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Case Report

Beyond the Ear, the Hidden Threat of *Paecilomyces* Neuro-Otological Infection: A Case Report

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BACKGROUND: *Paecilomyces* spp. are thermo-tolerant fungi found in decaying vegetables and soil. They can cause ocular, cutaneous, and miscellaneous infections. In otorhinolaryngology, most infections occur in the paranasal sinuses, while otological infection is uncommon.

METHODS: We report a case of 41-year-old diabetic, male army officer with warm, fluctuant left temporal swelling and reduced hearing. Otoscopy revealed an edematous external auditory canal (EAC) with posterior wall sagging and bulging tympanic membrane. Computed tomography revealed subperiosteal temporal abscess extending intracranially. The pus was drained surgically via an otological and a neurosurgical approach, and pus sent for culture grew *paecilomyces*. He showed clinical improvement after receiving oral antifungal treatment postoperatively. Retrospectively, his occupation as an army officer and his diabetic immunocompromised state may have predisposed him to the infection.

RESULTS: *Paecilomyces* middle ear infection leading to intracranial involvement of such magnitude is yet to be reported, and we showcase its successful management through a combined surgical neuro-otology approach and oral antifungal therapy. Fungal ear infections can lead to severe extracranial and intracranial complications if inadequately treated. Differentiating it from cholesteatoma also presents a diagnostic challenge clinically and radiologically. While both can lead to intracranial complications, our patient's brief history and lack of prior ear symptoms contrast with that of cholesteatoma.

CONCLUSION: The rarity of neuro-otological *paecilomyces* infections emphasizes the need for awareness and early identification. It is vital to recognize such infections, and prompt surgical management with appropriate antifungal drugs is warranted to prevent disastrous outcomes.

KEYWORDS: Abscess, mastoiditis, neurosurgery, otology, paecilomyces

INTRODUCTION

Paecilomyces spp. are filamentous and thermo-tolerant fungi found in decaying vegetables and soil.[1,2] It is known to cause human infections, such as ocular, cutaneous, subcutaneous, and other miscellaneous infections.[3] Miscellaneous infections include sinusitis, onychomycosis, lung abscess, pleural effusion, osteomyelitis, and disseminated infection.[1,3]

In otorhinolaryngology, most paecilomyces infections occur in the paranasal sinuses, such as maxillary and sphenoid sinuses.[4-8] Infection in immunocompetent and immunocompromised individuals can lead to severe consequences.[3] Surgical debridement in combination with antifungal drug therapy is the usual management.[3] Apart from a single otology case of chronic suppurative otitis media (CSOM) caused by a paecilomyces infection which was reported in India, to the best of our knowledge no other otological case has been reported.[9] We would like to report the first paecilomyces infection with neuro-otological involvement, which was managed via a combined surgical neuro-otology approach and oral antifungal therapy with clinical success.

CASE PRESENTATION

A 41-year-old male with underlying diabetes mellitus on tablet metformin, presented to us with left temporal swelling and reduced left-sided hearing of 3 weeks duration. He had no prior history of recurrent ear discharge or infection. Clinical examination revealed a left temporal swelling measuring 4 cm × 4 cm which was erythematous, warm, and fluctuant (Figure 1). Otoscopy revealed an

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edematous external auditory canal (EAC) with posterior wall sagging and bulging tympanic membrane. Facial nerve function was intact. The audiological assessment showed a type B tympanogram with left mild-to-severe mixed hearing loss on pure tone audiometry (Figure 2). Under the cover of intravenous cefuroxime, an urgent high-resolution computed tomography (HRCT) of the temporal bone revealed bony erosions within the mastoid portion of the left temporal bone, along with soft tissue attenuation within the left middle ear cavity and eroded left ossicles (Figure 3A and B). Axial and coronal view of contrast-enhanced CT in soft tissue window illustrated peripherally enhancing left temporoparietal scalp lesion with an extradural involvement (Figure 4A and B).

His blood viral screening of Hepatitis B, Hepatitis C, and Human Immunodeficiency Virus (HIV) infections were negative.

Following the posterior auricular incision by the otology team, 50 cc pus was drained from the subperiosteal layer over the temporal and post-auricular region. Temporalis muscle had necrosed and had sloughed with underlying osteomyelitic bone. The neurosurgical team then performed a left craniotomy and burr hole drainage to evacuate frank pus from the temporal base. The pus was found at the extradural surface with the dura intact. There was no keratin material seen intraoperatively. Postoperatively, a diagnosis of a subperiosteal temporal abscess (Luc's abscess) secondary to left otomastoiditis with intracranial involvement was made. Subsequently, a pus culture sent intraoperatively grew paecilomyces spp.

