ISSN: 0975-3583,0976-2833 VOL13,ISSUE07,2022

A RARE CASE REPORT OF ASPERGILLOUS OTOMASTODITIS IN AN IMMUNOCOMPETENT PATIENT WITH REVIEW OF LITERATURE

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Abstract :

Aspergillus otomastoiditis are extremely rare, even in immunocompromised patients. We report a case of a 32-year-old immunocompetent female, who underwent a tymanoplasty because of foul smelling purulent discharge since childhood with congenital cholesteatoma and attic type of perforation in the tympanic membrane. The facial nerve was not involved. Histopathology yielded cholesteatoma sac with *Aspergillus colonies*. This patient's age seems to be younger as per other cases reported. Aspergillus otomastoiditis involving the external auditory canal is again a rarity. When confronted with otitis/ear complaints with an unexpected clinical course a high index of suspicion is required to facilitate early diagnosis and appropriate therapy of a possible lethal Aspergillus infection, even in immunocompetent patients.

Key word : *Aspergillus,* congenital cholesteatoma, otomastoiditis , external auditory canal

Introduction :

Sade [1] in 1993 defined cholesteatoma as “the presence of squamous epithelium in the tympanic cavity producing macroscopic amounts of keratin inadequately cleared”. This circumstance influences youngsters extra aggressively than adults. It is of two types, congenital and acquired cholesteatoma. Clinical features include a release of foul - smelling discharge from the infected ear, loss of hearing, and pain in the infected ear [2]. The disease advances slowly, and the symptoms are not always obvious. When the disease progresses and increasing bone lysis occurs, the patient develops symptoms. Cholesteatoma can occur in the external auditory canal, tympanum-mastoid segment and extending into the facial recess. Cholesteatoma may affect adjoining structures (lateral sinus, facial nerve, posterior cranial fossa). Therefore, a CT scan is recommended for all patients. Diagnosis is made through physical examination and radiological investigation. The various causes of cholesteatoma are congenital, trauma and infection in the upper respiratory tract. The aetiology of cholesteatoma isn't understood, however numerous

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research has found that more than a few things cooperate synergistically for the purpose forming of this nonneoplastic keratinizing lesion. These elements encompass continual microbial contamination ensuing in persistent inflammation, consecutive invasion through cells of the immune system, Eustachian tube dysfunction, aggregation of cell debris, improved viscosity of centre ear effusions, increase of blood vessels, auditory ossicle resorption, and epithelial hyperplasia [3]. Most of the time it is due to bacterial infection. Very few case reports are available on cholesteatoma associated with fungal infection. Here we report a case of 32 years female with a disease of congenital cholesteatoma involving the external auditory canal, middle ear and mastoid segment associated with Aspergillus infection.

Case report :

A 32-year-old Hindu female reported to us in the ENT outpatient department of our tertiary care teaching hospital with the chief complaints of foul- smelling purulent discharge from the left ear and pain in the ear for the last 2months. She had an intermittent history of ear discharge since childhood. The right ear had no complaints. Other constitutional symptoms were absent. The patient had no comorbidities such as diabetes mellitus, hypertension or any other chronic systemic illness. General physical examination shows normal vitals. Examination of the left ear revealed chronic suppurative otitis media with attic perforation in the tympanic membrane with characteristic cholesteatoma flakes involving the auditory canal, middle ear and mastoid segment. The facial nerve assessment was done and seems not to be involved. Audiometry was done and showed moderately severe mixed hearing loss in left ear with normal hearing in right ear. A computed tomography scan was done, which revealed left side mastoid air cells collage to form a larger cavity of size 76 x 17 x 16 mm with a collection. The collection was communicating with the middle ear eroding the anterior and superior wall. The surrounding area of mastoid bone showed sclerosis. A diagnosis of left chronic suppurative otitis media with attic perforation and attico-antral (unsafe) type ear disease was made, and the patient was treated with modified radical mastoidectomy with type IV tympanoplasty. The excised cholesteatoma sac was sent for histopathological examination and it revealed fully differentiated stratified squamous epithelium, dead keratin squames and fungal balls. Periodic-acid-Schiff (PAS-stain) stain shows tightly packed noninvasive laminated fungal hyphae with acute angle branching, suggesting Aspergillus infection. Blood culture of the patient was done and no growth was seen. The patient was discharged and continued to receive oral Itraconazole. Her condition remains healthy, four months after the tympanoplasty.

