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Case Report

Nontuberculous mycobacterial pulmonary disease in a patient with unilateral pulmonary artery agenesis: Case report

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ABSTRACT

Nontuberculous mycobacteria (NTM) are ubiquitous organisms, but can cause a chronic pulmonary infection in some patients. Therefore, there could be host factors susceptible to this disease. A structural lung disease including damages of lungs caused by previous respiratory infection has been suggested as a host factor. Here we presented a case of NTM pulmonary disease which developed in a structural lung disease caused by a rare congenital lung disease. A 46-year-old male, was transferred to our hospital with an unexpandable lung after a closed thoracostomy due to spontaneous pneumothorax. His chest computed tomography showed an absence of left pulmonary artery at the time of admission. Mycobacterial culture in sputum, bronchial washing fluid, and pleural fluid showed the growth of NTM. *Mycobacterium intracellulare* was isolated from all positive cultures in the specimens. Combinations of drugs for *M. intracellulare* pulmonary disease including azithromycin, rifampin, and ethambutol were administered for 16 months. Amikacin intra venous treatment used for 6 months after treatment initiation. Culture conversion was achieved at 4 months of treatment. There was no evidence of recurrence of NTM pulmonary disease for 6 months after treatment. In conclusion, patients who have structural lung disease need to be careful monitoring about development of NTM pulmonary disease.

List of abbreviation

AFB	Acid fast bacilli stains
CT	Computed tomography
NTM	Nontuberculous mycobacteria

1. Introduction

Nontuberculous mycobacterial (NTM) pulmonary disease is a chronic respiratory infection caused by non-tuberculous mycobacteria, which inhabit in environment such as soil and water. NTM pulmonary disease is the most common form of infection accounts for

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more than 90% [1]. Recently, the prevalence of NTM diseases has been increased worldwide [1]. In South Korea, the frequency of NTM isolation from clinical specimens and NTM pulmonary disease have also been increasing [2,3].

NTM is a ubiquitous organism, and exposure to these bacteria is inevitable to all people, but the development of NTM infection is not common. Therefore, there could be host factors which cause a person be susceptible to NTM infection. Structural lung diseases have been known as host factors of this disease including chronic obstructive pulmonary disease, cystic fibrosis, and bronchiectasis [4,5].

Here we present a case of NTM pulmonary disease with a structural lung disease which was caused by a rare congenital lung disease.

2. Case presentation

A 46-year-old man was transferred to our hospital due to an unexpandable lung after a closed thoracostomy for his spontaneous pneumothorax (Fig. 1A and B). It had occurred 4 days before the visit of the other hospital. He had a history of recurrent pneumonia. He had a gout without receiving corticosteroid therapy and was a 30 pack-year current smoker. His chest computed tomography (CT) scans showed a cavitary consolidation in the left upper lobe and micronodules in the right upper lobe (Fig. 1C). His left lung showed structural changes including emphysema and hypoplasia (Fig. 1C). Incidentally, left pulmonary artery agenesis was noticed in his CT scans (Fig. 1D). We inserted an additional chest tube. We did acid fast bacilli stains (AFB), mycobacterial cultures, and *M. tuberculosis* PCR test in his sputa and pleural fluid. We also did a bronchoscopy test for further evaluation of TB due to negative results of AFB stain and *Mycobacterium tuberculosis* PCR test in his sputa and pleural fluid. His bronchoscopic specimens also showed negative results of AFB stain and *M. tuberculosis* PCR test. We removed his chest tubes after confirmation of fully expended his left lung on a chest radiography. He discharged from our hospital.

One month after his discharge, he revisited to our emergency department due to dyspnea. We found out a recurrent pneumothorax in his left lung and inserted a chest tube. NTM was isolated from all mycobacterial cultures in his sputa, bronchoscopic specimen, and pleural fluid, and *M. intracellulare* was identified from all positive NTM cultures. We started azithromycin (250 mg/day), rifampin (600mg/day), and ethambutol (800mg/day), and amikacin (1g intravenous tree times per week) for treating his *M. intracellulare* pulmonary disease and pleural infection. His lung was expanded well after a closed thoracostomy and discharged after chest tube removal.

