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Case Report

Bilateral Nontuberculous Mycobacterial Otitis Media: A Case Report

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Established treatment strategies for nontuberculous mycobacterial (NTM) infections are currently lacking, and whether surgical treatment should be applied in combination with antibiotic therapy remains debatable. Here, we report a case of bilateral otitis media caused by *Mycobacterium abscessusa*, a highly antibiotic-resistant bacterium. Many reported cases of NTM otitis media are unilateral, in which hearing of the contralateral ear is preserved. In the present case, strategies to improve hearing outcomes were considered, as both ears were affected. A 27-year-old woman presented with bilateral otorrhea that had lasted for the past 9 months. Bacterial culture showed *M. abscessus* in both ears. Based on drug sensitivity tests, clarithromycin, amikacin, and imipenem were administered. Three days after treatment initiation, diseased tissues were removed from the right middle ear, which had impaired hearing. On day 38, otorrhea stopped in both ears, and the hearing improved. Computed tomography revealed air in both middle ears. No apparent recurrence was detected. Under the same antibiotic therapy, resolution of diseased tissues and improvement in hearing were similar between the ears with and without surgery, suggesting that surgery is not always necessary. This finding may be incorporated into the treatment guidelines for NTM infections in the future.

KEYWORDS: *Mycobacterium abscessus*, middle ear surgery, drug sensitivity, treatment guidelines, hearing loss, otology

INTRODUCTION

Nontuberculous mycobacteria (NTM) is a term used to collectively describe acid-fast bacteria other than *Mycobacterium tuberculosis*. NTM are ubiquitous organisms found in soil, water, and certain foods.[1] Infections with NTM are commonly associated with pulmonary diseases and are very rare in the otorhinolaryngology field.

We report a case of bilateral otitis media caused by *Mycobacterium abscessus*, a rapid grower reported to be particularly resistant to antibiotic therapy.[2] A treatment strategy for NTM infections remains unestablished, and there is an ongoing debate regarding the appropriateness of combining surgical treatment with antibiotic therapy.

Many reported cases of NTM otitis media are unilateral, in which hearing of the contralateral ear is preserved. In the present case, strategies to achieve better hearing outcomes were considered, as both ears were affected.

CASE PRESENTATION

A 27-year-old woman presented to our hospital with bilateral otorrhea that had persisted for 9 months. While she was treated with ofloxacin (OFLX) ear drops in this period, she experienced alternating periods of remission and relapse. Otorrhea was not ameliorated even with the use of OFLX ear drops for 2 months before the visit to our hospital. She was referred to our hospital after the tympanic membranes had been perforated in both ears and when granulomatous masses developed in both external auditory canals (Figure 1A and B).

The bacterial culture test for otorrhea was performed twice, detecting *M. abscessus* in both ears both the times. Thus, she was diagnosed with bilateral refractory otitis media or externa caused by this bacterium. Pure-tone audiometry revealed bilateral mixed

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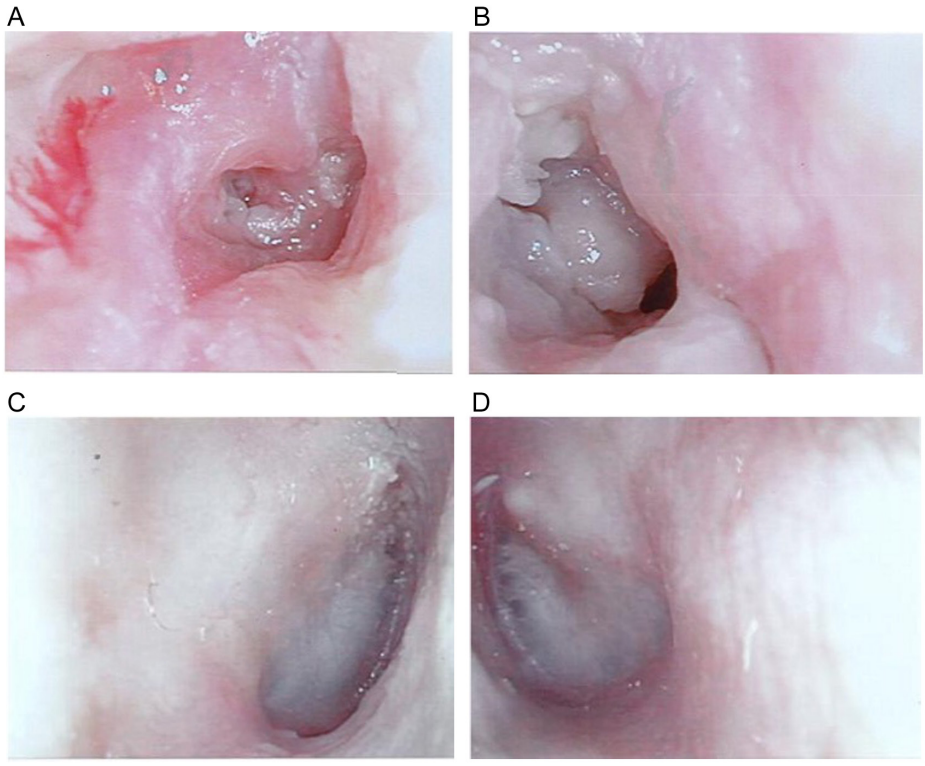