Discussion :

Fungal infections of the head and neck region are uncommon. Fungal infection is often seen in immunosuppressed individuals and organ transplant cases. Infections of the external auditory canal and paranasal sinuses in an immunocompetent patient is a rarity and usually associated with host immunodeficiency. Our's case is 32 years female who presented with foul - smelling discharge since childhood. The lady's blood culture showed no growth. A chest X-ray and an abdominal CT scan were performed to rule out any systemic lesion, both of which were negative. The lady was not immunosuppressed. The lady was not on an immunosuppressive drug or

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steroid. So this is a case of Chronic suppurative otitis media with congenital cholesteatoma in an immunocompetent individual.

Almost all patients with documented invasive external otitis resulting from Aspergillus reported in the literature were noted to have facial nerve involvement and most of the patients are old. In contrast, our's patient is a young 32 years lady . One more interesting thing is despite the involvement of the external auditory canal, middle ear and mastoid by Aspergillus and also an attic type of perforation in the tympanic membrane, there was no facial involvement.

Bacterial otitis and fungal otitis were similar in terms of symptoms, clinical signs, laboratory investigation and radiological features. As the diagnosis of fungal appendicitis cannot be made by clinical and radiological manifestations alone, a high index of suspicion is therefore very essential, especially in immune-depleted cases. Patients with invasive aspergillosis can have a similar clinical presentation to that infection caused by Pseudomonas aeruginosa. But the demonstration of fungal colonies will lead to correct diagnosis and proper treatment. Different case reports are available showing mucormycosis [4], aspergillosis [5], and candidiasis as the aetiology of fungal otitis. The diagnosis is made by the demonstration of fungal elements and tissue reaction in histopathology section. The patient was successfully treated with type IV tympanoplasty in combination with antifungal therapy. She is being followed for eight weeks, and she is doing well.

Conclusion :

Otomastoditis related to Aspergillosis involving the external auditory canal is very rare in occurrence and is seen in both immunosuppressed and immunocompetent patients. Due to the invasive nature of this life-threatening disease, prompt diagnosis and aggressive management including surgical debridement and antifungal therapy are necessary. When confronted with otitis/ear complaints with an unexpected clinical course a high index of suspicion is required to facilitate early diagnosis and appropriate therapy of a possible lethal Aspergillus infection, even in immunocompetent patients.

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ISSN: 0975-3583,0976-2833 VOL13,ISSUE07,2022

Pictures

Fig 1: Photomicrograph : Audiometry of the patient showing hearing loss in the left ear

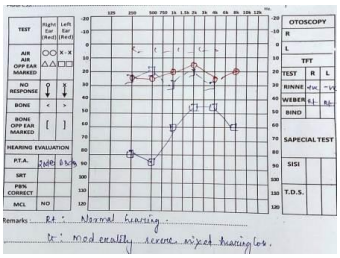

Fig 2: Photomicrograph : CT scan of mastoid showing left side mastoid air cells collage to form a larger cavity of size 76 x 17 x 16 mm with a collection

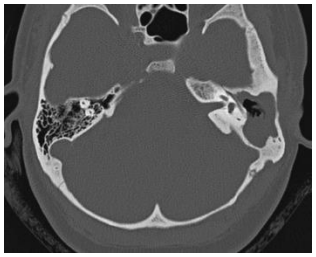

Fig 3: Photomicrograph : H & E pictures showing squamous epithelium, dead keratin squames and fungal balls in 10x

Fig 4: Photomicrograph : PAS stain showing Aspergillous in 40x