

Case Report

Allergic Fungal Otomastoiditis: A Case Report

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Allergic mucin is described as thick, peanut butter-like mucus impacted in the paranasal sinuses of patients with allergic fungal rhinosinusitis. The presence of allergic mucin in the middle ear has never been reported. We encountered a 65-year-old female with allergic mucin found impacted in her left middle ear and mastoid cavity during revised tympanoplasty surgery at our institute. Bilateral endoscopic sinus surgery performed 3 months later showed no evidence of fungal infection or allergic mucin in her paranasal sinuses. We report the case herein and propose the term *allergic fungal otomastoiditis* for this disease entity.

Key Words: Otitis media, otomastoiditis, allergic fungal rhinosinusitis, allergic mucin, fungal mastoiditis.

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INTRODUCTION

Allergic mucin was first described by Katzenstein et al. in 1983 as grossly white-tan mucoid material impacted in the paranasal sinuses of a group of patients.¹ The mucin is histologically indistinguishable from mucoid impaction of the bronchi in patients with allergic bronchopulmonary aspergillosis (ABPA) and is characterized by clumps of necrotic eosinophils, occasional Charcot-Leyden crystals, and scant fungal hyphae within a background of amorphous mucin. This disease entity presents a clinical and pathological picture similar to that of ABPA but involves mainly the paranasal sinuses instead of the lower airways, and the terminology eventually changed from allergic aspergillus sinusitis to allergic fungal rhinosinusitis (AFRS).¹⁻³ Allergic mucin has never been found in the middle ear and mastoid cavity; herein we report the first case of allergic fungal otomastoiditis.

CASE REPORT

A 65-year-old woman had been troubled by left hearing impairment and intermittent otorrhea for 10 years. She is immunocompetent without any systemic disease. Reviewing her history, left ear drum perforation was first noted in 2001, and she underwent a paper patch myringoplasty in December 2002. Due to recur-

rences of the left ear drum perforation and otorrhea, she received tympanoplasty type IV in August 2004 and revised tympanoplasty type IV with canal wall down mastoidectomy and ossiculoplasty at another medical center in February 2009. The pathology report in February 2009 showed fibrous stroma infiltrated by numerous lymphoplasma cells and scattered eosinophils without keratinized squamous epithelium.

Left otorrhea and otalgia relapsed again, which led the patient to our clinic in June 2010. Otoscopic exam showed a bulging left ear drum with some sticky mucopus in the external auditory canal. High-resolution computed tomography of the temporal bone revealed postmastoidectomy change with soft tissue density filling the left middle ear and opened mastoid cavity (Fig. 1). Revised tympanoplasty with mastoidectomy was performed again on July 13, 2010, for suspected recurrent cholesteatoma. Interestingly, besides pearl-like cholesteatoma, there was also much peanut butter-like mucus in the middle ear and mastoid cavity (Fig. 2). The pathology report showed some tissue fragments composed of amorphous mucin infiltrated by eosinophils, compatible with allergic mucin. Further fungal staining (periodic acid-Schiff staining) also revealed typical septate fungal hyphae with acute-angle branching scattered in the mucin, which is pathognomonic for aspergillosis (Fig. 3). Some of the tissue fragments were found to be granulation tissue with laminated keratin material, compatible with cholesteatoma.

In addition, the patient underwent bilateral endoscopic sinus surgery for chronic rhinosinusitis in October 2010. However, no allergic mucin was found in the paranasal sinuses, whether intraoperatively or pathologically.

The patient was regularly followed-up at our outpatient clinic and treated with topical steroid otic drops

