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CASE REPORT

Aspergillus mastoiditis, presenting with unexplained progressive otalgia, in an immunocompetent (older) patient

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Abstract *Aspergillus* mastoidits and skull-base osteomyelitis are extremely rare, even in immunocompromised patients. We report a case of an 81-year-old immunocompetent man, who underwent a mastoidectomy because of unexplained, progressive otalgia in spite of a noninflamed and air-containing middle-ear space. Histopathology yielded *Aspergillus fumigatus.* When confronted with otitis with an unexpected clinical course a high index of suspicion is required to facilitate early diagnosis and appropriate therapy of a potential lethal *Aspergillus* infection, even in immunocompetent patients. This seems to be more so in older patients with an open middle-ear cavity and/or when there is facial nerve involvement.

Keywords *Aspergillus* · Matoiditis · Immunocompetent · Otitis

Introduction

Invasive skull-base mastoiditis, also known as malignant or necrotizing otitis externa, typically affects immunocompromised patients (diabetes, leukemia, AIDS, prior treatment with cytotoxic medication and or corticosteroids) and is usually caused by *Pseudomonas aeruginosa*.

Although being an uncommon condition, it is often well recognized and at one moment differentiated from an 'ordinary' otitis due to alarming (progressive) symptoms such as extreme/deep otalgia, vertigo-hearing loss (labyrinthitis), facial nerve dysfunction and sometimes dysfunction of other nerves of the cerebellopontine angle (Gradenigo syndrome).

Although human exposure to *Aspergillus* is common, infections are infrequent. *Aspergillus* mastoiditis and skull-base osteomyelitis are extremely rare, even in immunocompromised patients [5], the first patient being described in 1985 [8]. We report a case of invasive *Aspergillus fumigatus* skull-base mastoiditis in an immunocompetent patient, only first recognized at surgery.

Case report

An 81-year-old man presented with a 5-week history of right-sided otalgia and otorrhoea (Table 1). His hearing in the right ear was subjectively reduced; there was no history of tinnitus or vertigo. Examination revealed whitish debris filling the right external auditory canal. The left ear was normal. His medical history included a symmetrical presbyacusis, for which he had been wearing hearing aids bilaterally since 1999. He did not take any medication. Initially, the diagnosis of a right-sided external otitis was made and the patient was treated with oral antibiotics and acetic acid ear drops.

One month later the patient returned with progressive, intermittent severe/undurable, right-sided otalgia. Examination showed some mild otorrhoea as well as a quiet, central perforation in the tympanic membrane

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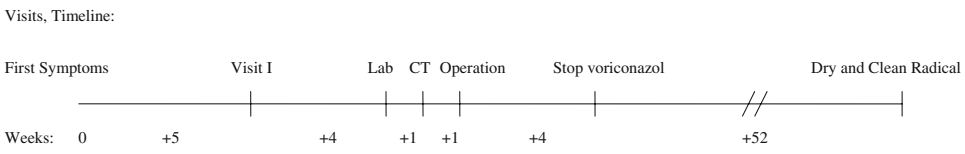
Table 1



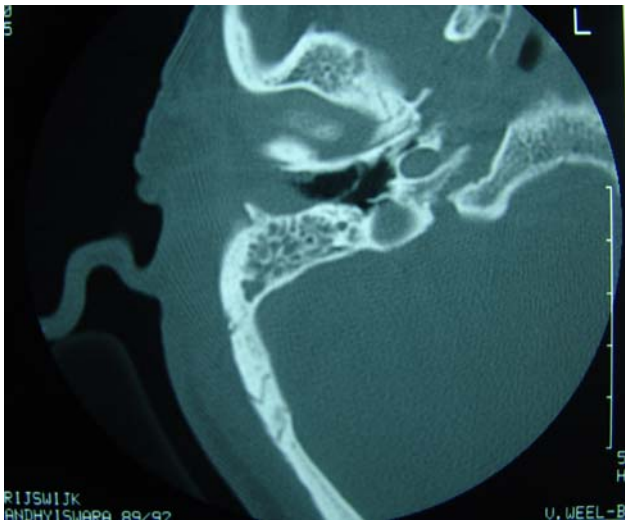


Fig. 1 Axial CT scan of the right temporal bone showing complete opacification of the mastoid air cells, an air-containing middle ear and swelling of the soft tissues surrounding the ear

with a noninflamed and air-containing middle ear space. The ear canal was slightly edematous. No granulation tissue could be seen in the external auditory canal or middle ear by microscopic examination. There was no post-auricular fluctuation and no erythema, tenderness or edema over the mastoid area. Facial function was normal. The Valsalva maneuver was negative.

