

Mycobacteroides abscessus infection in a patient with tetralogy of Fallot

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Abstract

A 58-year-old woman with tetralogy of Fallot was referred to our hospital with a 2-month history of hemoptysis and low-grade fever. The sputum smear on admission showed positive results for acid-fast bacilli, and comparative genomic analysis of the cultured sputum confirmed *Mycobacteroides abscessus* subsp. *abscessus* infection. Long-term combination antimicrobial therapy gradually improved the patient's symptoms. Although non-tuberculous mycobacteria infection is rarely observed in patients with cyanotic congenital heart disease, a worldwide increase in non-tuberculous mycobacteria infections may increase the incidence of this rare combination.

KEYWORDS

cyanotic congenital heart disease, *Mycobacteroides abscessus*, non-tuberculous mycobacteria, tetralogy of Fallot

INTRODUCTION

Over 150 non-tuberculous mycobacteria (NTM) species have been identified, and the number of NTM infections in Japan has rapidly increased in the past decade [1]. Although NTM infections are rarely observed in patients with cyanotic congenital heart disease, the current trend may increase the incidence of NTM in patients with congenital heart diseases. Here, we report a case of *Mycobacteroides abscessus* subsp. *abscessus* infection in a patient with tetralogy of Fallot.

CASE REPORT

A 58-year-old woman with a 2-month history of hemoptysis and low-grade fever was referred to our hospital. She had been diagnosed with tetralogy of Fallot at birth but had not undergone a revascularization procedure because her hemodynamic condition was stable. The patient's condition remained unchanged until the onset of the episode described here. On admission, physical examination revealed a body temperature of 37.9°C, percutaneous oxygen saturation of 83% without supplemental oxygenation, blood pressure of 108/61 mmHg, and heart rate of 120 beats per minute. Auscultation of the heart showed a harsh grade 4/6 systolic

ejection murmur at the left mid-sternal border. Fingernail clubbing and cyanosis were observed.

Chest radiographs showed a cavitory nodule measuring 4.8 cm in the left upper lung field, infiltrative shadows in the bilateral lung fields, and the boot-shaped heart typical of tetralogy of Fallot (Figure 1A). Chest computed tomography revealed a thickened cavity wall in the left upper lobe and granular nodules in both lungs (Figure 1C, E).

Sputum test results were positive for acid-fast bacilli. Although mycobacterium infection was strongly suspected, polymerase chain reaction results for *Mycobacterium tuberculosis* and *Mycobacterium avium* complex (MAC) were negative. Additionally, blood interferon-gamma release assay results were negative. Finally, the patient was diagnosed with *M. abscessus* subsp. *abscessus* infection, confirmed based on the results of DNA sequencing analysis of two cultured sputum samples. The susceptibility analysis showed a minimal inhibitory concentration (MIC) of 0.5 µg/mL for clarithromycin and MIC of 16 µg/mL for amikacin, showing sensitivity to both antibiotics.

The patient received combination antimicrobial therapy with clarithromycin, amikacin, and imipenem/cilastatin. Her symptoms and radiological findings gradually improved after 2 months of treatment (Figure 1B, D, and F). The antibiotics were then switched to clarithromycin, amikacin, and

