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CASE REPORT

A rare cause of malignant otitis externa and skull base osteomyelitis

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Abstract

An elderly male with type 2 diabetes presented with a 2-month history of otalgia and severe headaches. He was diagnosed with malignant otitis externa (MOE) and was commenced on empirical treatment with oral ciprofloxacin. Pseudomonas is the most common cause of MOE. A baseline CT scan was undertaken that demonstrated skull base osteomyelitis (SBO) due to findings of bone erosion at the mastoid tip and an infiltrating soft tissue mass eroding the clivus. Eight weeks later, he returned with worsening and bilateral symptoms of otitis externa, hearing loss, temporomandibular pain and dysfunction. Worsening and now bilateral malignant otitis externa were confirmed with an MRI scan that also demonstrated a small fluid collection in his left temporal region. The collection was aspirated and grew scedosporium apiospermum. He was diagnosed with fungal SBO and was commenced on treatment with the antifungal voriconazole, with significant improvement in symptoms and radiological findings. Fungal osteomyelitis is more likely in immunosuppressed patients, particularly those with type 2 diabetes. Fungal aetiology should be suspected in patients with progressive symptoms, despite treatment. A microbiology diagnosis of fungal SBO or MOE can be challenging to obtain and can lead to diagnostic delay. A sampling of the external auditory canal can aid in diagnosing MOE; however, scedosporium may also be isolated as a commensal organism. Aspirations from accessible fluid collections, infratemporal fossa needle sample and bone biopsy can provide material for diagnosis. Scedosporium is a rare cause of disease in humans, however, fungal infections are increasing in humans, due to an increase in susceptible populations. Scedosporium apiospermum is a rare cause of SBO and should be considered in patients not responding to standard treatment.

Key words: Malignant otitis externa; Skull base osteomyelitis; Scedosporium apiospermum; Fungal; Otitis externa

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Introduction

An elderly male with type 2 diabetes was diagnosed with malignant otitis externa (MOE) and was treated empirically with ciprofloxacin. Eight weeks later, he returned with worsening symptoms, as indicated by imaging findings and had developed a small fluid collection in his left temporal region. The collection was aspirated and grew scedosporium apiospermum. He was diagnosed with fungal skull base osteomyelitis (SBO) and commenced on treatment with the antifungal voriconazole, with significant improvement in symptoms and radiological findings. Scedosporium apiospermum is a rare cause of SBO and should be considered in patients not responding to standard treatment.

Case report

An 83-year-old man presented with a 2-month history of left otalgia and severe headaches that radiated into his neck and mandible. He had left temporomandibular joint (TMJ) pain, hearing loss, and a loss of sense of taste. He had a background of right presbycusis, poorly controlled type 2 diabetes, atrial fibrillation and hypertension. On examination, the left ear was tender over the tragus, and the external auditory canal appeared hyperaemic with mild canal stenosis and some debris, consistent with a diagnosis of otitis externa. A swab was taken with no bacterial growth on the culture. His blood results confirmed raised inflammatory markers (WCC 10.84×10^9/L, neutrophils 8.25×10^9/L, CRP 63 mg/L.)

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Case report (Continued)

The patient was diagnosed preliminarily with MOE. A computed tomography (CT) scan was undertaken (Figures 1 and 2) which demonstrated opacification of the left mastoid air cell complex and middle ear, with bone erosion at the mastoid tip and an infiltrating soft tissue mass eroding the clivus in the retropharyngeal region and extending into the left infratemporal region involving the left TMJ. A baseline magnetic resonance (MR) imaging study was also performed, confirming the CT findings. He was subsequently admitted to the hospital for osteomyelitis management and started on oral ciprofloxacin. He was discharged after 2 weeks in hospital with an 8-week course of ciprofloxacin.

At 8 weeks, the patient was reviewed in the clinic. He now complained of right otalgia and increasing left TMJ pain. His headaches had deteriorated, and he was now functionally deaf on audiology assessment. On examination, there was additional evidence of right otitis externa, the right mandible was tender on palpation, and he had malocclusion. All these findings were in keeping with a diagnosis of bilateral MOE and progressive SBO. He was readmitted to the hospital for intravenous antibiotic treatment and was commenced on intravenous vancomycin and meropenem.

An MR study was repeated (Figure 3). This showed the progression of SBO and the development of a small fluid collection in the left temporal region. Ultrasound-guided aspiration was performed and 3 ml of mucopurulent material was aspirated. The purulent material grew the fungus scedosporium apiospermum, indicating a fungal aetiology. Treatment was changed to the antifungal voriconazole orally, 200 mg twice a day.

