Fungal Otomastoiditis: A Case Series in Immunocompetent Adults

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Abstract Fungal otomastoiditis is a rare condition and is usually associated with host immunodeficiency. It is difficult to diagnose, and many cases are fatal. Treatment consists of surgical debridement, attempts to control the underlying immunological condition and antifungal chemotherapy. Individual case reports in immunocompetent patients have been published previously. We report a case series of seven with fungal otomastoiditis, all in immunocompetent patients.

Keywords Mastoiditis · Mucormycosis · *Aspergillus* · Otitis media

Introduction

Otomycosis refers to superficial fungal infection of the external ear, middle ear or open mastoid cavity and may account for up to six percentage of patients presenting with ear symptoms in an outpatient clinic [1]. Fungal infection in the ear is almost always restricted to the external auditory canal. Fungal otomastoiditis is a rare but reported entity, which is usually seen in immuno-compromised patients [2]. It is a potentially lethal infection, even in immunocompetent patients. A high index of suspicion is required to facilitate early diagnosis and appropriate therapy when confronted with otitis with an unexpected clinical course. This is more so in elderly patient with an open middle ear cavity or when there is facial nerve involvement

[3]. We report a series of seven cases of fungal tympanomastoiditis, all occurring in immunocompetent adults.

Case Reports

From November 2009 to July 2011, 227 ear surgeries were performed at our hospital and granulations and soft tissue from middle ear and mastoid was sent for histopathological examination in all the cases. A histopathology report of fungal otomastoiditis was obtained in seven patients. Patient details are presented in Table 1.

Discussion

Invasive fungal infection is widely reported in rhinologic literature, but sparsely in otologic [4]. Invasive temporal bone infection is a rare but serious entity most commonly seen in immunocompromised patients. *Aspergillus fumigatus* is the most frequently isolated species. Other species found are *Mucor*, *Blastomyces*, *Scedosporium apiospermum*, and *Lecythophora hoffmani*. In our series, five patients had *Aspergillus* infection, two patients had an associated Mucormycosis.

The route of entry of fungal infection to middle ear may be tympanogenic, meningogenic, haematogenic or nasopharyngeal [5]. The Porte de entrée to tympanic cavity and middle ear in all the seven cases reported here could be through the perforated tympanic membrane.

Hall and Farrior classified *Aspergillus* infection in temporal bone [6, 7] as three types:

- (i) Non invasive-localized and does not invade tissue, responds to conservative removal.

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Table 1 Patient details

	Case 1	Case 2	Case 3	Case 4	Case 5	Case 6	Case 7
Age	42 years	56	55	64	23	57 years	49 years
Immunity status	Immunocompetent	Immunocompetent	Immunocompetent	Immunocompetent	Immunocompetent	Immunocompetent	Immunocompetent
Chief complaints	Otalgia (L)-2 years	Otalgia-2 weeks Otorrhoea B/L-20 years	Otalgia (L)-3 months Giddiness-1 weeks	Otalgia and otorrhoea-2 years Facial weakness (R)-2 months Giddiness-3 days	Otalgia (L)-1 month	Otorrhoea and otalgia (R)-20 years	Giddiness, headache, and worsening of hearing loss B/L-3 weeks
Past history	Nil	Nil	Ear surgery right-30 years back Postero-superior marginal perforation with deep postero-superior retraction pocket-left Romberg's-negative	B/L ear surgery-20 years back Necrotic TM and deep canal skin and ossicles-right Nystagmus Romberg's-swaying to right LMN facial palsy-right-Grade IV	Nil	B/L ear surgery 20 years back TM-bilateral central perforation	B/L ear surgery
Clinical findings	Necrotic TM deep canal skin and ossicles-left	Central perforation with deep postero-superior retraction pocket with cholesteatoma bilateral					TM-large marginal perforation with cholesteatoma-right Total perforation with necrotic annulus and ossicles-left Romberg's negative
PTA	Mixed mod hearing loss-left	R-mod mixed hearing loss left-mild CHL	Right-mod SNHL profound SNHL-left	Profound hearing loss B/L	Mild conductive hearing loss B/L	B/L moderate mixed hearing loss	Right-profound hearing loss Left-severe hearing loss
HRCT-temporal bone	Soft tissue attenuation in left EAC with involvement of ossicles, oval window mastoid air cells	Not done	Not done	Soft tissue attenuation in right EAC, middle ear with erosion of facial canal	Not done	Not done	Bilateral chronic mastoiditis with otitis media
Management	EUM and debridement-refused further treatment	Canal wall down tympanoplasty-right	Canal wall down tympanoplasty-left	Modified radical mastoidectomy with facial nerve decompression-right	Canal wall down tympanoplasty-left	Cortical mastoidectomy and type I tympanoplasty	Canal wall down tympanoplasty left
HPR	Otomycosis	Cholesteatoma with filamentous fungal hyphae	Non-specific chronic inflammation with filamentous fungal colony	Cholesteatoma with filamentous fungal hyphae (Fig. 1)	Cholesteatoma with colonies of filamentous fungi and yeast like bodies	Otomycosis middle ear	Chronic otitis media with filamentous fungal colonies left ear <i>Aspergillus flavus</i>
Fungal culture	<i>Aspergillus fumigatus</i> and <i>Mucormycosis</i>	<i>Aspergillus flavus</i>	<i>Aspergillus fumigatus</i>	Mucormycosis and <i>Aspergillus fumigatus</i>	Not done	Not done	
Follow up	Lost follow up	1 year 6 months	1 year 5 months	1 year 4 months	11 months	4 months	2 months

