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Clinical Record

Septate fungal invasion in masked mastoiditis: a diagnostic dilemma

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Abstract

Invasive fungal mastoiditis is a rare entity, seen almost entirely in immunocompromized patients. It has been reported primarily in patients with leukaemia and more recently with acquired immunodeficiency syndrome. A literature search revealed only a few reports in diabetic patients, in whom the invasive fungus was identified as mucormycosis in all cases. We report the first case in the English literature of invasive septate fungal mastoiditis in a diabetic patient with intact tympanic membranes.

Key words: Mastoiditis; Aspergillus; Facial Paralysis; Diabetes Mellitus

Case report

A 59-year-old man from north India, a known diabetic for two years, presented complaining of fatigue, unsteadiness and right-sided otalgia for two months and right facial paresis for two weeks. Otalgia was initially acute, then later became a dull ache following treatment with various broad spectrum antibiotics. The hearing in the patient's right ear was subjectively reduced. There was no history of otorrhoea, vertigo or tinnitus.

Examination revealed a House-Brackmann grade four right facial palsy. Eye movements and corneal reflexes were normal and sensation over the cheek was intact. Otoscopic examination of the right tympanic membrane showed a mild hyperaemia and bulge in the pars flaccida and posterior superior quadrant of the pars tensa, with tenderness over the right mastoid. A pure tone audiogram showed a profound mixed hearing loss in the right ear. Magnetic resonance imaging (Figure 1) revealed high signal intensity in the middle ear and mastoid, suggesting an inflammatory process.

In view of the above findings, mastoid exploration followed by canal wall down mastoidectomy were performed. The facial canal was dehiscent, with two soft, red tissue masses appearing to infiltrate the nerve sheath, one involving the horizontal course and the other the vertical course of the nerve. The ossicles were normal and no pus or cholesteatoma was noted in the middle ear.

Histopathological examination revealed inflammatory granulation tissue and a spicule of necrotic bone. Septate fungal filaments suggestive of aspergillus were seen infiltrating the bony tissue (Figure 2). Following surgery, the patient was symptomatically better and was commenced on oral itraconazole. He was reviewed after two weeks and at that time his mastoid cavity was clinically healthy and healing well. There was no immediate improvement in the facial palsy.

Discussion

Otomycosis refers to superficial fungal infection of the external ear, middle ear or open mastoid cavity and may account for up to 6 per cent of patients presenting with ear symptoms in an out-patient clinic.[1] In contrast, fungal mastoiditis is a very rare, invasive otomycosis variant. The routes of entry to the temporal bone may be tympanogenic, meningogenic, haematogenic and nasopharyngeal.[2] A tympanogenic route is thought to exist when infection spreads from the external canal or middle ear into the temporal bone.[2] Meningogenic temporal bone invasion follows meningitis via the internal auditory canal, while haematogenic temporal bone mycosis follows fungaemia. Nasopharyngeal spread along the eustachian tube has been described in mucormycosis.[2]

In our patient, the only possible route of entry of fungus would have been via the eustachian tube, as he had an intact tympanic membrane and no symptoms or signs of meningitis or septicaemia. Different modes of entry of fungal spores into the middle ear via the eustachian tube have been proposed. Strauss and Fine[3] proposed adenoid colonization followed by reflux otitis media as a means of entry for fungal spores. Patients with acquired immunodeficiency syndrome (AIDS) often develop lymphoid hyperplasia which includes the adenoids. This may lead to eustachian tube obstruction, with secondary serous or acute otitis media. The anaerobic conditions so produced, along with host immune-incompetence, favour fungal colonization of the middle-ear cleft via the eustachian tube.[4]

Aspergillus is the most common fungal pathogen in immunocompromized patients. Over 90 per cent of cases of aspergillosis in the immunocompromized occur in the lung, whereas the incidence of invasive aspergillus sinusitis is about 4 per cent.[5] In immunocompetent hosts with chronic suppurative otitis media, aspergillus has been

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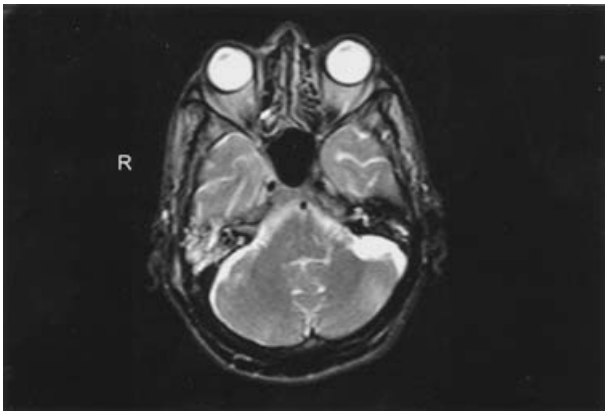

