

A Surgically Treated Case of Chronic Necrotizing Aspergillosis with Pleural Invasion

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Aspergillus is a ubiquitous fungus and can cause many levels of disease severity. Chronic necrotizing aspergillosis is a rare disease and few cases have been reported in Korea. We experienced a case of pleural aspergillosis that was treated successfully with medical and surgical interventions. The 52-year-old man who was diagnosed with chronic necrotizing pulmonary aspergillosis underwent surgical treatment including a lobectomy, decortication, and myoplasty. The patient was also medically treated with amphotericin B followed by voriconazole. Pleural irrigation with amphotericin B was also performed. A multi-dimensional approach should be considered for treating chronic necrotizing pulmonary aspergillosis.

Key words: 1. Pleural disease
2. Fungal infection
3. Decortication
4. Lung surgery
5. Aspergillosis

CASE REPORT

A 52-year-old male patient complained of pleuritic pain of the right chest, a persistent cough, and yellowish blood-tinged sputum for two months. He had been diagnosed with pulmonary tuberculosis and completely recovered after medical treatment; he had no other underlying diseases. The patient was a heavy daily drinker and had lost 7 kg over the previous six months before admission to the hospital. His blood pressure was 125/80 mmHg, pulse rate was 100/min, and body temperature was normal. Right breathing sounds were decreased with crackles. Chest radiographs showed old tuberculosis scars and a newly developed empyema in the right lung field (Fig. 1A). Tomography showed several cavitary le-

sions of different sizes, some of which had fluid collection, which suggested an infected cyst, an old tuberculosis scar, or active tuberculosis. Partial calcification of the pleura, pleural thickening, and pleural effusion were also found (Fig. 2A). A yellowish substance was located in the lumen of the right upper lobe apical segmental bronchus at bronchoscopy and a pathologic examination of the substance was performed. The pathologist reported the substance to be pulmonary aspergillosis (Fig. 3B).

The empyema was drained through a percutaneous catheter and air leakage was observed. Amphotericin B (AMB) was injected after the patient was diagnosed with pulmonary aspergillosis. After the administration of AMB, a fever that the patient had since the second day of admission subsided,

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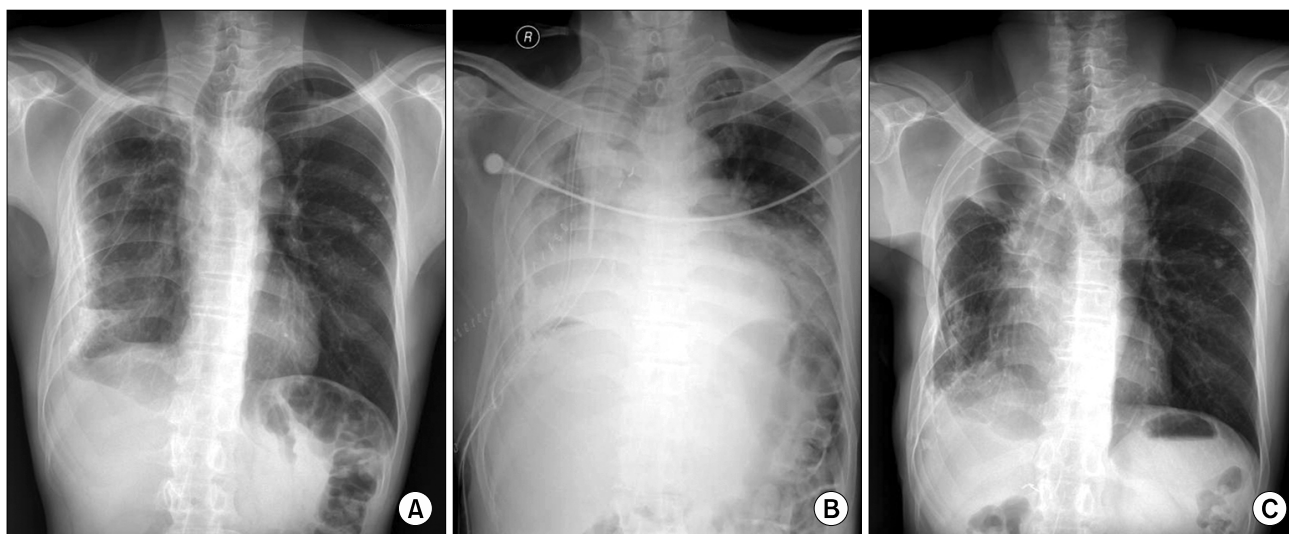


Fig. 1. Chest radiograph before operation showed empyema in the right pleural cavity (A), Postoperative chest radiograph (B), and recent chest radiograph (C).

