Pulmonary sequestration infected with nontuberculous mycobacteria: a report of two cases and literature review

Won-Jung Koh 1*, Goohyeon Hong 1*, Kwhanmien Kim 2, Soomin Ahn 3, Joungho Han 3

1 Division of Pulmonary and Critical Care Medicine, Department of Medicine, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea
2 Division of Thoracic and Cardiovascular Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea
3 Division of Pathology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea

1. Introduction

Pulmonary sequestration is a rare congenital malformation of the lower respiratory tract. It consists of a nonfunctioning mass of lung tissue that lacks normal communication with the tracheobronchial tree and that receives its arterial blood supply from systemic circulation [1,2]. A history of recurrent bacterial infection is a frequent feature of pulmonary sequestration. However, few cases of pulmonary sequestration infected with nontuberculous mycobacteria (NTM) have been reported [3-8]. Here, we report two cases of intralobar sequestration associated with infection with Mycobacterium avium (M. avium) and Mycobacterium abscessus (M. abscessus).

2. Case report

2.1. Case #1

A 37-year-old man was referred to our hospital for non-resolving pneumonia. One month prior, the patient presented to a private clinic with fever, cough, and sputum. Chest radiograph revealed pneumonic consolidation in the right lower lobe (Figure 1a). A chest computed tomography (CT) revealed a pneumonic consolidation in the right lower lobe containing a geographic necrotic portion and air-filled cavity, which received systemic blood supply from the descending aorta (Figure 1b). Direct smears of sputum demonstrated many acid-fast bacilli (2+). The patient was started on a four-drug regimen of isoniazid (300 mg/day), rifampicin (600 mg/day), ethambutol (1 200 mg/day), and pyrazinamide (1 500 mg/day) for presumptive diagnosis of pulmonary tuberculosis complicated with pulmonary sequestration.
Four weeks later, numerous mycobacterial colonies were cultured from three sputum specimens on Ogawa’s egg medium. All of these colonies were subsequently identified as M. avium. The identification was determined via a polymerase chain reaction–restriction fragment length polymorphism (PCR–RFLP) method, based on the rpoB gene[9]. Treatment was changed to clarithromycin (1 000 mg/day), rifampicin (600 mg/day), and ethambutol (800 mg/day). While on antibiotic treatment for two months, the patient clinically improved; however, his right lower lobe consolidation remained unchanged. The patient underwent right lower lobectomy by video-assisted thoracoscopic surgery (VATS). An anomalous feeding artery supplying the right lower lobe was found. Microscopic examination revealed multiple caseating granulomas (Figure 1c).

![Figure 1. A 37-year-old man with pulmonary sequestration infected with M. avium.](image)

(a) Chest radiograph showed a mass–like consolidation in the right lower lung. (b) Chest CT revealed a pneumonic consolidation in the right lower lobe, containing a geographic necrotic portion and air–filled cavity, which received systemic blood supply (arrow) from the descending aorta. (c) Histopathology of the lung showed caseating granuloma with necrosis (H–E stain, ×100).

The patient`s postoperative course was uneventful. He continued on treatment with the same antibiotics for 6 months following surgery. The patient was doing well 40 months after surgery.

2.2. Case #2

A 26–year–old woman visited our hospital because of fever and cough. She had been well until 5 days before visiting our hospital. A chest radiograph showed a large mass–like consolidation in the right lower lung (Figure 2a). Chest CT revealed a multicystic mass–like consolidation in the right lower lobe (Figure 2b), which was supplied by an anomalous systemic artery originating from the descending aorta (Figure 2c). A sputum smear for acid–fast bacilli was negative. She was diagnosed with pulmonary sequestration complicated with bacterial infection and received intravenous ceftriaxone plus azithromycin. Her fever and symptoms improved after antibiotic therapy. After 7 days of antibiotic therapy, the patient underwent right lower lobectomy by VATS. An aberrant artery originating from the descending aorta just above the diaphragm and entering into the right lower lobe was found. Microscopic examination revealed multiple caseating granulomas.

![Figure 2. A 26–year–old woman with pulmonary sequestration infected with M. abscessus.](image)

(a) Chest radiography showed a large mass–like consolidation in the right lower lung. (b) Chest CT revealed a multicystic mass–like consolidation in the right lower lobe, which was supplied by (c) an anomalous systemic artery (arrow) originating from the descending aorta.

Ten days after surgery, preoperative sputum culture was reported positive for NTM and M. abscessus complex (sensu lato) was identified by a PCR–RFLP method using the rpoB gene[10]. Further differentiation among M. abscessus complex members was done using sequence analysis targeting the rpoB and hsp65 genes and M. abscessus (sensu strict) was confirmed[10]. Oral clarithromycin was administered for 3 months after a 1–week intravenous treatment of amikacin and cefoxitin. The patient was doing well 36 months after surgery.

