histopathological study of a sample taken directly from the mastoid bone<sup>3,4</sup>.

One of the most feared, but rarest complications of otomycosis is otomastoiditis, described in a small series of cases<sup>5</sup>, where the prolonged use of antibiotics and topical steroids is common. Fungal disease of the temporal bone has been classified into three types: type 1 or non-invasive, Type 2 or with bone invasion (especially in immunocompetent patients), and Type 3 or fulminant angio-invasive (more common in immunosuppressed patients)<sup>6</sup>; The case presented here corresponds to Type 2.

There is no consensus on the most effective agent for the treatment of otomycosis; there are a few publications on its treatment with topical antifungals. It is important to note that some of these studies do not report clinical or mycological details cure rates, or routes of administration. However, everyone agrees on the importance of local mechanical debridement of visible fungal elements and the use of topical or systemic antifungals. Systemic antifungals are reserved for fungal cases of mastoiditis or cerebral extension<sup>7</sup>.

For extrapulmonary forms of aspergillosis, the first treatment option is triazoles, among which the preferred is voriconazole, or alternatively, posaconazole or isavuconazole. If it is not available, or if the patient is intolerant or refractory to such treatments, Amphotericin B is considered the next therapeutic option<sup>8</sup>. New antifungals are in development, such as fomanogepix, ibrexafungerp, olorofim, and rezafungin, which could be good therapeutic options in the near future<sup>9</sup>.

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## Conflicts of interest

The authors declare no conflicts of interest.

## Ethical disclosures

**Protection of humans and animals.** The authors declare that no experiments on humans or animals have been performed for this research.

**Confidentiality of data.** The authors declare that no patient data appear in this article.

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