He was started on oral voriconazole of 400 mg twice daily for 1 day, and 200 mg twice daily for 4 days. He was then continued on oral posaconazole 300 mg twice daily for 1 day, followed by 300 mg once daily for 2 weeks. Subsequent follow-ups showed clinical improvement and a repeat HRCT temporal bone 2 months post-treatment showed post-operative changes with no evidence of new collection. Post-operatively, his hearing level showed left mild mixed hearing loss, which was an improvement compared to the initial presentation (Figure 5). He has since remained disease free at 6 months follow-up at the time of writing. Written informed consent was obtained from the patient for anonymized information to be published in this article.

MAIN POINTS

- We share our encounter of the first paecilomyces infection with neuro-otological involvement, that we have managed successfully in cooperation with our neurosurgery colleagues.
- We would also like to highlight the diagnostic challenge encountered in managing our case, in terms of differentiating it from a case of cholesteatoma, as cholesteatoma can present similarly with associated intracranial complications.
- Despite disease diagnoses in the modern days being aided with radiological investigation, computed tomography finding in our case gave a false impression of a cholesteatoma in view of bone erosion.
- Nevertheless, the intraoperative and microbiological findings assisted us in reaching a final diagnosis, which was further justified by the fact that the patient has responded clinically and audiologically to the post-surgical antifungal therapy.

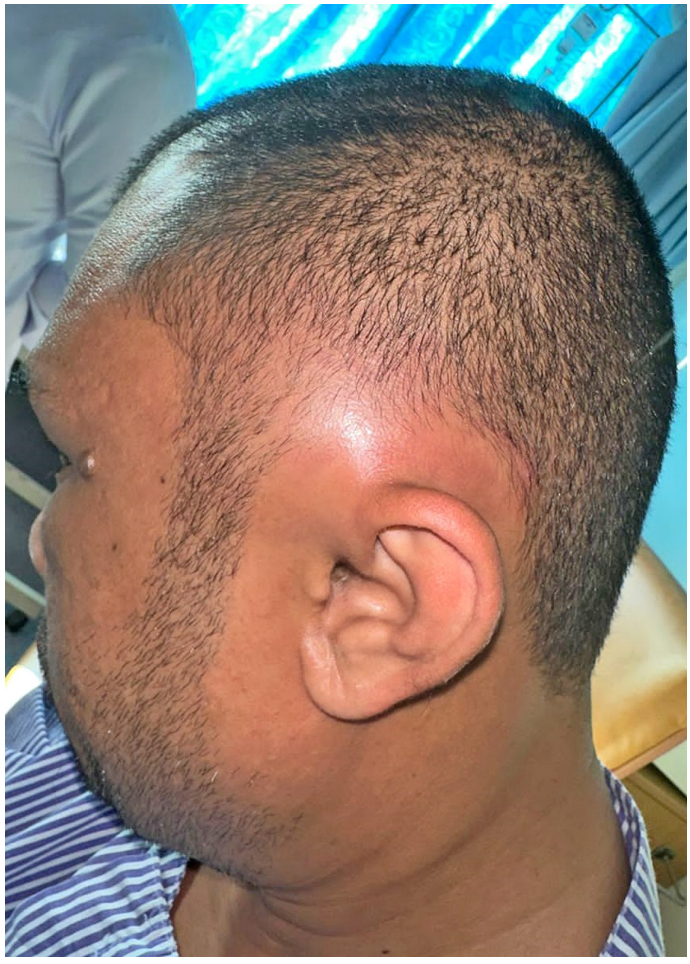

Figure 1. Clinical picture showing the left temporal swelling.

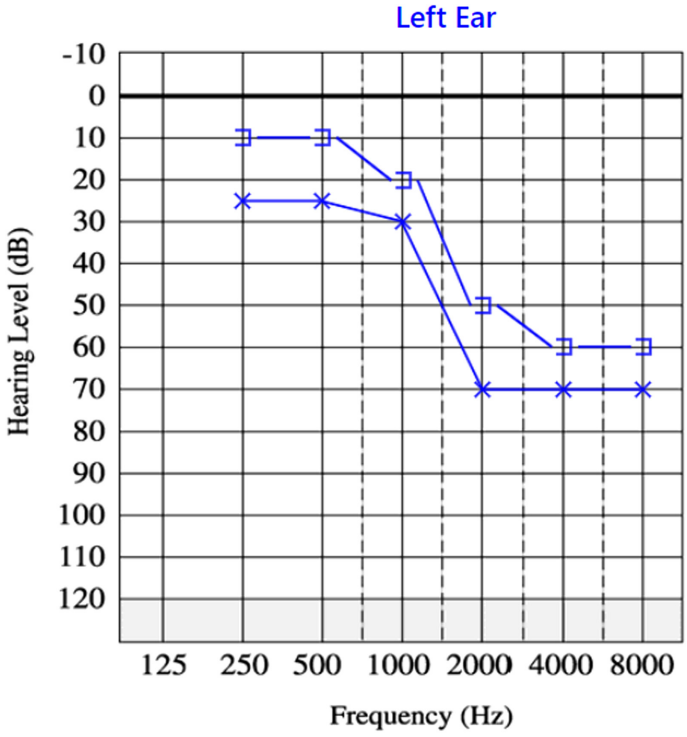

Figure 2. Pure tone audiometry (PTA) showing the left mild-to-severe mixed hearing loss.

