CASE REPORT

Scedosporium apiospermum: a rare cause of malignant otitis externa

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SUMMARY

A 79-year-old man, with a history of well-controlled diabetes mellitus, presented with left-sided otalgia. With an initial diagnosis of simple otitis externa, he was discharged on topical drops. He represented 2 months later with worsening otalgia and discharge. A diagnosis of malignant otitis externa was made based on clinical and radiological findings. Intravenous Tazocin and Gentamicin were given based on previous bacterial culture from ear swabs. The patient failed to improve and developed left-sided facial nerve palsy. His condition stabilised following a change in antimicrobial therapy and his management continued in the community on intravenous Meropenem with twice weekly aural toilet. Repeated nuclear medicine imaging failed to demonstrate resolution. A bony sequestration was removed from the external auditory canal in the outpatient clinic, which following extended culture grew *Scedosporium apiospermum*; his management was subsequently changed to oral Voriconazole. This led to rapid clinical improvement and disease resolution over a 6-week period.

BACKGROUND

Fungal pathogens are a rare and potentially life-threatening cause of malignant otitis externa (MOE), most commonly found in immunocompromised individuals.¹ They can be difficult to isolate and treat effectively.¹ Mortality rates increase with disease spread, highlighting the importance of early diagnosis and initiation of appropriate antimicrobial therapy. With delays in recognition and treatment, infection can spread through the temporal bone causing cranial nerve palsies, intracranial abscesses and sinus thrombosis.

Aspergillus and *Candida* species are the most common causes^{1 2} of fungal MOE and treatment consists of an intravenous antifungal, often Caspofungin or Amphotericin-B. Though generally effective at treating fungal MOE, both have large side-effect profiles and significant toxicity, particularly Amphotericin-B that is known to commonly cause side effects.³

Scedosporium apiospermum can be a colonising microbe but has been identified as a causative pathogen for MOE in two previous cases,⁴ both involving patients with HIV; in this case, the patient was not HIV positive and his diabetes mellitus was well controlled, making this the first reported case of MOE caused by *S. apiospermum* in a patient without HIV. Despite the extensive spread of disease, causing conductive hearing loss and a facial nerve palsy, he was managed

conservatively with antimicrobial therapy and regular microsuction with no surgical debridement. Not only was complete resolution achieved, but facial nerve function returned to normal. This demonstrates that despite extensive disease spread, conservative management without surgical debridement can still achieve good clinical outcomes.

CASE PRESENTATION

A 79-year-old man with diabetes mellitus, treated with metformin, presented with a 3-week history of left-sided otalgia and reduced hearing. Examination showed a narrowed and erythematous external auditory canal with significant debris. He had no facial nerve palsy and was systemically well.

An ear swab was taken and, pending culture, the patient started topical Gentamicin drops. This swab grew *Pseudomonas aeruginosa* but despite focused topical antimicrobial therapy, his symptoms failed to improve. A CT scan of the temporal bones showed opacification of the mastoid air cells and middle ear, middle ear dehiscence of the facial nerve but no osteolysis (figure 1). He was discharged on oral ciprofloxacin and topical Gentamicin drops.

Two months later he re-presented, systemically unwell with worsening otalgia and discharge from the left ear. Based on previous culture, treatment involved intravenous antibiotics (Gentamicin and Tazocin) as well as continuing topical Gentamicin drops. A further CT scan of the temporal bone demonstrated opacification of the mastoid air cells, soft tissue oedema and osteolysis consistent with a diagnosis of MOE (figure 2). His condition failed to improve and, over a 2-day period, he developed a grade IV House-Brackmann left facial palsy secondary to toxic effects on facial nerve dehiscence; he experienced no other cranial nerve neuropathies. Single-photon emission CT (SPECT-CT) using Technetium-111 radioisotope showed metabolically active disease in the left temporal bone (figure 3).

Antimicrobials were changed to Meropenem and Caspofungin, with the addition of Clotrimazole eardrops. After 4 weeks of treatment, repeated imaging (CT and SPECT-CT) showed static disease. With a lack of disease progression, the patient was discharged on intravenous Meropenem to be given in the community with weekly ENT follow-up.

Regular follow-up confirmed persistent disease; a bony sequestration was removed from his external auditory canal and sent for extended culture. This grew *S. apiospermum*, a rare fungal pathogen that has caused MOE in only two previously

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Figure 1 Initial CT scan, axial view, demonstrating opacification of the middle ear and mastoid air cells but no osteolysis. Ossicles remain intact.

documented cases causing malignant otitis externa (both with HIV). In this case, the patient did not have HIV, making this possibly the first documented case of MOE caused by *S. apiospermum* in an individual without HIV. As a result of this culture, his antimicrobial therapy was changed from intravenous Meropenem to oral Voriconazole.