He was discharged on a 6-month course with planned regular follow-up and imaging. At 8 weeks there was significant improvement in his symptoms, his bilateral otitis externa had resolved and otalgia, headaches and TMJ function all improved. Interval MR imaging confirmed an excellent response to treatment.

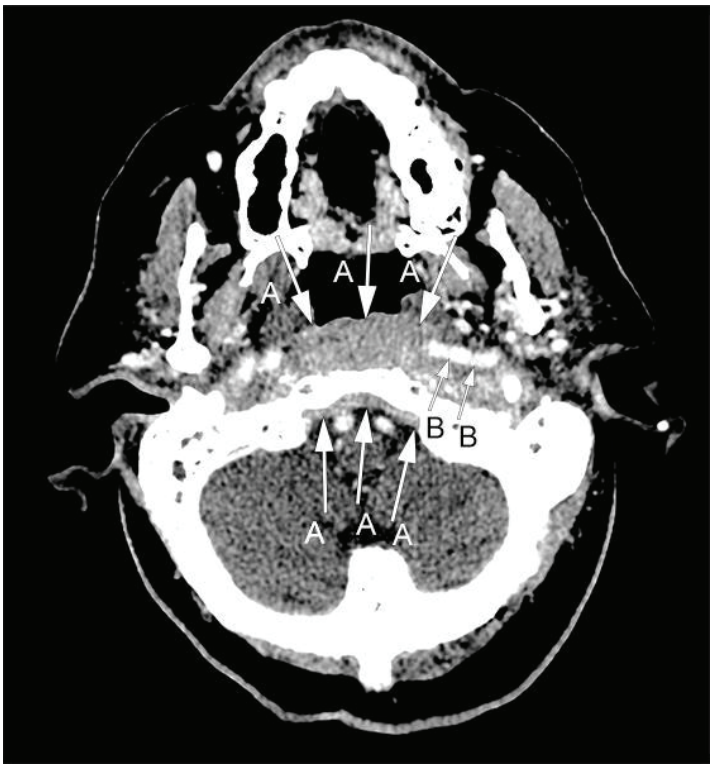

Figure 1. Axial computed tomography image post-contrast administration at the level of the skull base confirms an extensive soft tissue mass (A) in the prevertebral region and extending posteriorly adjacent to the vertebral arteries. The mass involves the left petrous temporal bone and encases the left internal carotid artery (B).

Discussion

Named for its invasive nature and elevated mortality rate, MOE is a debilitating and potentially fatal infection affecting the external auditory canal and skull base (Auinger

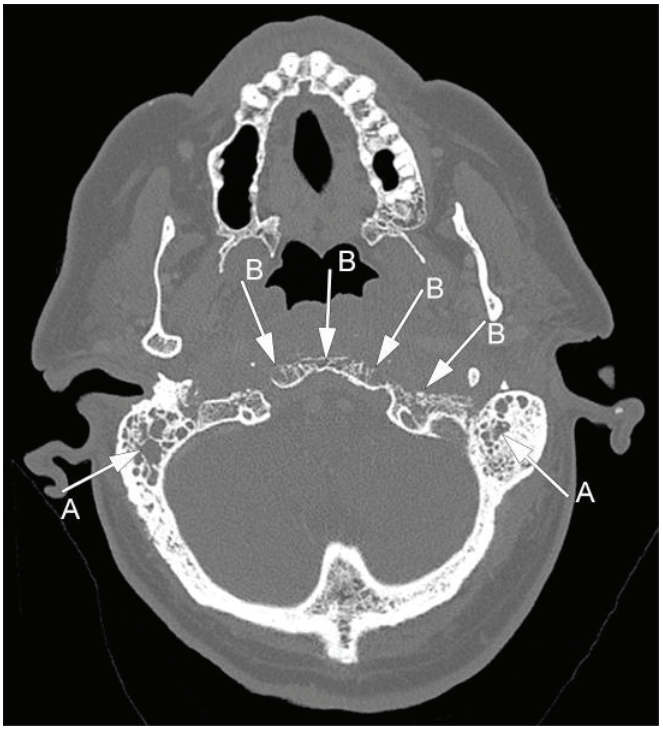

Figure 2. Axial computed tomography image on bone window setting at the same level as Figure 1, note bilateral opacified mastoid air cell complexes (A). There is diffuse erosion/destruction of the skull bone adjacent to the mass involving the left petrous temporal bone and clivus (B).