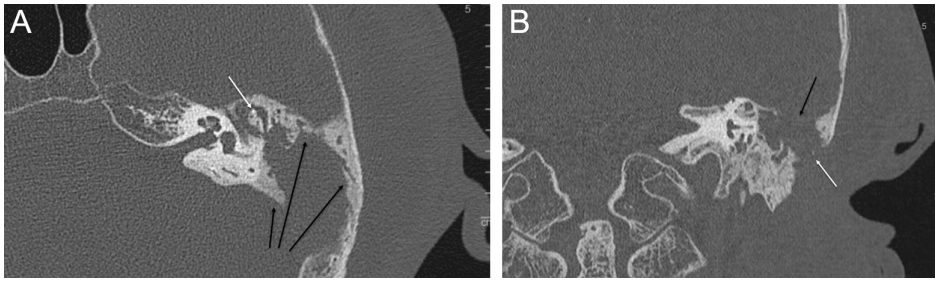

Figure 3. A, B. (A) Axial view of high-resolution computed tomography (HRCT) of the left temporal bone in the bone window showing bony erosions within the mastoid part of the left temporal bone (black arrows), soft tissue attenuation within the left middle ear cavity, and eroded left ossicles (white arrow). (B) Coronal view showing similar findings with eroded tegmen mastoideum (black arrow) and eroded mastoid tip (white arrow).

DISCUSSION

Paecilomyces spp. is a hyaline hyphomycete known to cause various infections in immunocompromised and immunocompetent individuals. Various species causing human infections include *Paecilomyces lilacinus*, *Paecilomyces variotti*, *Paecilomyces marquandii* and *Paecilomyces javanicus*.[2,3] Infections can occur following direct inoculation, eye contact, indwelling catheters or inhalation.[3] Common infections include ocular and cutaneous infections. Ocular infections are predisposed by intra-ocular lens implantation, non-surgical trauma, and contact lens usage.[3] For cutaneous and subcutaneous infections, predisposing factors include solid organ and bone marrow transplantation, corticosteroid therapy, malignancies, primary immunodeficiency, retroviral diseases, and diabetes mellitus.[3] In these individuals, fungal colonization of clinical materials like catheters and plastic implants serve as entry routes.[3] Among miscellaneous non-ocular, non-cutaneous infections, sinusitis is the commonest.[3] In otorhinolaryngology, paecilomyces infections involving paranasal sinuses like maxillary and sphenoid sinuses were reported.[4-8]

For our patient, diabetes mellitus and his occupation as an army officer might have predisposed him to the infection.[2,3] His harsh working environment could have exposed him frequently to soil contaminated with the fungus, creating opportunities for the fungus to gain access and overcome his immune system. His infection likely originated from the middle ear, which then proceeded to erode the skull base, specifically the tegmen tympani and tegmen mastoideum. With regards to the portal of entry, the inhalation route explains cases of sinusitis, while a perforated tympanic membrane likely provided entry in the reported case of CSOM.[9] Clinical findings of intact tympanic membrane with no prior skin trauma in our patient negates the possibility of the fungal entry through EAC or from a skin breach. Hence the only anatomically feasible entry route into the middle ear would be via inhalation via the eustachian tube. However, with paranasal sinuses uninvolved, it is still unfathomed how middle ear infection with intracranial complication could have occurred without causing any significant infection in the paranasal sinuses. The hematological route is also a possible route of infection, however, his blood culture taken did not grow anything. Nonetheless, we are thankful to be able to manage the disease prior to it causing a severe neurological or labyrinthine complication.

Paecilomyces infections of other anatomical regions are managed with surgical intervention and/or antifungal therapy with various degrees of success.[3] Antifungals are used as a single agent or in combinations.[2,3] They include amphotericin B, flucytosine, fluconazole, miconazole, itraconazole, caspofungin, terbinafine, voriconazole and ketoconazole.[2,3] Due to Paecilomyces' low susceptibility and poor clinical outcome to conventional antifungal drugs, recent literature reports the application of novel triazoles, such as ravuconazole, voriconazole, and posaconazole with better clinical outcomes, with or without surgical intervention.[3,8,10,11] Besides surgical debridement and antifungal therapy, management of predisposing factors is also vital.[3] We believe the optimization of our patient's sugar control, our management via combined surgical debridement and dual oral antifungal courses, has aided his clinical success.

