

Otopolyposis With Middle Ear Allergic Mucin in a Patient With Allergic Fungal Rhinosinusitis

Annals of Otolaryngology, Rhinology & Laryngology
1–4

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DOI: 10.1177/0003489416660112

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Abstract

Objective: The purpose of this study is to report a case of otopolyposis and middle ear allergic mucin in a patient with allergic fungal rhinosinusitis (AFRS) and no history of middle ear disease and introduce these as possible otologic manifestations of the AFRS.

Methods: A case of a 31-year-old female with the aforementioned findings is reported. A review of the pertinent literature was performed.

Results: We report a case of a 31-year-old female with a history of AFRS but no history of middle ear disease or hearing loss who presented to our institution complaining of aural fullness. Physical exam was significant for middle ear masses of unknown etiology. Surgical exploration revealed the presence of allergic mucin and middle ear polypoid histologically identical to tissue sampled during prior sinonasal surgeries at the same institution. Aspiration of the middle ear space did not resolve the otologic symptoms.

Conclusion: Otopolyposis and middle ear allergic mucin are extremely rare but possible otologic manifestations of AFRS. We encourage otolaryngologists to consider this in the clinical differential diagnosis of patients with a history of AFRS with new onset otologic symptoms.

Keywords

allergic fungal rhinosinusitis, otitis, polypoid, allergic mucin

Introduction

Allergic fungal rhinosinusitis (AFRS), first described in 1983 by Kazanstein et al¹ is a disease characterized by the presence of thick, inspissated, dark-tan mucus, and polyps in the nasal cavity and paranasal sinuses. This same mucus, known as allergic mucin, is found in the bronchi of patients with allergic bronchopulmonary aspergillosis (ABPA) and is histologically characterized by branching fungal hyphae, eosinophils, and Charcot-Leiden crystals composed of eosinophil breakdown products. Although *Aspergillus* was initially thought to be the offending organism of AFRS, it is now known that a variety of dematiaceous fungi that colonize the respiratory tract are also responsible.² Diagnosis is made by meeting all 5 of Bent and Kuhn's criteria for AFRS: history of type I hypersensitivity, presence of nasal polypoid and eosinophilic mucin without invasion, positive fungal stain, and characteristic computed tomography (CT) findings of heterogeneous opacification of the sinuses.³ Presence of allergic mucin outside the respiratory tract was first reported in 2013, when Chen and Chiang⁴ described the first

case of allergic fungal otomastoiditis. Otopolypoid has been reported previously in a patient with Samter's triad and another with chronic eosinophilic otitis media in 2009 and 2015, respectively.^{5,6} Here, we present a case of allergic mucin and polyps found within the middle ear in a patient with AFRS without a history of middle ear disease.

Case Report

A 31-year-old female with a history of asthma but without a history of aspirin intolerance presented to our clinic in

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December 2014 complaining of a sensation of fullness in both ears, more pronounced on the left side. She denied otorrhea, otalgia, vertigo, or hearing loss. She had a long-standing history of AFRS requiring 4 prior endoscopic sinus surgeries over a 13-year span. The most recent of these surgeries was 2 months prior to our otologic evaluation. She had known medical allergies to penicillin and sulfonamide and various environmental allergies previously addressed with immunotherapy. She had no personal or family history of autoimmune or vascular disease.

On otoscopic exam, tan polypoid masses were visible medial to and abutting the tympanic membranes bilaterally. Computed tomography of the temporal bones confirmed bilateral soft tissue densities in both middle ears (Figure 1). These findings were present on imaging in 2011, with an increase in opacification in the interim. Preoperative audiogram was normal (Figure 2A).

The patient was taken to the operative room for surgical exploration of the left middle ear for tissue biopsy to establish diagnosis. The left side was prioritized as it was more symptomatic for the patient. Upon elevating the tympanic membrane, thick, yellow mucoid secretions were found and aspirated from the middle ear space and Eustachian tube. There was a well-organized fibrous and polypoid tissue mass adherent to the medial aspect of the tympanic membrane, filling the entire mesotympanum and encasing the ossicles. Pathologic examination confirmed this tissue to be an inflammatory process consistent with polyposis. Allergic mucin, containing Charcot-Leiden crystals and inflammatory cells including abundant eosinophils and plasma cells, was also identified (Figure 3). Grocott-Gomori's methenamine silver (GMS) fungal staining of the mucin did not reveal any fungal organisms. These histologic findings were identical to the allergic mucin and sinonasal polyps sampled from prior sinonasal surgeries at our institution.