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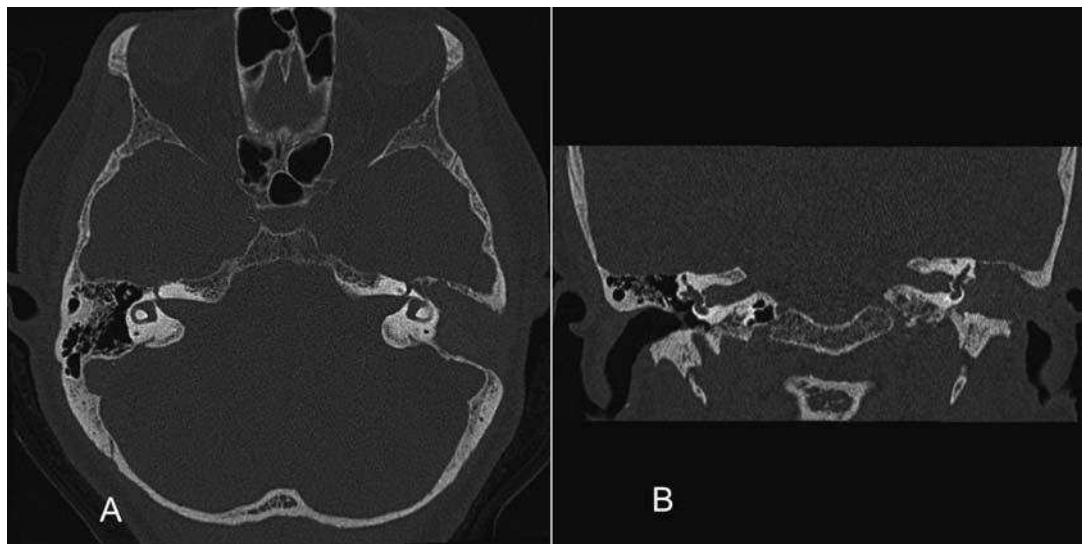


Fig. 1. Axial view (A) and coronal view (B) of high-resolution computed tomography of the temporal bone revealed postmastoidectomy change of the left ear and some soft tissue density filling the mastoid cavity.

(Rinderon eye solution [Shionogi & Co., Ltd., Osaka, Japan], betamethasone disodium phosphate solution, 1 mg/mL) during the following months. The middle ear and opened mastoid cavity remain free of allergic mucin to date.

DISCUSSION

Allergic fungal rhinosinusitis is characterized by allergic mucin impacted in the paranasal sinuses. The thick, dark, peanut butter-like mucus is similar to the inspissated mucus found in the bronchi of patients with ABPA, whether grossly or microscopically.¹ The pathogenesis, also similar to ABPA, is postulated to be type 1 and 3 hypersensitivity to multiple fungi, with elevated serum IgE and IgG against specific fungal antigens in most of these patients.^{1,4-6} Theoretically, this allergic reaction could happen anywhere within the respiratory mucosa, and the peanut butter-like mucus produced could accumulate in the spaces of the respiratory tract, such as the paranasal sinuses and lower airways, resulting in the symptoms of AFRS and ABPA.

Interestingly, this female patient had allergic mucin in her left middle ear instead of the paranasal sinuses or bronchi. The endoscopic sinus surgery performed 3 months later excluded the presence of AFRS. The ABPA could also be excluded, as the patient had no symptoms or signs of lower airway obstruction and had a normal chest radiography report. Therefore, it is assumed that fungi inhabiting the external auditory canal could have run through the perforated ear drum into the middle ear and evoked the hypersensitivity reaction locally. The allergic mucin was thus produced by the respiratory epithelium of the middle ear and resulted in the problematic otorrhea.

Reviewing the pathology report performed in another medical center in February 2009, eosinophils were found to be scattered in the fibrous stroma from the middle ear. The histological finding should have sug-

gested an allergic reaction in the middle ear occurring at least since 2009, although fungal staining had not been performed, and no amorphous mucin or fungal hyphae were mentioned in the report.

Fungal mastoiditis is a rare disease entity and occurs almost exclusively in immunocompromised patients. In immunocompetent patients, the disease has been only sporadically reported.⁷ As far as we know, fungal infection of the middle ear in the form of allergic mucin has never been reported in the literature. Therefore, we present this case and propose the term *allergic fungal otomastoiditis* for this disease entity, analogous to allergic fungal rhinosinusitis and allergic bronchopulmonary aspergillosis.