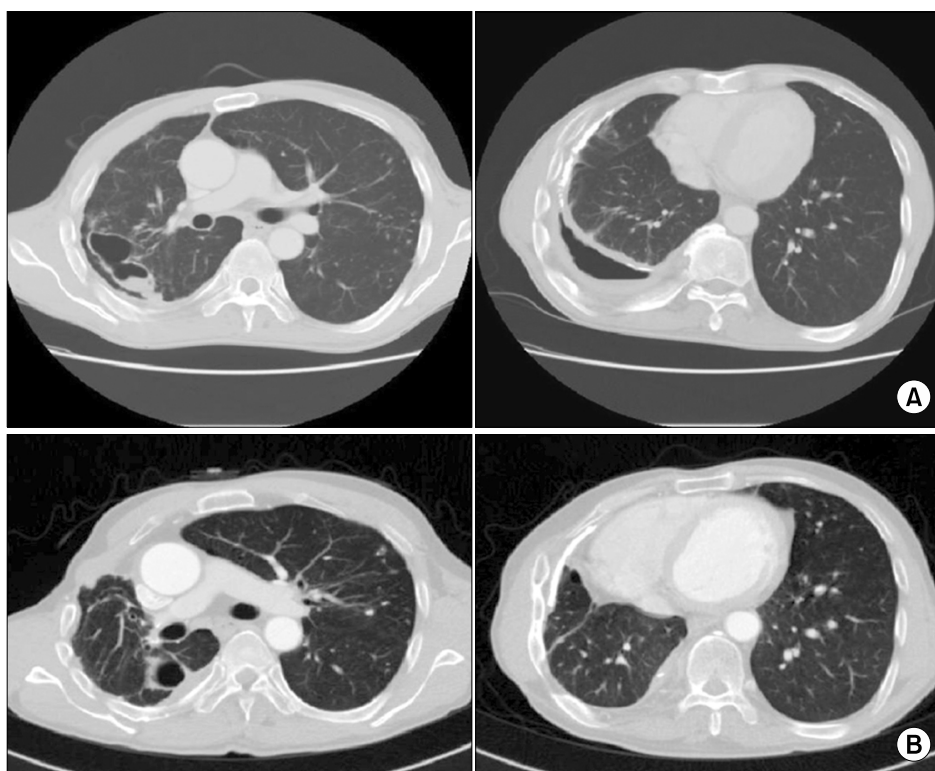


Fig. 2. (A) Tomography before the operation shows cavitory lesions and pleural lesions in the right pleural cavity. (B) Recent tomography shows a small cavity and no pleural effusion.

the white blood cell count decreased from 12,900/mL to 7,900/mL, and C-reactive protein decreased from 22.9 mg/dL to 4.4 mg/dL. On the fourteenth day of admission, a 3 French Fogarty catheter was introduced to the right upper lobe apical segmental bronchus and the bronchus was obli-

terated with the balloon under general anesthesia. After the obliteration of the bronchus, the air leakage decreased dramatically. The location of the fistula was confirmed with a fistulography by using a percutaneous catheter and a blockage test with the balloon of the Fogarty catheter. We attempted

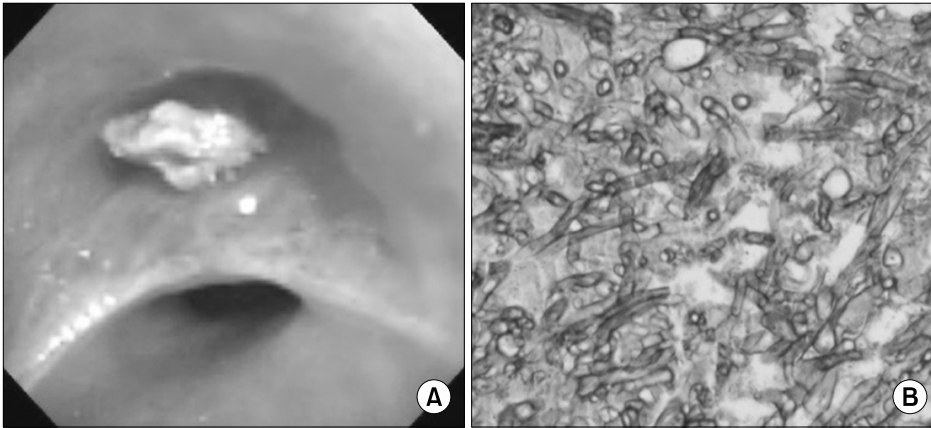


Fig. 3. (A) Bronchoscope showed a mass of *Aspergillus* in the right upper lobe apical segmental bronchus. (B) Methenamine silver stain of *Aspergillus*, $\times 400$.

fistula occlusion with BioGlue and a coil while the bronchus was blocked with the Fogarty catheter balloon, but the occlusion of the fistula failed due to the existence of multiple broncho-pleural fistulae.

