Case Report

Aspergillus invasive otitis externa as a complication of herpes zoster oticus

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\textbf{A B S T R A C T}

Aspergillus invasive otitis externa (IOE) is rare and often occurs in the immunocompromised hosts. An immunocompetent case of Aspergillus IOE complicated by herpes zoster oticus is described in this report. The patient presented with refractory otorrhea and otalgia that continued for a long time after resolution of the herpes zoster oticus. She was successfully treated for 8 weeks with oral voriconazole. To our knowledge, this is the first description of an immunocompetent case of Aspergillus IOE complicated by herpes zoster oticus.

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1. Introduction

Herpes zoster has been described in all age groups and the lifetime risk is estimated to be 10%–20%. The incidence of herpes zoster is about 150–300 cases per 100,000, with the incidence dramatically increased in patients older than 60 years [1]. Herpes zoster oticus is a type of herpes zoster that involves external ear. In addition to causing skin lesions, it may affect the geniculate ganglion, causing facial palsy, tinnitus, hearing loss, nausea, vomiting, vertigo, and nystagmus [2,3].

Although herpes zoster disrupts the integrity of the skin and mucosa, secondary infection is rare. When this happens, the responsible organism is often \textit{Staphylococcus aureus}, \textit{β}-hemolytic \textit{Streptococcus}, or \textit{Hemophilus influenzae} [4–6]. Aspergillus invasive otitis externa (IOE) complicated by herpes zoster oticus has not been reported previously. Here, we report an immunocompetent case of Aspergillus IOE complicated by herpes zoster oticus, who presented with prolonged otalgia and otorrhea.

2. Case report

The patient was a 50-year-old woman and a tea farmer who ran a tea market. She had been healthy and never taken any medicine until 12 months ago. She presented with severe left ear pain, vertigo, and headache. She went to her local clinic, where a physical examination showed erythematous swelling and vesicles affecting her left ear canal and pinna that were compatible with herpes zoster oticus. She was transferred and admitted to our hospital, where intravenous acyclovir 500 mg every 8 hours and clindamycin 600 mg every 8 hours were administrated. Her vesicles crusted and the swelling of ear canal subsided gradually. After a 13-day hospitalization, she was discharged. However, she was admitted 1 week later because of severe vertigo. Physical examination showed that the shallow wounds in her left ear canal had reached the healing stage. Brain magnetic resonance image revealed no organic lesions. She received symptomatic treatment and hydrocortisone 100 mg every 8 hours for 2 days. The vertigo became milder and she was discharged. Because of itching sensations within her left ear canal, she often used Q-tips to clean her ear.

Two months later, she began to suffer from left otalgia, otorrhea, and hearing impairment. A physical examination revealed an ulcer and yellowish discharge on the posterior area of her left external ear canal. The laboratory data did not show leukocytosis or elevated C-reactive protein. Intravenous cefazolin 1,000 mg every 8 hours and gentamicin 80 mg every 12 hours were administrated with curettage of the local necrotizing tissues. A pus culture yielded coagulase-negative \textit{Staphylococci} and mold-like pathogens. Her symptoms became milder and she was discharged. One month later, the otorrhea and otalgia flared up again. A physical examination still showed the presence of the same ulcerative wound with yellowish discharge in her left ear canal. Intravenous amoxicillin/clavulanic acid 1,200 mg every 8 hours was administrated with curettage of the local necrotizing tissues. A wound cultured yielded a mold-like pathogen. The otorrhea became less and she was discharged.
Four months later, the same symptoms flared up with the same physical examinations with normal laboratory data. Intravenous ciprofloxacin 400 mg every 12 hours and gentamicin 80 mg every 12 hours were administrated with curettage of the local necrotizing tissues. Because of lessening of the otorrhea, she was discharged. However, the same symptoms flared up again 3 months later with the same physical examination. Intravenous ceftazidime 2 g every 12 hours and oral doxycycline 100 mg every 12 hours were administrated with curettage of local necrotizing tissues. Her otorrhea became less and she was discharged.