Initial laboratory workup showed signs of an infection [ESR 46 mm/h, CRP 21.3 mg/l, normal WBC (8.6×10^9/l; 16% lymphocytes)]. Liver enzymes were normal. Fasting blood sugar was 6.1 mmol/l. Anti-dsDNA, ENA, anti-proteinase3 and anti-myeloperoxidase were all negative. Tone and speech audiometry showed a superimposed conductive hearing loss of the right ear. Because of the ununderstood invalidating earache (not matching with the quite external auditory canal, tympanic membrane and middle ear) a computed tomography (CT) scan was planned. This scan (1 week later) demonstrated complete opacification of the right mastoid air cells, swelling of the soft tissues surrounding the ear but no signs of bony erosion (Fig. 1).

Considering the aforementioned and the still progressive, sometimes undurable otalgia, despite the use of broad spectrum antibiotics and strong analgesics, we decided to perform a cortical mastoidectomy to obtain mastoid-material for histopathologic investigation.

At surgery subcutaneous edema and a black discoloration of the mastoid cortex and the bony ear canal was encountered. This was the first time invasive fungal mastoiditis or other bone infiltrating pathology was suspected. Therefore, we decided to perform directly a canal wall down (radical mastoidectomy) procedure to remove as much infected bone as possible and to ensure a good drainage. Purulent fluid and widespread granulation tissue filled the mastoid air cells. Intraoperative specimens of tissue were sent for culture and histopathological analysis.

After surgery an infectious disease consult was obtained to medically complement surgical eradication of the fungal infection. Postoperatively, the patient was treated with local miconazol dressing on cotton strips and oral voriconazol (200 mg twice daily) for 4 weeks.

Histolopathological analysis revealed necrosis, inflammation/granulocytes, and numerous septate fungal hyphae. Cultures of mastoid material obtained during surgery yielded *Aspergillus fumigatus.*

At follow-up visits after surgery, a well-healed incision was seen and the mastoid cavity re-epithelialized normally in several months with no evidence of residual or persistent disease by microscopic examination (last visit 1 year after surgery).

Discussion

Mycotic infections of the head and neck region are uncommon. Infections of the external auditory canal and paranasal sinuses are the most encountered sites.

The classification of *Aspergillus* infections has been described for paranasal sinus aspergillosis, and has been expanded to include temporal bone infection [5].

Documented cases of *Aspergillus* mastoiditis are exceedingly rare and all but six [1–4, 6, 7] of the reported cases to date involved immunocompromised patients.

Patients with invasive aspergillosis can have similar clinical presentation and physical findings as an infection caused by *Pseudomonas aeruginosa.* Failure to identify *Aspergillus* as the causative pathogen of invasive temporal bone infection is the principal reason for delay in initiating potentially life-saving therapy.

In our patient mainly the mastoid was involved with the infection and not so the well-aerated middle ear (Fig. 1). This

unusual finding might be explained with an obstruction of the aditus ad antrum through pathologic tissue (granulation tissue) in combination with the tympanic membrane perforation [10].

Almost all patients with documented invasive external otitis resulting from *Aspergillus* reported in literature were noted to have facial nerve involvement. Evidence of facial nerve involvement in patients with mastoiditis—particularly those who are immunocompromised and do not respond to antibiotics—should cause one to consider *Aspergillus* in the differential diagnosis [1–6, 8].

Another noteworthy co-incidence in the seven documented immunocompetent patients with *Aspergillus* mastoiditis to date (our patient included) is their old age (52–85, average 75 years) and the fact that they all had a port d'entrée to the tympanic cavity or mastoid: six had a tympanic membrane perforation [1–4, 6] and the seventh (with the age of 52) was already known with a cholesteatoma (for which she refused surgery) 9 years before the mastoiditis [7].

Treatment of *Aspergillus* mastoiditis is threefold. Aggressive surgical debridement and resection is required and antifungal therapy should be instituted once the diagnosis is made. Hyperbaric oxygen therapy should be considered post-operative although clear evidence to demonstrate effectiveness is still missing [9].

Further, attempt should be made to find and control any underlying immunologic conditions.

Conclusion

Mastoiditis caused by *Aspergillus* species is very uncommon. Because of the invasive nature of this life threatening disease, prompt diagnosis and aggressive management including surgical debridement and antifungal therapy are necessary.

When confronted with otitis/ear complaints with an unexpected clinical course a high index of suspicion is required to facilitate early diagnosis and appropriate therapy of a potential lethal *Aspergillus* infection, even in immunocompetent patients. This seems the more so in older patients with an open middle ear cavity (tympanic membrane perforation, cholesteatoma) and/or when there is facial nerve involvement.

This is for we know the seventh immunocompetent patient with invasive *Aspergillus* mastoiditis described in the world literature.

Conflict of interest statement Both authors indicate that they do not have a financial relationship with the organization that sponsored the research. Also there has not been any organization that sponsored the research.

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