Figure 1. Physical examination and audiometric findings of the ear. A, B) At the initial visit, the skin of both (A: right ear, B: left ear) external auditory canals was edematous with pale polyps and otorrhea. The tympanic membranes were invisible. C, D) After the treatment, the edema and polyps disappeared from the skin of both (C: right ear, D: left ear) external auditory canals, and otorrhea also stopped.

conductive–sensorineural hearing loss of 63.8 dB in the right ear and 47.5 dB in the left ear (Figure 2 A). Computed tomography (CT) revealed that the tympanum and mastoid air cells were filled with soft tissue attenuation in both ears (Figure 3A and B).

We tested the drug sensitivity of the acid-fast bacterium using BrothMIC NTM, which does not target rapid growers, as it was the only available drug sensitivity test kit. The results showed that the bacterium was sensitive only to clarithromycin (CAM) and amikacin (AMK) and resistant to all other drugs. The patient was admitted and started with treatment involving intravenous administration of AMK 800 mg and oral administration of CAM 800 mg. On hospital day 3, diseased tissues were immediately removed from the tympanum and mastoid air cells of the right ear, which had impaired hearing. Although the tympanum and mastoid air cells were filled with granulation tissues, the ear ossicles were intact. The incision line healed without complications. Subsequently, we obtained BrothMIC RGM, which targets rapid growers, and performed the tests. Since the bacterium was found to be intermediately sensitive to imipenem (IMP) and sitafloxacin (STFX), in addition to CAM and AMK, the intravenous

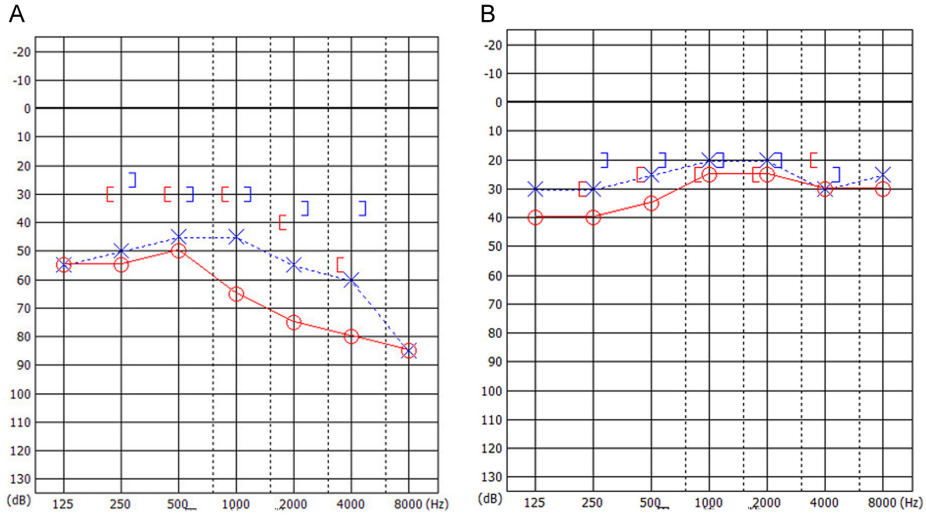

Figure 2. Audiometric findings. A) At the initial visit, pure-tone audiometry revealed bilateral mixed conductive–sensorineural hearing loss with 63.8 dB in the right ear and 47.5 dB in the left ear. B) After the treatment, air conduction hearing improved in both ears.

