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Case Report

A Case of Endobronchial Actinomycosis with a Broncholith cured by Cryotherapy through a Flexible Bronchoscope

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기관지 방선균증 및 결석을 굴곡 내시경을 통한 냉동요법으로 치료한 증례

유진석 · 조은주 · 구상건 · 권혜정 · 임성경 · 장태원 · 옥철호

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We report the case of a 53-year-old man who presented with obstructive pneumonitis and broncholithiasis. We attempted to remove the broncholith with forceps through a flexible endoscope, but the potential for bleeding due to partial synechia did not allow this. We succeeded in removing it with cryotherapy. The histopathological diagnosis was thoracic actinomycosis associated with broncholithiasis. Endobronchial actinomycosis with a broncholith is very rare. We successfully treated a patient with endobronchial actinomycosis with a broncholith by administering short-term antibiotics after broncholithectomy via cryotherapy through a flexible bronchoscope.

Key Words: Actinomycosis, Broncholith, Bronchoscopy, Cryotherapy

Endobronchial actinomycosis is a rare suppurative granulomatous infection of the bronchus, and association with foreign body aspiration or broncholith is very rare. It is sometimes misdiagnosed as endobronchial carcinoma. In cases of endobronchial actinomycosis with a broncholith, the broncholith is removed via a thoracotomy and then the actinomycosis is treated in Korea.[1-3] Only one case of endoscopic removal of a broncholith has been reported in Japan.[4] To treat endobronchial actinomycosis, intravenous antibiotics are recommended for 2-6 weeks, followed by oral antibiotics for 6-12 months.[5] However, in a case of endobronchial actinomycosis with a foreign body or broncholith, it was recently reported that the administration of antibiotics for less than 2 months after removing the lesion in the bronchus resulted in a cure.[6] We successfully treated a case of endobronchial actinomycosis with a broncholith by administering antibiotics for the short term after performing a broncholithectomy via cryotherapy through flexible bronchoscope.

CASE REPORT

A 53-year-old man visited our hospital because of a fever and dry cough for 10 days. There was no sputum, hemoptysis, dyspnea, or weight loss. The family history was non-contributory. He smoked

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three packs of cigarettes daily for 30 years and drank one bottle of soju daily for 30 years. He was a supervisor at a container factory. He had a blood pressure of 120/80 mmHg, pulse of 80/min, respiratory rate of 20/min, and body temperature of 36.5℃. On physical examination, his general condition was normal. His heart and lung sounds were normal on chest auscultation. The peripheral blood examination showed hemoglobin 12.6 g/dL, hematocrit 36.6%, 7800/mm^3 leukocytes, and 469,000/mm^3 platelets. The serum biochemistry included total protein 6.8 g/dL, albumin 3.4 g/dL, total bilirubin 0.6 mg/dL, AST 23 IU/L, ALT 26 IU/L, rGTP 151 U/L, blood urea 18 mg/dL, creatinine 1.0 mg/dL, and LDH 267 IU/L. The serum electrolytes were sodium 139 mEq/L, potassium 4.4 mEq/L, and chloride 102 mEq/L. The chest x-ray showed a focal infiltration in the right middle lobe (Fig. 1). Chest computed tomography (CT) showed a calcified stone in the bronchus and atelectasis of the right middle lobe (Fig. 2). Bronchoscopy showed a calcified stone in the same area and a tissue biopsy was obtained in this region. We tried to remove the broncholith with forceps, but this was impossible because of narrowing proximal to the broncholith due to edema, which increased the risk of bleeding (Fig. 3).