The patient is compliant with scheduled interval follow-up examinations and reports no significant change in symptoms, still experiencing intermittent fullness in the ears, which she notes correlates with her nasal symptoms. Otoscopic exam shows recurrence of the mass just as prior to surgery (Figure 4). Postoperative audiogram is stable compared to preoperative exam (Figure 2B). Given that the source of disease is centered within the sinonasal tract, the patient is presently followed conservatively without intervention on the contralateral ear. The focus of treatment shifts to management and control of the sinonasal disease.

Discussion

Allergic fungal rhinosinusitis is characterized by the presence of allergic mucin and polyps in the nasal cavity and paranasal sinuses. This thick, dark tan-colored mucin is histologically identical to that found in the bronchi of patients with allergic bronchopulmonary aspergillosis. It was previously believed

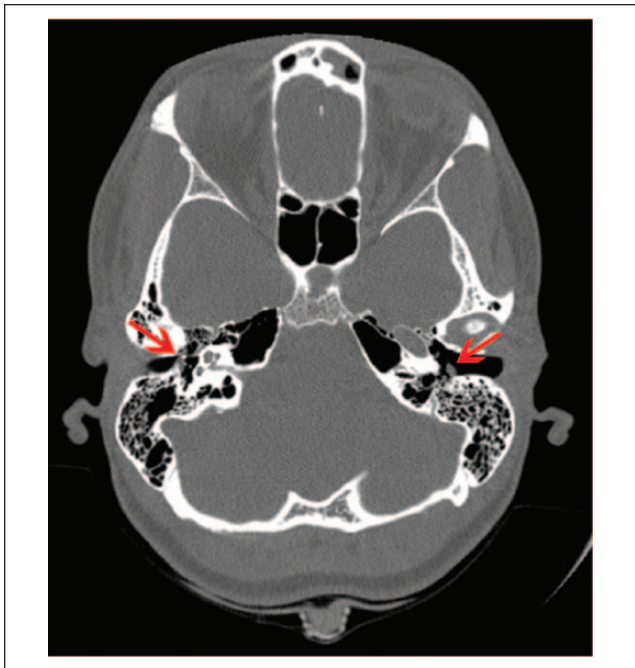


Figure 1. High-resolution computed tomography of the temporal bones revealed soft tissue abutting the tympanic membrane and encasing the ossicles bilaterally.

that this inflammatory response was caused by an IgE and IgG-mediated type 1 and 3 hypersensitivity to fungal antigens—the same pathophysiology as ABPA.^{7,8} However, recent studies have shown that AFRS may be a considerably more complex process, involving a cytokine-mediated local Th2 immunologic response to various fungi and even *Staphylococcus aureus* by the respiratory epithelium.⁹

Otic polyps were reported in 2009 by Brobst et al⁶ in a patient with Samter's triad and significant otologic complaints. Allergic mucin had never been reported outside the respiratory tract until 2013 when Chen and Chiang⁴ reported the presence of mucin in the middle ear and mastoid cavity of a patient with a 10-year history of hearing impairment and otorrhea refractory to multiple interventions, to which they coined the term *allergic fungal otomastoiditis*. Azadarmaki et al⁵ later reported a case of otic polyps present in a patient with eosinophilic otitis media, a form of chronic otitis media characterized by eosinophil-rich mucin in the middle ear in the absence of fungal organisms and refractory to conventional treatments for otitis media.¹⁰ Their patient had a longstanding history of middle ear disease and profound hearing loss, having failed multiple sets of tympanostomy tube placements.

Here we present the first case of otic polyps in a patient with AFRS and no prior history of middle ear disease. Aerated middle ear and mastoid space on serial imaging speaks to the absence of acute or chronic otitis. Unlike previous cases in the literature, our patient's only symptom