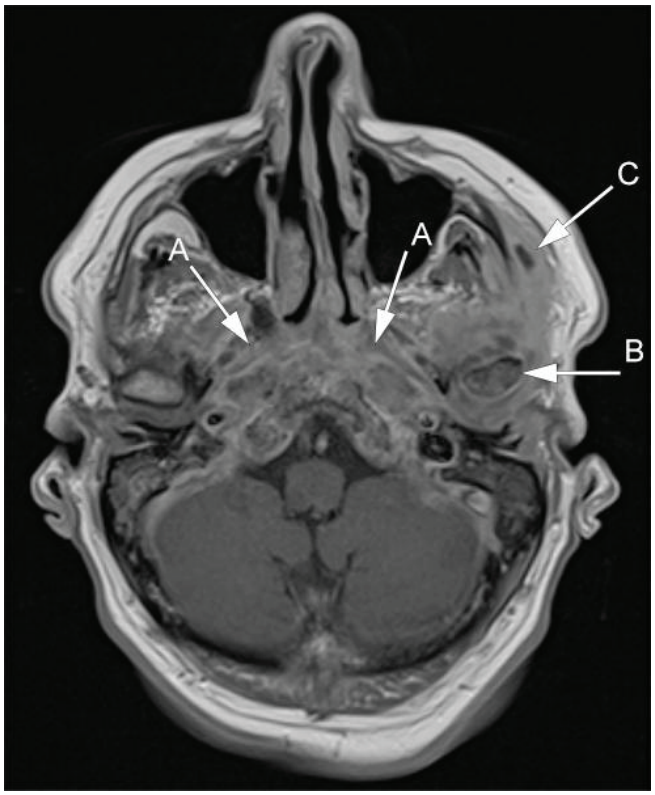

Figure 3. Axial T1-W post-contrast fat saturated magnetic resonance image at a similar level to computed tomography image. This illustrates diffuse abnormal soft tissue and pathological enhancement in relation to the skull base/clivus (A), infiltrating into the left infratemporal region/temporomandibular joint (B=left mandibular-condyle head). Note the small left temporal fluid collection (C) which was aspirated.

et al, 2021). It causes deafness by destroying the auditory ossicles and can involve the facial nerve (Yao and Messner, 2001). A poor prognosis of MOE is likely in patients with bilateral symptoms and cranial nerve involvement (Eveleigh et al, 2009). Distinguishing features of MOE include pain that worsens at night, severe temporal and/or occipital headaches and prolonged otitis externa despite treatment (Mion et al, 2015)

SBO occurs when the infection spreads to involve the sphenoid, occiput, and clivus (Khan et al, 2018). SBO usually occurs in elderly patients who are immunocompromised or have type 2 diabetes (Auinger et al, 2021). Clinically bacterial and fungal SBO are indistinguishable from each other. Bacterial SBO caused by MOE is significantly more common, pseudomonas aeruginosa accounting for 80% of cases (Prasad et al, 2014). Fungal SBO incidence is rare, caused primarily by aspergillus then candida (Khan et al, 2018). Treatment is started empirically with the fluroquinolone ciprofloxacin (Yao and Messner, 2001). Often, it is difficult to obtain tissue or fluid samples for a definitive diagnosis (Auinger et al, 2021).

Scedosporium apiospermum, a ubiquitous environmental mould, is now considered a major human pathogen, and is one of the most common moulds that can cause infections in humans (Koehler et al, 2014). It is an opportunistic infection that causes invasive fungal disease, especially in immunocompromised patients. First line treatment is with voriconazole (Koehler et al, 2014; Huguenin et al, 2015). Monitoring of disease response should include imaging, a multidisciplinary team and the clinical presentation of the patient (Auinger et al, 2021).

The common presentation of otitis externa caused by scedosporium is chronic otitis externa lasting months or years that does not respond to usual treatments (McLaren and Potter, 2016). Obtaining a microbiological diagnosis can be challenging and this often leads to diagnosis delay (McLaren and Potter, 2016; Huguenin et al, 2015). A sampling of the external auditory canal can aid in diagnosing MOE; however, scedosporium may also be isolated as a commensal organism. Aspirations from accessible fluid collections, infratemporal fossa needle samples and bone biopsy can provide material for diagnosis (Huguenin et al, 2015; McLaren and Potter, 2016).

Learning points

- MOE is a life-threatening infection and requires prolonged treatment with antimicrobials.
- Most cases are bacterial and treated empirically with antibiotics.
- Although fungal MOE is rare, a failure to respond to treatment should alert the clinician to this possibility and the anti-fungal treatment instigated.

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Availability of data and materials

All the data of this study are included in this article.

Author contributions

MAH identified the case and was the principal clinician for the patient. FH collected the data and wrote the first draft of the case report. MAH and DH collected the data and annotated the images and was the principal editor. All authors made editorial changes to the manuscript. All authors read and approved the final manuscript. All authors participated sufficiently in the work and agreed to be accountable for all aspects of the work.

Ethics approval and consent to participate

The participant signed an informed consent form.

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Conflict of interest

The authors have no conflicts of interest to declare.

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