FIG. 1
Axial T2-weighted magnetic resonance image shows high signal intensity in the middle ear and mastoid on the right side.

cultured from the middle ear[6] and also from mastoid granulation[7] but has never been found to invade underlying bone.[8] Invasive fungal mastoiditis has been reported primarily in patients with leukaemia and AIDS.[2–4] The few reports of cases in diabetic hosts have always involved mucormycosis.[9,10] The current case is the first report of a septate fungal mastoiditis in a diabetic host, following spread of infection from the nasopharynx.

Clinically, our patient was diagnosed with masked mastoiditis following prolonged treatment elsewhere with various broad spectrum antibiotics. Had there been a high index of suspicion of fungal mastoiditis, he would have undergone mastoid exploration earlier, which may have prevented facial paralysis.

As fungal involvement was not suspected during surgery, tissue was not sent for fungal culture. Histopathological examination revealed septate fungal filaments, suggestive of aspergillus, infiltrating the bony tissue. In retrospect, had we sent a sample for culture, we would have been able to identify the species correctly.

Treatment of these mycoses consists of three parts: control of underlying immunologic conditions; surgical debridement of necrotic tissues; and antifungal chemotherapy. Amphotericin B is the most reliable agent against most fungal pathogens, including aspergillus.[11] Dupont,[12] in a study of 49 patients with aspergillosis, reported that itraconazole in doses of 200 to 400 mg once daily could be an alternative to amphotericin B in the treatment of invasive aspergillosis. Our patient was advised to commence a course of intravenous amphotericin B, but as he lived far away he could not stay for the complete treatment. The patient was thus commenced on oral itraconazole 200 mg daily, to be taken for one year.

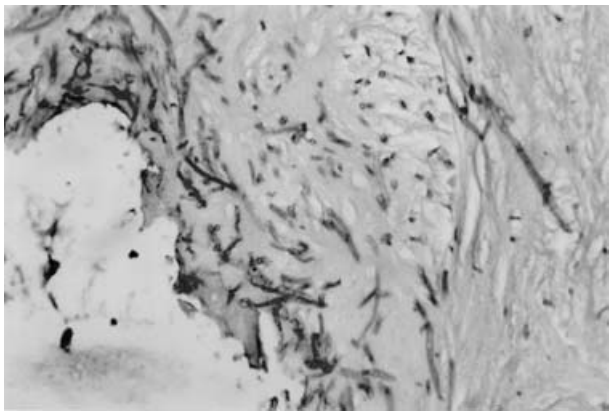

FIG. 2
Septate, branching fungal filaments in necrotic bone. (PAS; ×400)

In diabetic patients presenting with masked mastoiditis and facial palsy with no cholesteatoma detected during mastoid exploration, we recommend sending tissue for fungal culture along with histopathological examination. A high degree of suspicion is required to ensure an early diagnosis and effective treatment of this potentially lethal infection.

Conclusion

Invasive fungal mastoiditis is a rare entity seen almost entirely in immunocompromized patients. It has been reported mostly in patients with leukaemia and AIDS. The few reports in diabetic patients have always involved mucormycosis. This report aims to alert clinicians to invasive fungal mastoiditis as a differential diagnosis in diabetic patients with masked mastoiditis and facial paresis who are not responding to broad spectrum antibiotic therapy. We also emphasize the need to send tissue for fungal culture, especially when no cholesteatoma has been detected during mastoid exploration.

- **Invasive fungal mastoiditis is a rare entity, seen almost entirely in immunocompromized patients. It has been reported primarily in patients with leukaemia and more recently with acquired immunodeficiency syndrome (AIDS)**
- **In this case report, masked fungal mastoiditis caused by aspergillus spp presented with a facial paralysis and an intact tympanic membrane**
- **Treatment was with mastoidectomy followed by systemic itraconazole**
- **The diagnosis and treatment of fungal mastoiditis is discussed**

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Dr G Kuruvilla takes responsibility for the integrity of the content of the paper.
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