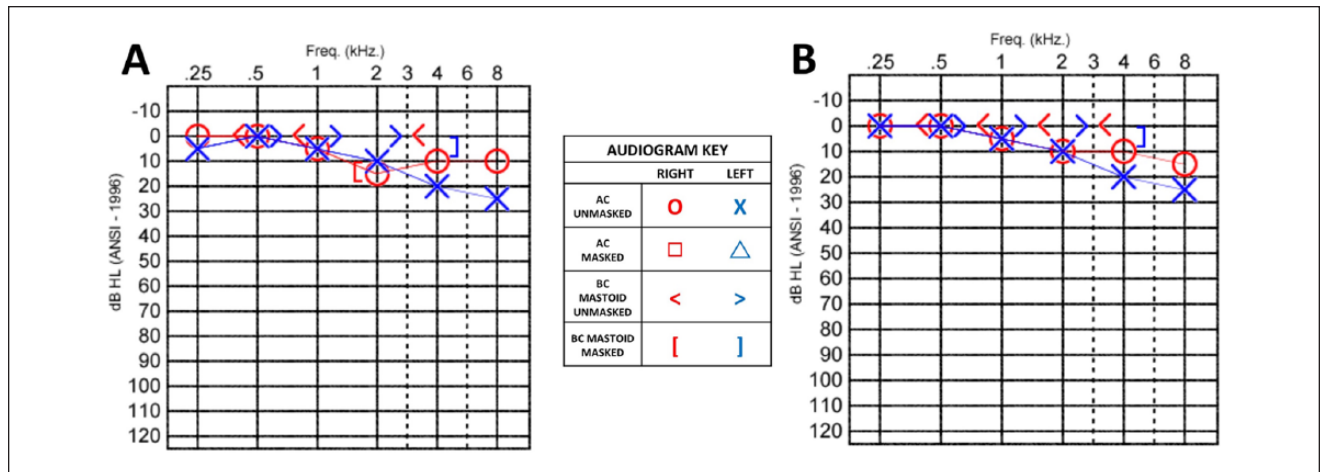


Figure 2. (A)Preoperative audiogram, which remained stable (B) postoperatively.

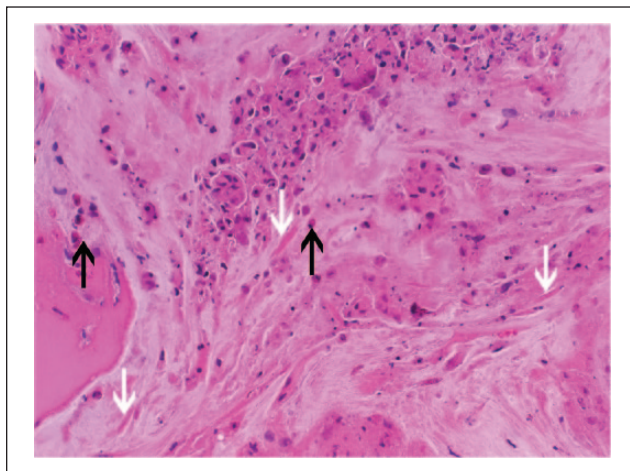


Figure 3. Hematoxylin and eosin (H&E) staining of the inspissated mucoid secretions at 400× magnification reveals eosinophils (black arrows) and Charcot-Leyden crystals (white arrows) admixed with mucin.

was fullness in the ears without concomitant hearing loss. The decision was made to proceed with surgery based on the presence of clinical symptoms and an uncharacterized middle ear mass. The sensation of fullness did not respond to surgical intervention. As mucin was aspirated from the Eustachian tube intraoperatively, we suspect the otic polyps were a direct extension of preexisting sinonasal disease and consequently an incidental finding during the workup of her otologic complaints.

Conclusion

We present a case of otopolyposis and allergic mucin in a patient with a history of sinonasal polypsis. Unlike prior reports, our patient did not have a history of recurrent otitis

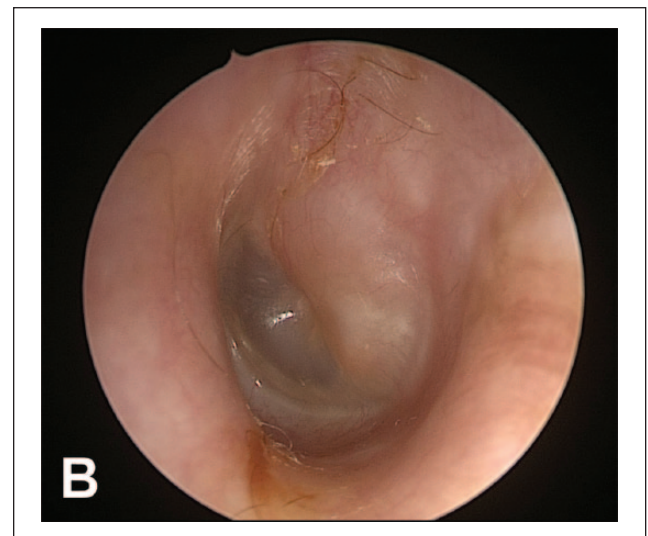


Figure 4. One year postoperatively, “recurrent” mass is seen abutting the posterior aspect of the tympanic membrane just as prior to surgery.

media or hearing loss. The only symptom was aural fullness that did not respond to removal of the otitic polypoid disease. Although an extremely rare disease entity, we encourage otolaryngologists to consider this in the clinical differential diagnosis of patients with a history of allergic fungal rhinosinusitis with new onset otologic symptoms.

Declaration of Conflicting Interests

The author(s) declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: The authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

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