In the case of AFRS, corticosteroids, both systemic and topical, are the mainstay treatment to prevent the recurrence of polypoid disease and mucus trapping after sinus surgery. Topical and systemic antifungal agents have been tried to decrease the fungal antigenic burden in the sinonasal cavities, but convincing data are still

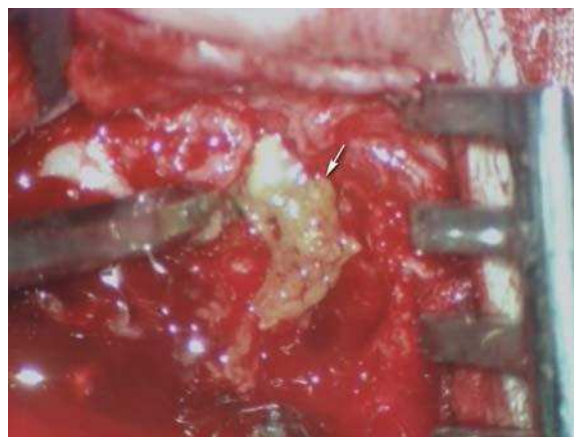


Fig. 2. Intraoperatively, sticky peanut butter-like mucus was found in the mastoid cavity and middle ear (white arrow).

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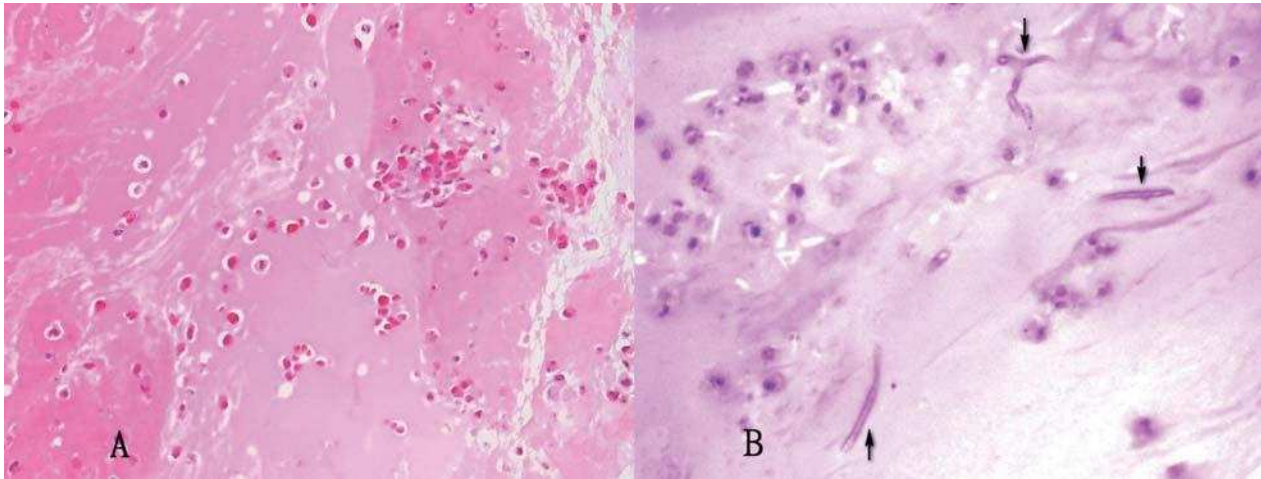


Fig. 3. In the hematoxylin and eosin stain (A), eosinophils can be seen to have infiltrated the amorphous mucin. In the periodic acid-Schiff stain (B), septate and acute angle-branching fungal hyphae are shown (black arrows), compatible with the *Aspergillus* species.

lacking.³ In this case, we prescribed topical steroid otic drops (Rinderon solution) postoperatively to prevent recurrence of allergic mucin production, and over the 20-month follow-up period the middle ear and opened mastoid cavity remained free of allergic mucin.

CONCLUSION

We present the first case report of allergic fungal otomastoiditis. Similar to AFRS and ABPA, allergic reactions to fungus in the middle ear could induce formation of allergic mucin which further accumulates in the middle ear and results in the reported symptoms. Otolaryngologists should be aware of the existence of allergic mucin during practice in the clinic or operating

theater. Topical steroid otic drops can be used for control of the problematic otorrhea postoperatively.

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