Case Report

A Concomitant Allergic Fungal Otomastoiditis with an Allergic Fungal Rhinosinusitis

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ABSTRACT

Background: Allergic fungal otomastoiditis is a rare ear condition characterized by the presence of a specific middle ear mucus usually found in allergic bronchopulmonary aspergillosis. Methods: We report a case of concomitant allergic fungal otomastoiditis and rhinosinusitis in an immunocompetent 27-year-old man complaining of right otorrhea and nasal discharge for 6 months resistant to any medical treatment. Examination revealed a sticky mucus arising from a polypoid middle ear associated with a right middle meatus purulent discharge. Surgical treatment revealed no cholesteatoma but thick yellowish mucus and a fungal ball, as targeted by imaging, both of which containing Aspergillus species. Result: The patient received short postoperative systemic corticotherapy and showed no recurrence 6 months after. Discussion: Allergic fungal otomastoiditis should be considered in the face of a sticky mucus arising from perforated chronic otitis, especially when associated with allergic fungal rhinosinusitis. Complete surgical debridement is essential.

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1. Introduction

Allergic fungal rhinosinusitis (AFRS), first described in 1976 by Safirstein et al. [1, 2], is a distinct phenotype of chronic rhinosinusitis with nasal polyps. It is suggested by the presence of a specific mucus in one or more sinuses impaired with mucociliary clearance, similar to that found in the bronchi of patients with allergic bronchopulmonary aspergillosis (ABPA). The presence of that specific mucus outside the respiratory tract was first reported in 2013 when Chen and Chiang [3] described the first case of Allergic Fungal Otomastoiditis (AFO). Here, we present the fourth case [4, 5], of AFO in a patient with AFRS without any history of middle ear disease.

2. Case Report

We report the case of an immunocompetent 27-year-old non-smoker Caucasian man, who was complaining for 1 year about a right nasal obstruction and nasal discharge preceding the onset of a right isolated otorrhea six months ago. He had no known allergies, neither medical nor surgical history other than bilateral turbinoplasty 6 months ago. Before patient consultation, two systemic and local antibiotics short courses, combined with anti-inflammatory eardrops, where attempted with a transient efficiency. The right otoscopy revealed a total eardrum perforation with a thick otorrhea and the left one, an inflatable retraction pocket which deeper part was being seen. No facial or vestibular impairment was noticed. The nasal endoscopic examination found thick nasal discharge and polyps arising from the right middle meatus. Pure-tone audiometry showed right mild-to-moderate conductive hearing loss. The high-resolution temporal bone computed tomography (HTCT) and magnetic resonance imaging (MRI), reported respectively in (Figures 1 & 2), suggested a right cholesteatoma. The chest radiography was normal. Sinus computed tomography (Figure 1) suggested a right maxillary sinus fungal ball with many sinuses’ soft tissue opacifications, especially in the left sphenoid sinus.

The patient underwent a conservative wall up technique mastoidectomy revealing no clinical evidence of cholesteatoma, but a huge polypoid promontory reaction stuck in a thick peanut butter-like mucus fulfilling all middle ear cavities. Multiple samplings were performed. Simultaneously, a right maxillary sinus antrotomy, bulla opening and a left sphenoidotomy were performed, allowing suction and sampling of a thick allergic-type mucus in every sinuses and extraction of a maxillary
sinus fungal ball. The patient received 10 days of oral amoxicillin with clavulanic acid and voriconazole. Bacterial and mycological external ear canal and middle ear swab cultures revealed respectively, a wild-type Propionibacterium acnes and Aspergillus flavus (>10^6 CFU/ml). Ear histologic report found epidermal islets sustained by a thin inflammatory fibrous layer infiltrated with numerous plasmocytes, lymphoplasm cells and eosinophils. The fungal ball and thick sinus allergic-type mucus culture identified 2 Aspergillus species, respectively A. fumigatus and A. flavus. No fungal tissues invasion was reported. Total IgE, A. flavus and A. fumigatus IgE specific levels were elevated, and skin prick tests were negative except for Aspergillus mix species.

We observed right otorrhea recurrence 15 days after treatment. He was re-operated to perform a type III tympanoplasty. Pro tympanum and eustachian tube were washed under pressure, allowing the removal of a eustachian tube shaped mucous plug (Figure 3). Swab mycological cultures were still positive to Aspergillus flavus, unlike bacteriological ones, which were all negative. The second histological report found neither epidermal cells nor fungal invasion. The patient received a 15 days systemic steroids course (1mg/kg of prednisolone) combined with antibiotic prophylaxis and was normally followed up with no clinical sign of nasal or ear recurrence 6 months after. We noticed audiometric normalization. Written patient consent was obtained to report his case.

3. Discussion

AFO is an uncommon ear condition [4, 5], whose pathogenesis is quite similar with AFRS and ABPA as it could be type 1 and type 3.
hypoallergenic to multiple fungi [2] with elevated serum IgE and IgG against specific fungal antigens. As skin tests, biological and mycological results suggested in that case, Aspergillus species were involved as usually reported in AFRS [1, 2], but also in AFO [3, 5], when fungal elements can be identified. Firstly described by Chen et al. [3] in 2013, this diagnosis is supported by seeing a sticky and sandy peanut-butter-like mucus coming out from a tympanic perforation which may have been present for several years with multiple failed surgical closure attempts [3, 5]. HTCT (Figure 1) and specifically MRI (Figure 2) suggested cholesteatomatous chronic otitis, except dark heterogeneous T2-weighted images, as it can be seen in AFRS because of ferromagnetic elements inside the fungal mucus [1]. The first histological report was consistent with an early stage epidermal invasion, but underlying presence of hypersensitivity cells suggested the intertwining of both pathologies in the form of squamous cell epithelial metaplasia.

Commensal fungi in the external auditory canal could have run through the perforated eardrum into the middle ear and led to the hypersensitivity reaction. As targeted by preoperative MRI, we removed an Aspergillus colonized eustachian tube shape mucous plug in the protympanum (Figure 3). This plug was probably the AFO thick mucus accretion result through a dysfunctional eustachian tube, and its non-removal in the first surgery could have been a risk factor for recurrence [4] as it was noticed in a recurrent AFO.

In our case, AFRS was confirmed as all 5 Bent and Kuhn’s criteria were present. Concurrent AFRS has previously been described [4] and could be an alternative cause of its middle ear spread, not only because of reported symptoms chronology but also the fungus strains similarity found in both places.

As there is a lack of evidence concerning AFRS medical treatment, extensive surgical debridement is still the gold standard and, as previously reported [3, 5], was performed on our patient on his sinuses and ear to remove any fungal element, limit recurrence risk and improve middle ear and sinus ventilation. As AFRS [1], patients who received a combination of oral and topical corticosteroids postoperatively for AFO [3, 5], had better clinical outcomes and no disease recurrence compared with those undergoing surgery alone [4].

4. Conclusion
Although it is uncommon, otolaryngologists have to consider allergic fungal otomastoiditis when examining immunocompetent patients with a history of chronic rhinosinusitis with new-onset otologic symptoms, especially those with a sticky peanut-butter-like otorrhea. In case of surgical exploration, the protympanum has to be specifically checked as a mucous plug could block the eustachian tube clearance and led to recurrence.

References