We achieved a culture conversion after 4 months of treatment initiation. We stopped the intravenous amikacin after 6 months due to a development of hearing loss. We maintained the regimen for 16 months, and culture conversion maintained at the end of treatment. There was no recurrence at 6 months after treatment completion.

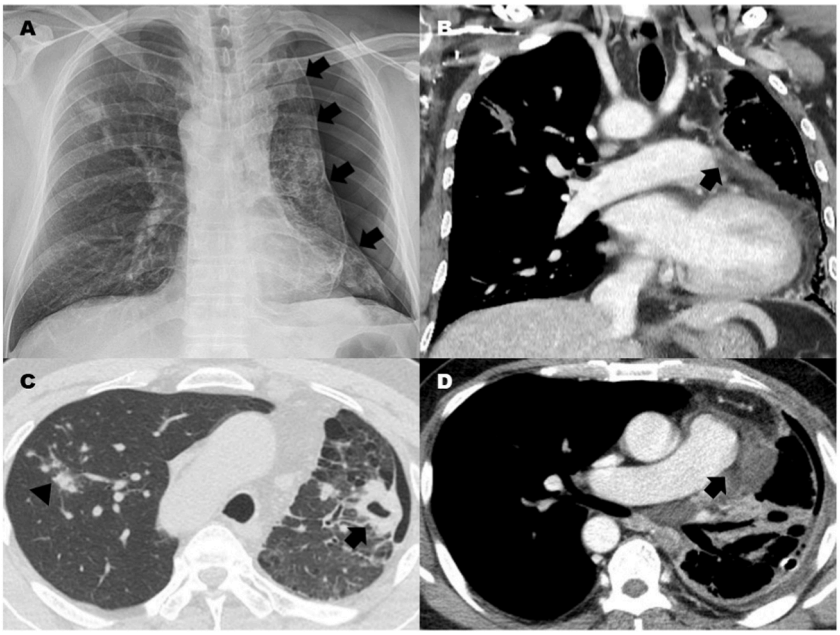

Fig. 1. Chest radiography and computed tomography images of 46 year-old man with dyspnea
A. A chest radiography showed left pneumothorax. B. Incomplete expansion of left lung (arrows) after a chest tube insertion (arrow head). C. A cavity (arrow) in the left upper lobe and micronodules in the right upper lobe were shown in the CT scan. Right lung hypoplasia was also shown in the CT scan. D. An absence of left pulmonary artery was shown in contrast enhanced chest CT scan.

3. Discussion and conclusions

In this case report, we experienced a case of NTM pulmonary disease in a rare congenital lung disease which caused recurrent respiratory infection and a structural lung change.

NTM pulmonary disease can develop in patients who are susceptible to this disease. Several risk factors have been identified including old age, the use of immunosuppressant drugs including tumor necrosis factor-a blocker and oral and inhaled corticosteroids, cystic fibrosis, primary ciliary dyskinea, and alpha 1 antitrypsin deficiency. Structural lung diseases including g bronchiectasis, chronic obstructive lung disease, and interstitial lung disease were also well known as a risk factor for this disease due to decrease in the mucociliary clearance, abnormal stagnation of sputum and airway damage caused by persistent inflammation [4,5].

Unilateral pulmonary artery agenesis is a rare congenital lung disease. Patients with this disease have a normal pulmonary trunk, but unilateral absence of a pulmonary artery branch which can cause a recurrent pulmonary infection and structural lung disease [6,7]. Patients can also have hemoptysis, chest pain, and pleural effusion.

In our case, the patient had a right pulmonary artery agenesis which caused a structural lung disease including emphysema and hypoplasia of his right lung and recurrent respiratory infection. This could be the host risk factor for developing NTM pulmonary disease and finally the patient had *M. intracellulare* pulmonary and pleural infection.

In conclusion, we report a rare case of *M. intracellulare* pulmonary and pleural infection combined with a structural lung disease caused by a unilateral pulmonary agenesis. Development of NTM pulmonary disease need to be attention if patients have a structural lung disease.

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Data availability statement

The datasets generated during the current study are available from the corresponding author on reasonable quest.

Patient consent statement

Patient has provided informed consent for publication of the case.

Declaration of competing interest

The authors have no conflicts of interest to disclose.

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