Rapid improvement followed. After 6 weeks of oral Voriconazole, repeat SPECT-CT showed reduced tracer uptake in the left temporal bone (figure 4). Voriconazole was stopped and he ultimately regained full facial nerve function but was left with a significant conductive hearing loss.

INVESTIGATIONS

External auditory canal swab: *P. aeruginosa* sensitive to Gentamicin. Extended bone biopsy+culture: *S. apiospermum*.

Figure 2 Repeat CT scan, axial view, demonstrating soft tissue oedema and osteolysis of the left temporal bone. Erosion of ossicles can be seen.

Figure 3 Initial SPECT-CT, axial view, demonstrating increased tracer uptake in the left temporal bone. SPECT-CT, single-photon emission CT.

Haematology and biochemistry: raised inflammatory markers which normalised with treatment.

CT temporal bones: osteolysis, soft tissue oedema and opacification of mastoid air cells and the middle ear.

SPECT-CT: tracer uptake left temporal bone.

DIFFERENTIAL DIAGNOSIS

- ▶ Bacterial malignant otitis externa.
- ▶ Fungal malignant otitis externa.

TREATMENT

- ▶ Patient stabilised on intravenous Caspofungin and Meropenem and topical eardrops; with daily microsuction to

Figure 4 Repeat SPECT-CT, axial view, demonstrating reduced tracer uptake in the left temporal bone from previous imaging. SPECT-CT, single-photon emission CT.

the left ear, his improvement plateaued and in real terms, this 'stability' referred to a lack of deterioration.

- ▶ He underwent no invasive surgical intervention, only regular aural toilet and biopsy of available tissue.
- ▶ On advice from the local microbiology team, the patient was continued on intravenous Meropenem given in the community with weekly aural toilet using Gramicidin and Neomycin ointment to span hospital visits.
- ▶ Conversion to oral Voriconazole after culture of *S. apiospermum*.

OUTCOME AND FOLLOW-UP

Patient recovered with complete resolution of facial nerve function. He suffered significant conductive hearing loss in the affected ear due to osteolysis of the ossicles and tympanic membrane obliteration. This conductive loss has been amenable to in-ear hearing aids.

Two-weekly follow-up monitored for disease recurrence over a 6-month period and the patient currently has an open follow-up to avoid any future disease being missed. The total period of oral antifungal medication was 6 weeks.

DISCUSSION

S. apiospermum is an exceptionally rare cause of MOE. Previous published cases of MOE have involved patients suffering from HIV.⁴ Excluding MOE, *S. apiospermum* has been isolated as a cause of otitis externa in other case reports and case series;^{5–8} one case of otitis externa was treated solely with Voriconazole after 6 months of failed antibiotic therapy⁵ similar to this case; however, there was no evidence of MOE.

This case highlights the difficulty of initial pathogen isolation, and also the difficulty in starting an appropriate first-line antimicrobial; fungal causes are often discovered after multiple oral and topical antibacterial preparations have been tried.⁹ It would be wrong to ascertain from this case that all patients with a diagnosis of MOE should take antifungal medication and antibiotics; however, this case suggests that it should always be a consideration in individuals not responding to broad-spectrum intravenous antibiotic administration that the causative pathogen could be fungal. Tissue sampling has been shown to be effective in isolating pathogens when initial pus swabs have been sterile;² as was the case with this individual, the causative pathogen was identified via extended culture of sequestered bone from the external auditory canal, and this has also been the situation in refractory cases of otitis externa.⁵

Other case series reviewing the management of fungal MOE have shown Voriconazole to be effective and safe in treating MOE secondary to *Aspergillus* species with fewer side effects than Amphotericin-B. It could therefore be suggested that

Voriconazole is a safe and effective oral alternative to otherwise intravenous treatment of fungal MOE.¹

Learning points

- ▶ Malignant otitis externa is a diagnosis that should be considered in cases of otitis externa that are not responding to topical treatment.
- ▶ Initial management of malignant otitis externa should include microbiology sample (pus swab for example) and broad-spectrum intravenous antibiotics, including pseudomonal cover, pending microbiological sensitivities.
- ▶ Fungal causes of malignant otitis externa, despite being rare, are more pervasive in immunocompromised individuals. A high degree of suspicion in these individuals is required so as not to delay diagnosis.⁵
- ▶ Voriconazole is a safe orally administered antifungal agent that has the potential to expedite discharge in patients with fungal malignant otitis externa.

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