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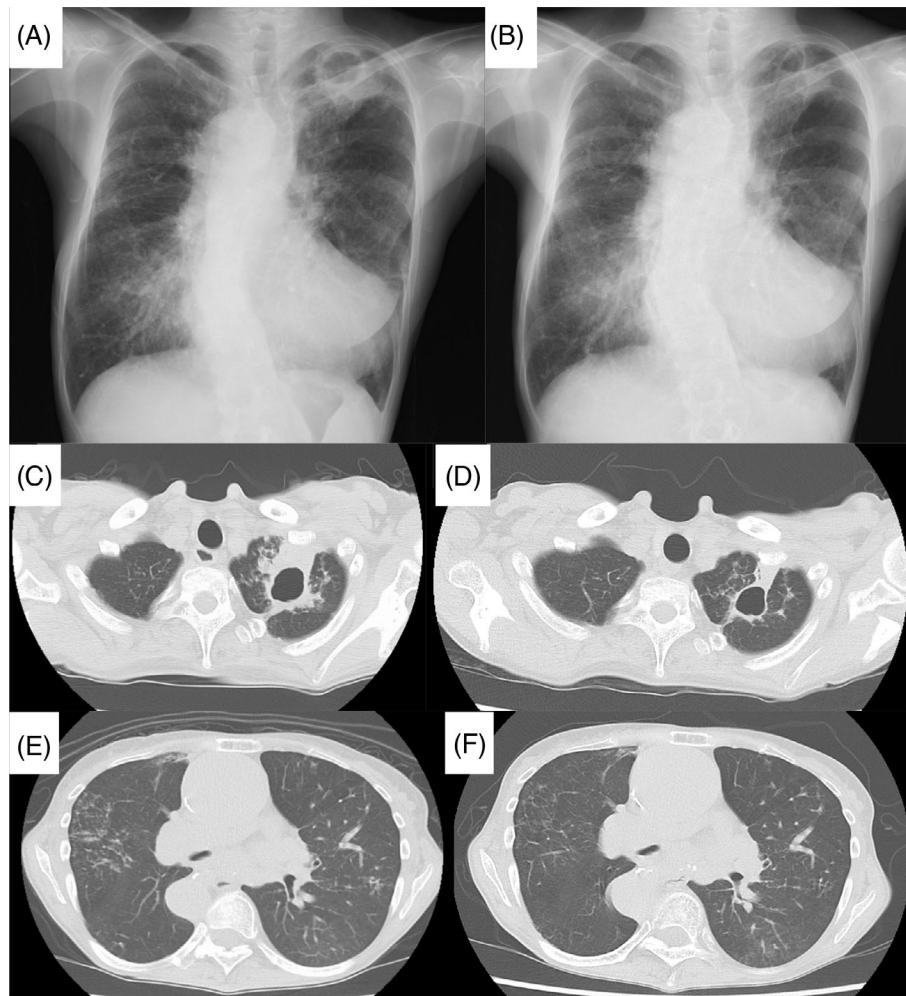


FIGURE 1 Chest radiograph (A) and computed tomography scan (C, E) on admission. Chest radiograph (B) and computed tomography scan 2 months after initial antibiotics treatment (D, F).

sitafloxacin, and her condition has remained stable for 3 years. In addition, repeated sputum cultures were all negative and radiograph findings have been unchanged.

DISCUSSION

Despite a worldwide increase in NTM cases, NTM infections are rare in patients with cyanotic congenital heart disease. Reduced pulmonary blood flow and local hypoxia have been shown to inhibit the growth of *M. tuberculosis* [2]. Therefore, patients with low-flow cyanotic heart lesions, such as tetralogy of Fallot, might be less likely to have NTM infection than general populations. Consequently, there are limited data on NTM infection in this population. To the best of our knowledge, this is the first report showing NTM infection in a TOF patient who did not undergo a revascularization procedure. However, the recent increasing trend in NTM infections may result in the increased occurrence of cases such as the one we have described.

M. abscessus complex (MABC), which can cause lung, skin, and soft tissue infections, is a rapidly growing group [3]. MABC comprises three subspecies: *abscessus*, *bolletii*, and *massiliense*. Although there are fewer MABC infection cases than MAC infection cases in Japan, the number of cases of infection with *M. abscessus* subsp. *abscessus* and *massiliense* is increasing [4]. Of these, respiratory infections caused by *M. abscessus* subsp. *abscessus* are highly resistant to treatment, and their prognosis is sometimes very poor. This is mainly because *M. abscessus* subsp. *abscessus* frequently acquires macrolide resistance through induction of the *erm(41)* gene [5]. Therefore, early diagnosis and treatment are essential. Moreover, considering the frequent acquisition of macrolide resistance, future studies should aim to determine the best combination regimen for treating infection caused by this species.

We believe there are no reports of MABC infection in patients with cyanotic congenital heart disease. This case emphasizes the need for cardiologists and paediatricians caring for patients with cyanotic congenital heart disease to be aware of the possibility of *Mycobacterium* infection. The treatment of MABC infections remains a significant

challenge, and early diagnosis and intensive antibiotic treatment are essential for good outcomes.

AUTHOR CONTRIBUTIONS

All authors qualified for authorship. Authors' main contributions regarding this manuscript are as follows. **Tomohiro Akaba:** drafting the draft, **Fumi Onizawa:** supervision, revising the draft, **Yuno Shiota:** revising the draft, **Naoko Arakawa:** revising the draft, and **Etsuko Tagaya:** supervision and final approval of the draft.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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