We considered broncholithectomy via cryotherapy possible and we succeeded in removing the broncholith by cryotherapy through a flexible bronchoscope under general anesthesia. The stone measured

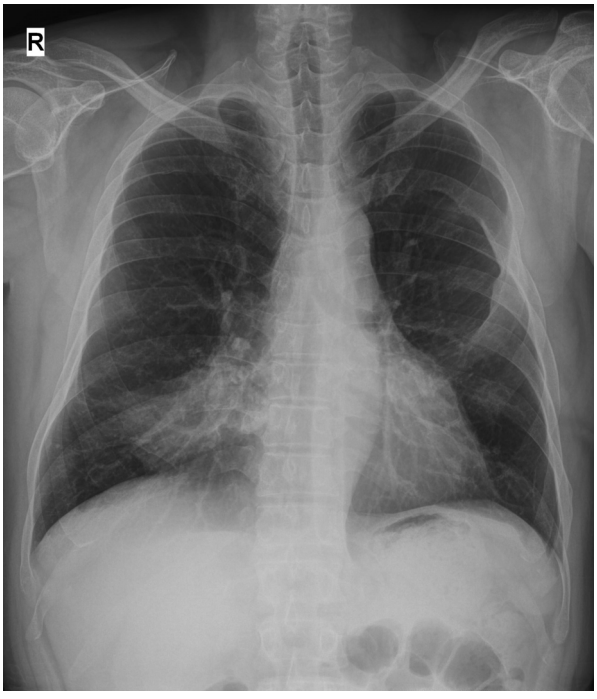

Fig. 1. Chest radiograph shows focal infiltration in the right middle lobe.

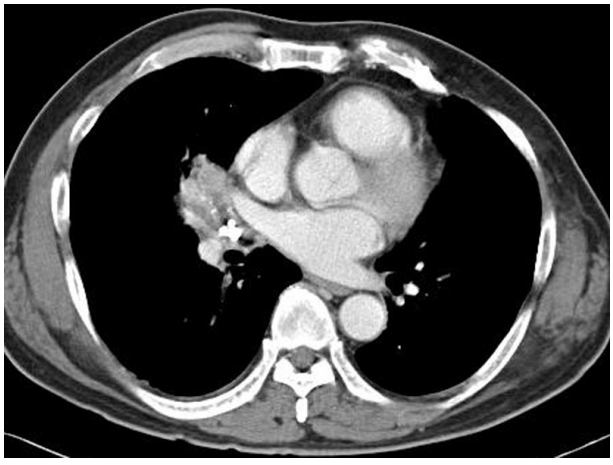

Fig. 2. Chest CT shows a calcified stone in the bronchus and atelectasis in the right middle lobe.

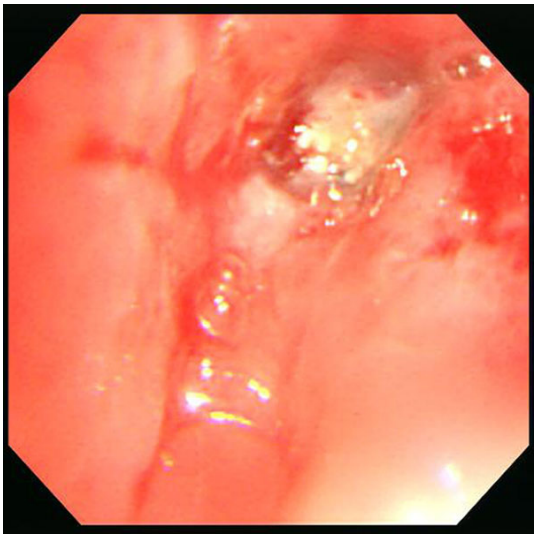

Fig. 3. Bronchoscopy shows near-total obstruction of the right middle lobe by broncholith.

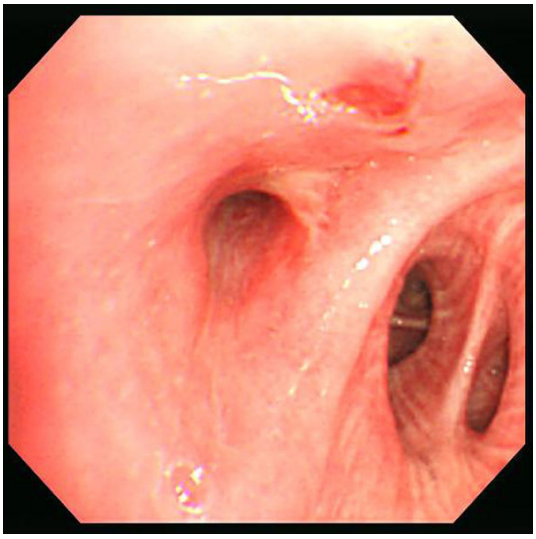

Fig. 4. There were no specific findings at follow-up bronchoscopy 5 days after the cryotherapy procedure.