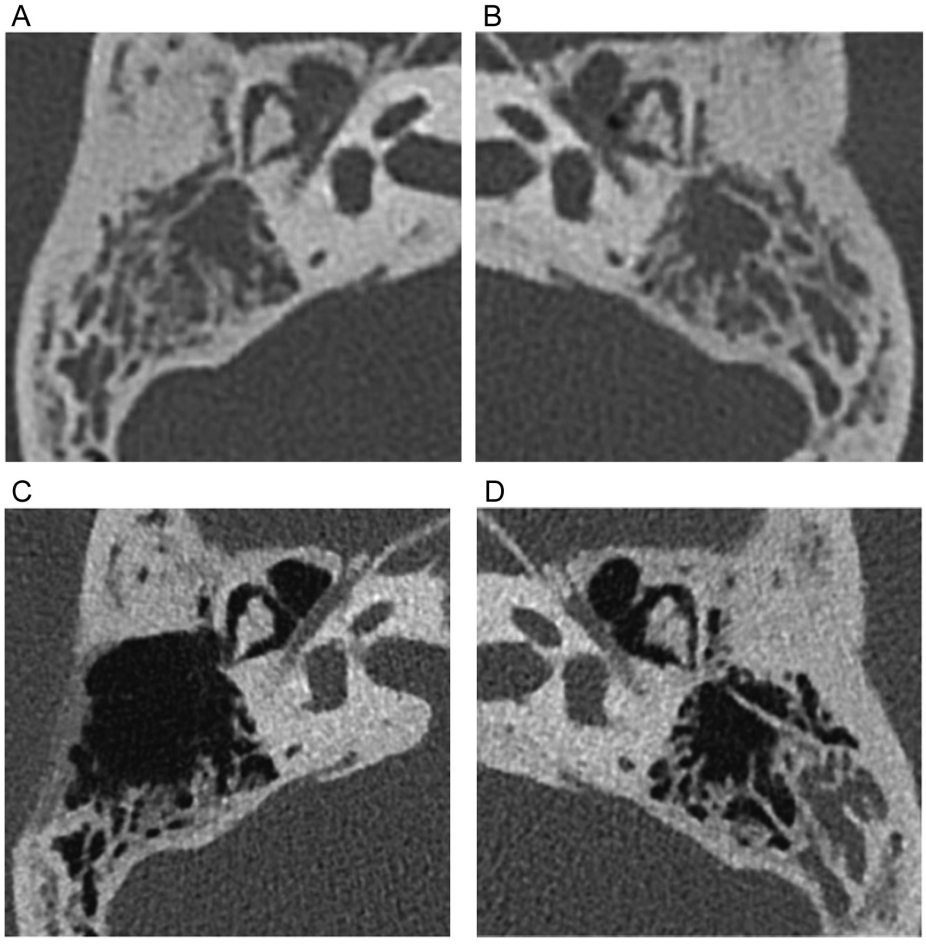

Figure 3. Computed tomography findings. A, B) At the initial visit, the tympanum and mastoid air cells were filled with soft tissue attenuation in both (A: right, B: left) ears. No apparent bone destruction was observed. C, D) After the treatment, air was observed in the tympanum and mastoid air cells of both (C: right, D: left) ears.

administration of IMP 2 g was also started on hospital day 15. On hospital day 38, otorrhea stopped in both ears (Figure 1C and D), and air conduction hearing also improved (Figure 2B). Since CT revealed air in the tympanum and mastoid air cells of both ears (Figure 3C and D), the antibiotic therapy was changed to the oral administration of CAM 800 mg and STFX 100 mg on hospital day 41. The patient was discharged from our hospital. Although itching of the ears persisted later, it disappeared 172 days after discharge. Subsequently, the oral administration of antibiotics was discontinued. No topical drugs were administered during the treatment period. Three years have passed since the treatment, and no apparent relapse has been detected. Informed consent was obtained from the patient.

DISCUSSION

Prolonged otitis media impacts hearing outcomes; therefore, early diagnosis and treatment are important. Patients who do not respond to routine treatment should undergo evaluation to assess the potential presence of acid-fast bacterial infections, even in the absence of granulation tissues. When an acid-fast bacterial infection is suspected, collaboration with the microbiology team becomes imperative to facilitate an early examination.

In this case, an earlier treatment was desirable because of the possibility of decreased hearing in both ears. Thus, the treatment was started when the bacterium was found to be sensitive to CAM and AMK using BrothMIC NTM. However, the treatment of NTM infections requires a combination of at least 3 antibiotics.[3] Treatment with 1 or 2 antibiotics causes resistance to NTM and makes treatment continuation difficult. Moreover, infection, particularly with *M. abscessus* among NTM, is refractory and difficult to cure with antibiotic therapy alone. Although no gold-standard treatment has been established, the American Thoracic Society/Infectious Diseases Society of America guidelines[4] also recommend treatment with multidrug combination chemotherapy and surgery. However, a total resection of infected tissues is difficult in the middle ear. Moreover, some patients should undergo multiple surgeries. Several sporadic reports indicate that surgery should be performed in patients who do not respond to antibiotic therapy.[5]

In the present case, whether surgical treatment should be performed was difficult to determine. Since the first drug sensitivity test with BrothMIC NTM revealed that the bacterium was sensitive to only 2 drugs, CAM and AMK, we were concerned about the possibility of decreased hearing in both ears in the case of difficulty in continuing treatment owing to the development of drug resistance. After consulting with the Department of Infectious Diseases, we decided to perform early surgery for the ear with impaired hearing.

On hospital day 38, otorrhea stopped in both the surgically treated right ear and the surgically untreated left ear. In addition, soft tissue

attenuation in the tympanum and mastoid air cells disappeared on CT, and the air conduction hearing improved. Thus, it appeared that surgical treatment was not always necessary, even for infection with *M. abscessus*, which is considered to be particularly resistant to antibiotic therapy among NTM.

CONCLUSION

We have detailed our encounter with a rare case of bilateral NTM otitis media, a condition lacking established treatment strategies. However, under the same content and duration of antibiotic therapy, resolution of diseased tissues and improvement of air conduction hearing were achieved without differences between the surgically treated right ear and the surgically untreated left ear. This suggests that surgical treatment is not always necessary. This finding may be incorporated into the guidelines for the treatment of NTM infections in the future.

Informed Consent: Informed consent was obtained from the patient who agreed to take part in the study.

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