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Allergic Fungal Otomastoiditis in a Patient without Allergic Fungal Rhinosinusitis: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Patient: Female, 27
Final Diagnosis: Allergic fungal otomastoiditis – AFOM
Symptoms: Left-sided otorrhea and hearing impairment for 2 years
Medication: Systemic steroid course
Clinical Procedure: Aural toilets and systemic steroid over 2 weeks
Specialty: Otolaryngology

Objective: Rare disease
Background: Allergic fungal rhinosinusitis is an inflammatory condition involving the paranasal sinuses and linings of the nasal passages that lasts 12 weeks or longer. In 2013 Chen et al. reported the first case of middle ear allergic otomastoiditis in a patient who tested negative for allergic sinus-nasal disease. To the best of our knowledge, this is the second such case report to be published.
Case Report: Our patient was a 27-year-old immune-competent woman presenting with chronic left-side otorrhea and hearing loss for 2 years. An audiogram showed low- and middle-frequency, mild-to-moderate left-sided conductive hearing loss. A coronal view computed tomography (CT) image of the temporal bone showed coalescent air cells in the left mastoid with opacification of the left middle ear, but no evidence of bony erosion. Diagnosis confirmed by swab culture taken at a clinic showed *Aspergillus flavus*. The main treatment in such cases is usually a combination of surgical and medical therapy.
Conclusions: We present the second case report of allergic fungal otomastoiditis, showing an allergic reaction to fungi in the middle ear and formation of mucin in a symptomatic patient. Otolaryngologists need to be aware of the presence of this sensitivity, both in the clinic and the operating theater.

MeSH Keywords: Antibodies, Fungal • Ear, Middle • Mastoiditis • Mucins

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Background

Unfavorably susceptible mucin was first portrayed by Katzenstein et al. in 1983 as white-tan mucoid material in the paranasal sinuses of a group of patients [1]. The mucin is histologically undefined, starting with mucoid impaction of the bronchi for patients with hypersensitive bronchopulmonary aspergillosis (ABPA). Such patients may have clumps of necrotic eosinophils and incidental Charcot-Leyden crystals, as well as insufficient contagious hyphae, alongside a foundation of amorphous mucin.

This illness displays a clinical and pathological picture similar to that of ABPA, but mostly involves the paranasal sinuses as opposed to the lower-level airways, with allergic *Aspergillus* sinusitis and allergic fungal rhinosinusitis (AFRS) [1–3]

The presence of unfavorably susceptible mucin outside the respiratory tract was first announced in 2013, when Chen and Chiang reported a case of allergic fungal otomastoiditis. Here, to give more prominent attention to this condition and its range of symptoms and severity, we present the second reported case of allergic fungal otomastoiditis, in a patient with no nasal symptoms or nasal procedure.

Case Report

A 27-year-old non-smoker Saudi woman presented at our clinic with constant left-sided otorrhea and hearing impairment for 2 years. She was immunocompetent, without any systemic disease. On otoscopic exam, she was noted to have left marginal eardrum perforation, with thick, purulent otorrhea. She did not have any facial shortcoming or vertigo. Pure tone audiogram

Figure 1. (A, B) Coronal view of different level of high-resolution computed tomography of the temporal bone revealed coalescent left mastoid air cells and opacification of left middle ear, with no evidence of bony erosion.

Figure 2. (A) Keratinized squamous epithelium. (B) Giant cells granuloma around the keratin.

Figure 3. Coronal view of different level of high-resolution computed tomography of the temporal bone revealed partial left mastoid bone resection with a large soft-tissue lesion eroding the bone around the lateral wall of the sigmoid sinus, associated with demineralization of the facial recess and jugular bulb (red arrow for eroding the bone around the lateral wall of sigmoid sinus).

Figure 4. (A, B) Coronal view of different level of magnetic resonance imaging of the temporal bone revealed a well-defined lesion occupying the left mastoid; high signal intensity air cells on T1 could represent proteinaceous material with diffusion restriction.

testing showing mild-to-moderate left-sided conductive hearing loss. A coronal view CT of the temporal bone showed coalescent air cells in the left mastoid with opacification of the left middle ear, with no evidence of bony erosion (Figure 1A, 1B).

The patient underwent cortical mastoidectomy and tympanoplasty type III. The pathology report revealed transitional epithelium-lined tissue with aggregates of lymphoplasmacytic cells and pools of keratinous material (Figure 2A, 2B).

Left-sided otorrhea and hearing impairment recurred once more, which prompted the patient to return our clinic in July 2017. On otoscopic exam, she was noted to have left subtotal eardrum perforation with some sticky mucopus in the external auditory canal. A coronal view CT of the temporal bone showed partial left mastoid bone resection with a large soft-tissue lesion eroding the bone, associated with demineralization of the facial recess and jugular bulb. The ossicles are not visualized on the left side and the internal ear structures were unremarkable (Figure 3).

Magnetic resonance imaging (MRI) of the temporal bone revealed a well-defined lesion occupying the left mastoid, with air cells showing high signal intensity in T1, which could indicate proteinaceous material with diffusion restriction with impression of left-side cholesteatoma after mastoidectomy and findings suggestive of residual/recurrence (Figure 4A, 4B).

The patient underwent revised left canal wall mastoidectomy tympanoplasty type IV for suspected recurrent cholesteatoma. Strangely, in addition to pearl-like cholesteatoma, there was also yellowish material coming from the mastoid antrum, with the appearance of cholesterol granuloma. The pathology report in July 2017 showed transitional epithelium-lined issue with aggregates of lymphoplasmic cystic cells and pools of keratinous material. There was eosinophilic mucin, and PAC staining for fungi was negative. A surgical specimen was not sent for bacterial and mycobacterial culture.

