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

Won–Jung Koh^{1**}, Goohyeon Hong^{1*}, Kwhanmien Kim², Soomin Ahn³, Joungho Han³

¹Division of Pulmonary and Critical Care Medicine, Department of Medicine, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea

²Division of Thoracic and Cardiovascular Surgery, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea

³Division of Pathology, Samsung Medical Center, Sungkyunkwan University School of Medicine, Seoul, South Korea

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ABSTRACT

We report two cases of pulmonary sequestration infected with nontuberculous mycobacteria (NTM): *Mycobacterium avium* and *Mycobacterium abscessus*. Chest computed tomography showed pneumonic consolidation in the right lower lobe, which received a systemic blood supply from the descending aorta in both patients. Video–assisted thoracoscopic surgeries were successfully performed and pathological examinations revealed multiple caseating granulomas. A review of the literature revealed only seven previous case reports of pulmonary sequestration infected with NTM, and no case with *Mycobacterium abscessus* has been reported.

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. His symptoms and chest radiographic findings did not improve, despite two weeks of antibiotic treatment.

A chest radiograph showed a mass–like consolidation in the right lower lung (Figure 1a). 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.

*Corresponding author: Won–Jung Koh, MD, Division of Pulmonary and Critical Care Medicine, Department of Medicine, Samsung Medical Center, Sungkyunkwan University School of Medicine, Irwon–ro 81, Gangnam–gu, Seoul, 135–710, Korea.

Tel: (+82) 2–3410–3429

Fax: (+82) 2–3410–3849

E–mail: wjkoh@skku.edu

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*: These authors contributed equally to this work.

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* gene⁹. 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).

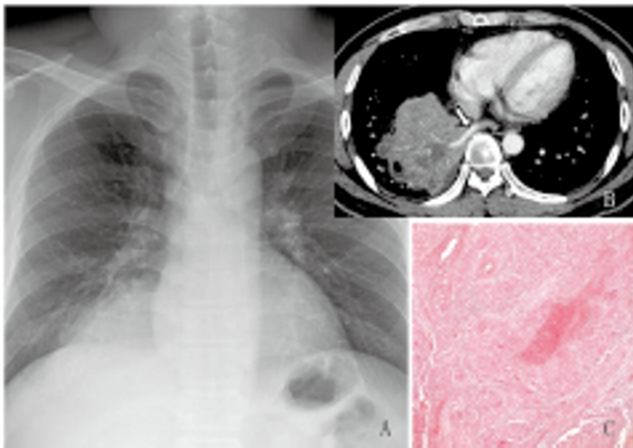


Figure 1. A 37-year-old man with pulmonary sequestration infected with *M. avium*.

(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.

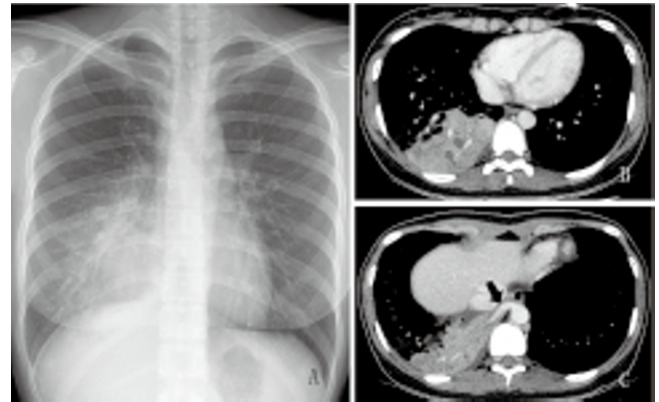


Figure 2. A 26-year-old woman with pulmonary sequestration infected with *M. abscessus*.

(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* gene¹⁰. Further differentiation among *M. abscessus* complex members was done using sequence analysis targeting the *rpoB* and *hsp65* genes and *M. abscessus* (sensu strict) was confirmed¹⁰.

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 incomplete⁵. Despite the high frequency of infections, few data are available regarding specific infecting organisms⁷.

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

Table 1

Pulmonary sequestration complicated by nontuberculous mycobacterial infection in the literature.

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

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[18].

In summary, we report unique cases of NTM infection in a sequestered 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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