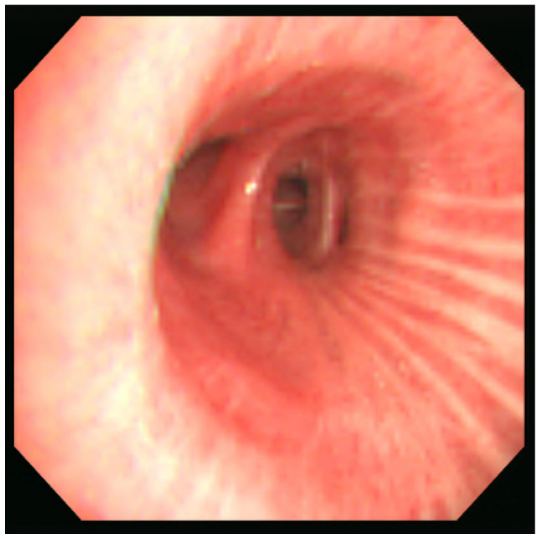

Fig. 6. There were no specific findings at follow-up bronchoscopy 8 weeks after the cryotherapy procedure.

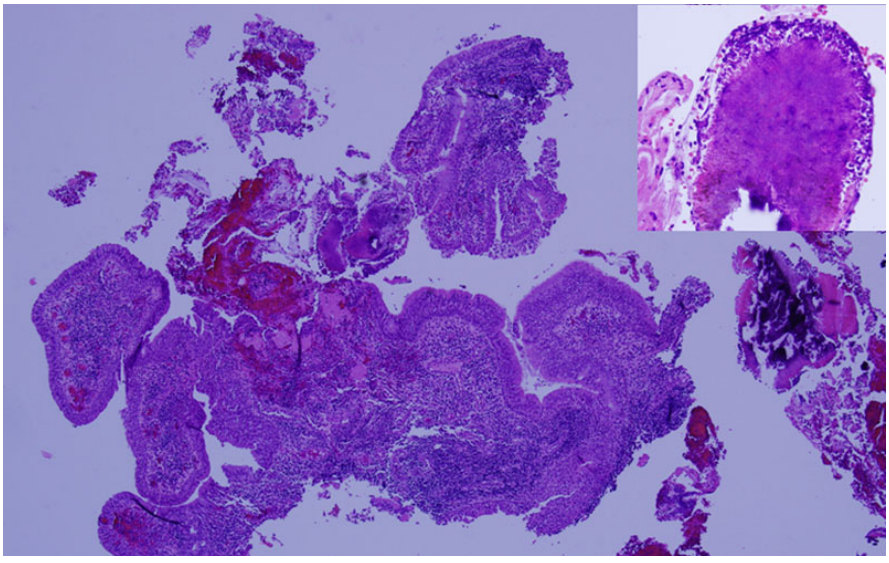

Fig. 5. The bronchoscopic biopsy showed marked lymphocyte infiltration and a few calcified fragments with radiating filamentous basophilic bacterial colonies (H&E, × 40).

1.3 cm in diameter. No complications arose during or after the cryotherapy procedure. Follow-up bronchoscopy 5 days after the cryotherapy procedure was normal (Fig. 4).

The tissue biopsy confirmed actinomycosis and showed no malignant cells, tuberculosis, or inflammation (Fig. 5). Intravenous cefoperazone/sulbactam 2 g every 12 h was given for 15 days starting from the initial visit, based on a diagnosis of obstructive pneumonia. After obtaining the tissue biopsy result, intravenous ampicillin 2 g every 6 h was given for 5 days. After discharge, oral amoxicillin was administered for 8 weeks. On follow-up endoscopic examination of the bronchus, there were no more lesions (Fig. 6). The patient showed no sign of recurrence 48 months later. We will continue to monitor the

patient.