Two months later, the left otorrhea and hearing impairment recurred again, which prompted the patient to return to our clinic. On otoscopic exam, she was noted to have left central eardrum perforation with some sticky mucopus in the external auditory canal. A swab culture was taken in the clinic, showing *Aspergillus flavus* without clinical evidence, and swabs sent from the clinic did not show any bacterial growth. We did not evaluate the atopic status of our patient, as diagnosis depends on the clinical picture and the growth of a fungal agent in the swab culture.

The patient was normally followed up at our outpatient clinic. She received topical steroid (dexamethasone), with minimal response, then received systemic steroid (prednisolone 1 mg/kg, tapering over 2 weeks), to which she responded within 3 days of initiation of medication. The middle ear has since remained free of unfavorably susceptible mucin to date.

Discussion

Unfavorably susceptible fungal otomastoiditis is an extremely uncommon condition, with only 1 case previously reported, in 2013 by Chen and Chiang [4], which involved mucin in the middle ear and mastoid cavity of a patient with a 10-year history of hearing debilitation and otorrhea unmanageable by different mediations, for which they coined the term “allergic fungal otomastoiditis”. This condition is characterized by thick, dull, nutty-spread-like bodily fluid, like the inspissated bodily fluid found in the bronchi of patients with ABPA, either grossly or microscopically [1]. Those pathogens, also comparable to ABPA, are postulated to be type 1 and type 3 hypersensitivity to various fungi, with raised serum IgE and IgG levels against particular contagious antigens [1,5,6] Theoretically, this allergic response can occur any place inside the respiratory mucosa,

and the thick, dull, nutty-spread-like bodily fluid transformed may accumulate in the spaces of the respiratory tract, such that the paranasal sinuses and lower airways, bringing about the indications of AFRS and ABPA.

Curiously, our patient had allergic mucin in her left middle ear rather than the paranasal sinuses or bronchi. Our patient's introduction contrasted from that of the patients depicted by Chen and Chiang (2013) and Bayer et al. (2014) in that she did not have any nasal symptoms or nasal procedures.

ABPA was excluded in our patient because she had no manifestations or indications of lower airway obstruction and she had a typical chest radiography report. Therefore, we suspected that fungus growing in the external auditory canal might have gone through the punctured ear drum into the middle ear.

Hall and Farrior classified *Aspergillus* infection in temporal bone [7,8] as 3 types:

- (i) Noninvasive-localized and does not invade tissue, responding to conservative removal;
- (ii) Invasive-bony invasion with granulomatous response and fibrosis, occurring in immunocompetent patients;
- (iii) Fulminant-tissue and angioinvasion with no granulomatous response, occurring in immunocompromised patients.

These signs might be useful in distinguishing AFOM from cholesteatoma, which normally does not have heterogeneous flag power and dissolves as opposed to extending. The nearness of intermittent thick otorrhea may likewise suggest AFOM rather than cholesteatoma.

In such cases, surgery is necessary. Careful extirpation of the ailment is viewed as the essential treatment for AFRS, with the objective of evacuating the activating antigen and enhancing ventilation and post-agent access for debridement. In the ear, this may require a canal wall tympanomastoidectomy. As is frequently the situation in AFRS, our patient had recurrence, with focal eardrum perforation and thick otorrhea at about 2 months after the medical procedure.

Topical steroids and intermittent debridements seem to have controlled the sickness throughout the previous couple of months. There is some evidence that IgE treatment might be useful, yet it is unclear whether immunotherapy and anti-fungal agents can effectively treat AFOM [2].

Conclusions

We introduce the second case report of unfavorably susceptible allergic fungal otomastoiditis. Like AFRS and ABPA, hypersensitive responses to a growth in the middle ear could incite

development of unfavorably susceptible mucin, which further aggregates in the middle ear and results in the presented symptoms, although there might be a high rate of intermittent illness in specific cases. Otolaryngologists need to be aware of

the presence of unfavorably susceptible mucin, both in the clinic and in the operating theater. Topical steroid otic drops can be used to control the hazardous otorrhea postoperatively.

References:

1. Katzenstein AL, Sale SR, Greenberger PA: Allergic aspergillus sinusitis: A newly recognized form of sinusitis. *J Allergy Clin Immunol*, 1983; 72: 89–93
2. Bent JP, Kuhn FA: Diagnosis of allergic fungal sinusitis. *Otolaryngol Head Neck Surg*, 1994; 111: 580–88
3. Ryan MW, Marple BF: Allergic fungal rhinosinusitis: Diagnosis and management. *Curr Opin Otolaryngol Head Neck Surg*, 2007; 15: 18–22
4. Chen CM, Chiang CW: Allergic fungal otomastoiditis: A case report. *Laryngoscope*, 2013; 123(4): 1040–42
5. Stewart AE, Hunsaker DH: Fungus-specific IgG and IgE in allergic fungal rhino sinusitis. *Otolaryngol Head Neck Surg*, 2002; 127: 324–32
6. Manning SC, Holman M: Further evidence for allergic pathophysiology in allergic fungal sinusitis. *Laryngoscope*, 1998; 108: 1485–96
7. Amonoo-Kuofi K, Tostevin P, Knight J: Aspergillus mastoiditis in a patient with systemic lupus erythematosus: A case report. *Skull Bas*, 2005; 15(2): 109–12
8. Hall PJ, Farrior JB: Aspergillus mastoiditis. *Otolaryngol Head Neck Surg*, 1993; 108(2): 167–70