Nutritional support and AMB injections continued for 1 month and during that period the patient gained 3 kg and his nutritional state improved. Pleural cavity irrigation with AMB mixed with 1 L of 5% dextrose was performed for 5 hours daily (the total amount of AMB was 1,300 mg for irrigation). However, the patient developed an intermittent fever again. Since the persistent air leakage and drainage of turbid fluid through the percutaneous catheter lasted for 1 month, a lobectomy and pleural decortication was performed. No air leakage was observed and the fever subsided after the operation. A pathologic exam showed *Aspergillus* invading the pleura.

Two weeks after the operation, the patient developed a fever, leukocytosis, and air leakage. Computed tomography showed an air space at the posterior pleural cavity. Irrigation with 12,000 mL of normal saline was performed in the operating room and the absence of air leakage through the bronchus stump was confirmed. However, air leakage was observed from the surface of the lung. The antifungal agent was converted to an oral voriconazole, and the patient was discharged with one chest tube to which a Heimlich valve was connected since there was air leakage and a small amount of a yellowish substance drained through the chest tube.

The patient developed massive bleeding (1,000 mL) through the chest tube 5 months after being discharged. The intra-operative finding was that the bleeding originated from the chest tube tract. Bleeding control and myoplasty with the

pectoralis major muscle and latissimus dorsi were performed to fill the dead space of the pleural cavity. There had been no dead space observed immediately after the operation, but dead space in the pleural cavity had eventually formed. However, the dead space gave rise to no clinical problems. The patient was discharged without the chest tube and no complications have arisen for 15 months post-retreatment.

DISCUSSION

Aspergillus is a ubiquitous fungus; about 200 species have been reported, but only a few are pathogenic.

Aspergillosis is classified into invasive aspergillosis, saprophytic aspergillosis, and the allergic form of aspergillosis. Invasive aspergillosis is a hematologic systemic infection or a direct infection from a former infection route. Although Walsh et al. [1] classified chronic necrotizing aspergillosis (CNA) within invasive aspergillosis, CNA is a different disease entity than invasive aspergillosis or the saprophytic form of aspergillosis [2].

Aspergillus does not usually cause life-threatening medical problems in immunocompetent patients. With HIV infection, chemotherapy for malignant cancer, hematopoietic stem cell transplantation, or organ transplantation, systemic infection of *Aspergillus* causes problems more frequently.

CNA is also called semi-invasive aspergillosis or subacute invasive aspergillosis; it causes progressive lung and pleural injury without prominent symptoms, and usually the pathogen is *Aspergillus fumigatus*. In particular, CNA that invades the pleura is rarely reported. CNA progresses for several months

and does not invade the blood vessels or other organs. CNA is characterized as a local invasion. These are the differences between CNA and invasive aspergillosis. Unlike saprophytic aspergillosis, CNA does not require prior lung injury [2].

As we reviewed in this case, the immunosuppressive middle-aged or elderly patients are usually vulnerable to CNA and complain of fever, cough, sputum, and weight loss. The standard diagnostic methods are a microscopic examination or culture, a pathologic confirmation of *Aspergillus* from the sample that is acquired from bronchoalveolar lavage, or a video assisted thoroscopic surgery biopsy. Galactomannan EIA and (1->3)-beta-D-glucans serum tests are of diagnostic help to some extent [1].

The oral form of itraconazole is recommended and voriconazole is suspected to be just as effective as itraconazole for CNA [1], but voriconazole is preferred due to the excellent coverage of a wide spectrum of fungus types [3].

Hargis et al. [4] performed an intracavitary instillation with AMB successfully and reported a decrease in the size of the cavity. When a patient is tolerant of pleural irrigation, treatment with AMB can be attempted.

Although surgical treatment of CNA is not usually recommended due to the risk of the surgery itself, persistently symptomatic patients or complications such as hemoptysis or frequent secondary infection through broncho-pleural fistula are indications for surgical intervention. Our patient showed empyema, bronchopleural fistula (BPF), and prolonged respiratory symptoms; aggressive surgical intervention was necessary. If a lung lesion adjacent to a large vessel and invaded pleura is observed, then the treatment of choice is a resection of the lung lesion and decortications [1].

Ahn et al. [5] recommended a myoplasty with the intercostals, pectoralis major, latissimus dorsi, and omentoplasty to

prevent BPF and dead space of the pleural cavity after a lung resection.

Although it was subclinical, our patient showed a partial cavitory recurrence after the obliteration of the cavity. We suspect that muscle atrophy and migration were the main causes. Grima et al. [6] suggested that a limited thoracoplasty combined with a myoplasty and a cavernostomy resulted in immediate good results and the prevention of late recurrence.

CNA is a rare disease but can cause many complications. Surgical treatment can be a good choice for treatment of CNA. In addition, we need to consider multiple treatment modalities including medical and surgical options, and local irrigation to treat CNA with pleural invasion.

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