Fig. 1 Photomicrograph showing fungal hyphae in H and E stain preparation at $\times 100$ magnification (oil immersion)

- (ii) Invasive-bony invasion with granulomatous response and fibrosis, occurs in immunocompetent patients.
- (iii) Fulminant-tissue and angioinvasion with no granulomatous response, occurs in immunocompromised patients.

In immunocompetent host, spread of fungal spores is controlled by innate immune factors such as phagocytic activity. In our series all cases had history of prolonged use of topical antibiotic -steroid medication which might have hampered the innate immunity and led to the spread of fungal infection.

The diagnosis of invasive fungal temporal bone infection is difficult and requires a high index of suspicion. In patients who have prolonged otorrhoea and rapidly progressing audio vestibular symptoms and signs in spite of adequate antibacterial therapy, possibility of fungal infection should be considered. Insufficient and delayed epithelization of the mastoid cavity post operatively can be due to such a disease. In our series fungal culture was sent for cases with such a history and those patients who per-operatively had extensive granulations with necrotic bone and ossicles. A single positive culture for *Aspergillus* is neither sensitive nor specific for invasive aspergillosis unless it was isolated from the blood stream or a deep tissue biopsy [5]. In our cases a biopsy and culture of granulations from the mastoid was done for cases 2, 3, 4, and 7 and from middle ear in case 1. In addition to culture, histopathological examination and temporal bone computerized tomography, bone scanning and PET-scan can help in the diagnosis [8]. Bone scan can demonstrate the osteoblastic reaction secondary to osteitis by high uptake of the isotope in the involved mastoid. PET scan can accurately demonstrate phase of inflammation in healing bones and progress of infection in osteomyelitic bones.

The infection can extend through the soft tissue and vascular planes into adjacent structures. Without intervention, the disease is usually fatal. Even after the recommended therapy, mortality and morbidity related to its complications are common [1]. In our patients, cases 3, 4, and 7 had giddiness which could be due to labyrinthitis. The labyrinthitis is considered to have been caused by leakage of toxic substances indirectly to the labyrinth as there was no microscopic evidence of semicircular canal fistula per-operatively. Bone erosion was noted in all the cases. Two causes can be postulated for bone erosion. One is erosion by the toxic products released by the fungi. The second possibility is by the cholesteatoma itself [2]. The conservative treatment with systemic and topical antibiotics might have facilitated fungal proliferation.

The treatment of fungal otomastoiditis consists of three parts: control of underlying immunocompromise if present, surgical debridement of necrotic tissues; and antifungal chemotherapy. In our series all were immunocompetent, surgical debridement was done for cases 2–7. Canal wall down Tympanoplasty for all cases except case 6 where a cortical mastoidectomy was done. In immunocompetent hosts with CSOM, *Aspergillus* has been cultured from the middle ear and mastoid granulation but has never been found to invade underlying bone [9]. In all the cases reported bone erosion surrounding the granulation was present.

Conclusion

Fungal tympanomastoiditis, although rare, has to be considered in differential diagnosis of patients with prolonged otorrhoea, otalgia (an uncommon symptom of CSOM) and rapidly progressing audio vestibular symptoms and signs, in spite of an adequate antibacterial therapy. Prolonged use of topical antibiotic -steroid medication might have hampered the innate immunity and facilitated fungal proliferation. Individual cases of fungal otomastoiditis have been reported previously. We report here a case series of seven, all in immunocompetent adults.

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