One month later, the same symptoms recurred and she was admitted. Physical examination showed ulceration and erythematous swelling with yellowish discharge on the posterior area of her left external ear canal. Her blood cell count, erythrocyte sediment rate, C-reactive protein, and serum electrolyte were all within normal limits. Serum autoimmune profile, such as antinuclear antibody, rheumatoid factor, C3, C4, antineutrophil cytoplasmic antibodies, immunoglobulin G (IgG), IgA, and IgM was normal. A chest film showed no specific finding. Computed tomography showed increased soft tissue density around the left external auditory canal close to the tympanic membrane (Fig. 1). A pus culture yielded mold-like pathogen, which was identified as Aspergillus nigra. Culture of the same sample for Mycobacterium yielded nothing. Intravenous ceftazidime 2 g every 8 hours and vancomycin 1 g every 12 hours were used with tropical ofloxacin for 4 weeks. However, her otorrhea and otalgia persisted. The antibiotic treatment was shifted to oral voriconazole 200 mg every 12 hours and intravenous clindamycin 600 mg every 8 hours. Her otorrhea and otalgia subsided gradually. Physical examination showed an improvement of the swelling and ulceration of left external canal. She was discharged and maintained on oral voriconazole 200 mg every 12 hours and amoxicillin 500 mg every 8 hours for 8 weeks. On follow up at 8 months, the patient was free of symptoms.

3. Discussion

IOE is an ear infection spreading from the external auditory canal to adjacent anatomical structures, including soft tissues, cartilage, and bone [7]. Aspergillus is a rare cause of IOE [8]. When Aspergillus IOE occurs, this pathogen slowly invades adjacent soft tissues and the mastoid air cells. Most cases with Aspergillus IOE involve persons who are immunocompromised, usually those who have long-lasting neutropenia, who are on long-term steroid therapy, or who have uncontrolled diabetes mellitus [9,10]. In immunocompetent patients, diagnosis is often delayed because Aspergillus is very rare cause of IOE, compared with, for example, P aeruginosa [8,11]. In our case, the pus cultured yielded mold-like pathogen at first but it was considered to be a colonized microorganism because her symptoms responded to treatment that did not include an antifungal agents. In addition, Aspergillus IOE is very rare in immunocompetent patients. Because of recurrent courses of her disease, Aspergillus was finally considered to be the pathogen and the correct treatments could be administrated. Tracing back her whole course, we found there had been no systemic inflammatory response syndromes, such as leukocytosis, fever, tachycardia, or elevated C-reactive proteins, in this immunocompetent patient with Aspergillus IOE. Local curettage of necrotizing tissues seems to be able to improve otorrhea transiently in patients with Aspergillus IOE but is impossible to eradicate pathogens completely. Even when the patient is immunocompetent, Aspergillus still should be considered as a possible pathogen of IOE, especially when the clinical course is refractory or recurrent under antibiotics treatment.

Herpes zoster oticus is characterized by the vesicles on the antihelix and external canal after onset of the preherpetic pain. Resolution of the inflammation and lesions occurs in 10–14 days. However, the patient may experience itching during this period. Patients are advised to avoid scratching the ear to avoid spreading the lesions or having the wound developing a secondary infection [12]. Aspergillus is commonly found in soil, dust, plants, and decaying vegetable matter. Our patient was a tea farmer and therefore it is quite possible that Aspergillus might have easily colonized her ear via her hands during her work. The Aspergillus may be inoculated into the unhealed wound in the external ear canal through scratching the ear with her hands. In addition, transient treatment with hydrocortisone for her vertigo during the secondary admission is a possible precipitating factor that might have assisted the colonizing Aspergillus to become invasive.

The treatment of Aspergillus IOE classically includes extensive surgical debridement and intensive long-term antifungal therapy, including amphotericin B and/or itraconazole. In a previous report, 24 cases with Aspergillus IOE were reviewed [13]. The most commonly used antifungal agent was amphotericin B. This drug is effective when treating of Aspergillus IOE, but its substantial toxicity profile must be taken into account. The median total dose of amphotericin B administered was 2 g (range, 0.8–2.5 g) and the patients needed prolonged hospitalization. Two patients in that report were successfully treated with long-term voriconazole therapy, like our patient. Voriconazole is currently considered the first-line therapeutic option for invasive aspergillosis, based on its high intrinsic anti-Aspergillus activity and its superiority when compared with intravenous amphotericin B in large randomized trials [14,15]. In addition, this broad-spectrum azole distributes itself throughout the body, including soft tissue and bone, where good diffusion has been recently documented [16]. Because of the drug having more than 90% oral bioavailability, patients treated with voriconazole do not need prolonged hospitalization [17]. Thus, voriconazole may be considered an attractive first-line therapeutic option for Aspergillus IOE.

In conclusion, we presented an immunocompetent case who suffered from Aspergillus IOE complicated by herpes zoster oticus. We successfully treated the patient using voriconazole without surgical debridement. Aspergillus should be considered as a pathogen for refractory IOE, even in an immunocompetent patient.

Fig. 1. The computed tomography discloses increased soft tissue density (arrow) around left external auditory canal close to the tympanic membrane.
Voriconazole is an attractive and effective first-line therapeutic option for Aspergillus IOE.

References