3. Discussion

Pulmonary sequestration is a rare malformation characterized by the presence of lung tissue with abnormal or absent communication with the bronchial tree and arterial blood supply from systemic circulation. Patients present with signs and symptoms of pulmonary infection of a lower lobe mass. It is believed that sequestrations become infected when bacteria migrate through the pores of Kohn or if the sequestration is incomplete[5]. Despite the high frequency of infections, few data are available regarding specific infecting organisms[7].

Patients with infected pulmonary sequestration due to NTM have been reported only rarely. In a review of the literature, only seven cases of pulmonary sequestration complicated by infection with NTM were found. All of the reported cases of pulmonary sequestration associated with NTM infection, including our cases, are summarized in Table 1. M. avium–intracellulare complex (MAC) were the most common etiologic organisms, and various NTM infections, including Mycobacterium kansasii and Mycobacterium gordonae, were possible etiologic agents in the previously reported cases. However, M. abscessus has never been reported in the literature on the etiology of infected pulmonary sequestration. Our second case is the first reported case with pulmonary sequestration infected with M. abscessus.

Combination antibiotic therapy was recommended
Pulmonary sequestration complicated by nontuberculous mycobacterial infection in the literature.

<table>
<thead>
<tr>
<th>Author (year)</th>
<th>Sex</th>
<th>Age</th>
<th>Type</th>
<th>Organism</th>
<th>Type of surgery</th>
<th>Antibiotics</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mooney et al. (1975)[3]</td>
<td>male</td>
<td>24</td>
<td>intralobar</td>
<td>M. avium complex</td>
<td>Open thoracotomy</td>
<td>Antituberculosis drugs</td>
</tr>
<tr>
<td>Sekine et al. (1998)[4]</td>
<td>female</td>
<td>30</td>
<td>NA</td>
<td>M. avium</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Shiota et al. (2002)[5]</td>
<td>female</td>
<td>29</td>
<td>intralobar</td>
<td>Mycobacterium intracellulare</td>
<td>Open thoracotomy</td>
<td>CLR RFP EMB for 6 mo</td>
</tr>
<tr>
<td>Miyazaki et al. (2004)[6]</td>
<td>female</td>
<td>28</td>
<td>intralobar</td>
<td>Mycobacterium avium</td>
<td>NA</td>
<td>Antibiotics*</td>
</tr>
<tr>
<td>Miyazaki et al. (2004)[6]</td>
<td>female</td>
<td>25</td>
<td>intralobar</td>
<td>M. avium</td>
<td>NA</td>
<td>Antibiotics*</td>
</tr>
<tr>
<td>Lin et al. (2005)[7]</td>
<td>male</td>
<td>33</td>
<td>intralobar</td>
<td>Mycobacterium kansasii</td>
<td>VATS</td>
<td>None</td>
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<tr>
<td>Umeda et al. (2009)[8]</td>
<td>female</td>
<td>72</td>
<td>extralobar</td>
<td>Mycobacterium gordonae</td>
<td>Open thoracotomy</td>
<td>None</td>
</tr>
<tr>
<td>Present study</td>
<td>male</td>
<td>37</td>
<td>intralobar</td>
<td>M. avium</td>
<td>VATS</td>
<td>CLR RFP EMB for 6 mo</td>
</tr>
<tr>
<td>Present study</td>
<td>female</td>
<td>26</td>
<td>intralobar</td>
<td>M. abscessus</td>
<td>VATS</td>
<td>CLR for 3 mo plus AMK and CFXT for one week</td>
</tr>
</tbody>
</table>

NA = not available, CLR = clarithromycin, RFP = rifampicin, EMB = ethambutol, VATS = video-assisted thoracoscopic surgery, AMK = amikacin, CFXT = cefoxitin.

* Details regarding antibiotic regimen and durations were not available.

For treatment of MAC or M. abscessus lung disease[11,12], however, surgical removal should be advocated in both symptomatic and asymptomatic cases of pulmonary sequestration, and the benefit of a period of perioperative antibiotic treatment has been controversial[7]. Some patients simply underwent a surgical resection and did not receive any antibiotic treatment in the published reports[7,8]. Surgery is the treatment of choice, usually performed through a posterolateral thoracotomy[13-15]. More recently, the traditional approach of thoracotomy has been successfully replaced by VATS[16,17]. Our patients successfully underwent VATS lobectomy without perioperative morbidity. VATS lobectomy has been performed with increasing frequency and applied to various lung diseases including benign inflammatory and congenital lung diseases. In our institution, VATS lobectomy was successfully performed in 14 patients with pulmonary sequestration, including the two cases in this report[8]. In summary, we report unique cases of NTM infection in a sequestrated lung in young adults. Surgical resection, in particular VATS resection, together with combination antibiotic therapy, established the correct diagnosis and removed the focus of infection.

**Conflict of interest statement**

We declare that we have no conflict of interest.

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**References**