Apart from a single case of CSOM, there have been no other cases reported in the literature.[9] Our case was rather severe and this has not been reported before. Our case required the combined efforts of both the otology and neurosurgery teams to evacuate the pus. Since fungal sinusitis is more frequently reported, awareness of complications of invasive fungal sinusitis is naturally higher, especially in immunocompromised individuals. Possible complications include orbital, cavernous sinus, and intracranial involvement.[12] However, fungal ear infections such as ours should not be underestimated, as various intratemporal and intracranial complications can be similarly debilitating if not treated adequately. The diagnostic challenge

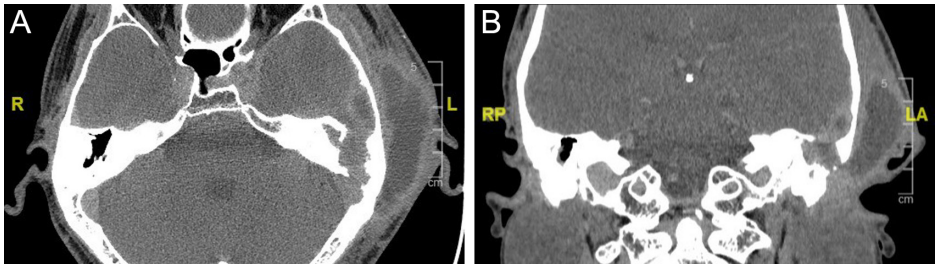

Figure 4. A, B. (A) Axial view of contrast-enhanced computed tomography (CECT) of left temporal bone in soft tissue window showing large, well-defined left temporoparietal peripherally enhancing scalp collection with extradural collection. (B) Coronal view showing similar findings, with visualized involvement of similar enhancing collection within the mastoid part of the left temporal bone with bony erosions.

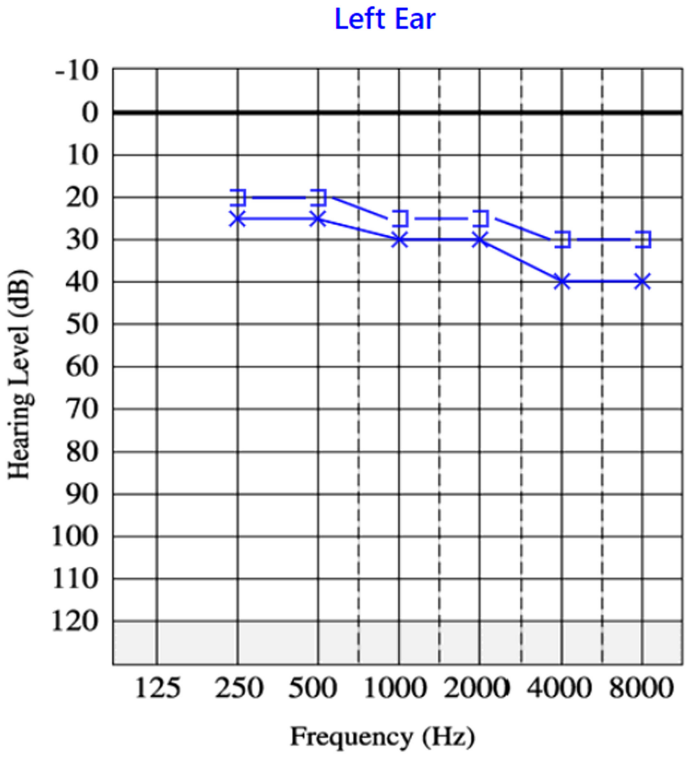

Figure 5. Pure tone audiometry (PTA) showing the left mild mixed hearing loss.

encountered in managing our case is the differentiation from cholesteatoma, as cholesteatoma can present similarly with associated intracranial complications.[13] However, our patient had a very short history prior to presentation with no previous ear symptoms. This is in contrast with cholesteatoma, where patients usually present with a chronic discharging ear that eventually leads to intracranial complications.[13] Radiologically, CT findings may falsely give the impression of a cholesteatoma due to bone erosion, however as reported above, our intraoperative findings differed.[13]

In conclusion, Paecilomyces infection is uncommon in otorhinolaryngology. While few paranasal sinus infections have been reported, we presume our encounter with neuro-otological paecilomyces infection is the first one reported. We hope our experience will increase awareness among Otolaryngologists-Head & Neck Surgeons and Neurosurgeons, with regards to the possibility of an extensive middle ear fungal infection. In view of the better response of paecilomyces infection to only certain antifungal drugs, constant awareness, and early identification are vital to prevent a potentially disastrous outcome.

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