DISCUSSION

Actinomycosis is a chronic suppurative infection caused by anaerobic Actinomyces species, a gram-positive member of the normal bacterial flora of the oral cavity, pharynx, digestive system, and female genitalia. Human infection results mainly from Actinomyces israelii. Actinomycosis arises mainly on the face, neck, chest, and abdomen. Thoracic actinomycosis constitutes 20% of all actinomycosis, and endobronchial actinomycosis with a broncholith is very rare.[7,8]

In Korea, seven reported cases of actinomycosis have been diagnosed by biopsy after a broncholithectomy via a thoracotomy.1 In Japan, Watanabe et al. removed a broncholith via endoscopy and found a sulfur granule on biopsy, diagnosing actinomycosis.4 Hiroyoshi et al. also removed a broncholith at thoracotomy and diagnosed actinomycosis based on a biopsy.[9]

The mechanism of thoracic actinomycosis related to broncholithiasis is as follows: an existing broncholith or aspirated foreign body is infected by actinomycosis, which causes inflammation, and it grows and obstructs the bronchus, leading to obstructive pneumonia.[10] In one case, there was no foreign body; the tube-shaped broncholith had the shape of the bronchus and an Actinomyces colony was found in the broncholith, indicating that the broncholith was generated secondarily following chronic inflammation due to an Actinomyces infection.[11]

The symptoms of actinomycosis include a cough, sputum, fever, chest pain, weight loss, and hemoptysis, which are all non-specific. The radiological findings of thoracic actinomycosis are also non-specific. With pulmonary parenchyma actinomycosis, CT shows chronic segmental airspace consolidation, with a hypointense signal and contrast enhancement, with adjacent pleural thickening.[12] In endobronchial actinomycosis with a broncholith, CT shows a calcified stone located in the proximal bronchus and post-obstructive consolidation of the distal pulmonary lobe or pulmonary segment.10 This is often misdiagnosed as pulmonary tuberculosis, lung cancer, or pneumonia. The diagnosis is confirmed by identification of typical bacteria or identifying sulfur granules in conjunction with Gram staining and culture of a biopsy specimen. Endoscopic bronchial lavage is not recommended because the physiological saline blocks the proliferation of Actinomyces.[6]

For treatment, high-dose antibiotics are administered, typically penicillin 180,000-240,000 units/day intravenously for 2-6 weeks and then oral penicillin or amoxicillin for 6-12 months.[5] When penicillin treatment fails or in patients allergic to penicillin, then tetracycline, clindamycin, rifampin, lincomycin, or streptomycin can be administered. Pregnant women can be given erythromycin. In recent reports, actinomycosis was cured with short-term antibiotics. To treat endobronchial actinomycosis with a foreign body in the bronchus, Kanako et al. removed the foreign body via endoscopy and then administered oral amoxicillin for 1 month. They reported no recurrence for 14 months thereafter. This suggests that the long-term administration of antibiotics is not always necessary if the foreign body can be removed.[13] In Korea, a case of endobronchial actinomycosis was cured without recurrence for 31 months with the administration of antibiotics for less than 40 days after removing the lesion. This reinforces the argument that

endobronchial actinomycosis can be cured by shorter-duration antibiotic regimens after removal of the lesion.1 In our case, there were no signs of recurrence over 48 months after a total of 11 weeks of antibiotic treatment; this agrees with reports that long-term antibiotics are not required after broncholith removal.

A broncholithectomy can be performed surgically or endoscopically. The success rate of endoscopic removal of a broncholith depends on whether the broncholith is attached to the bronchial wall: it is 48% in cases of partial synechia and 100% in cases of non-synechia. With a mobile broncholith or partial synechia that cannot be removed with forceps through a flexible bronchoscope, cryotherapy is recommended as an alternative.[14] When a broncholith is too big to pass through the bronchus, a YAG laser can be considered when it is difficult to break the broncholith with forceps. In the case of partial synechia, the endoscopic removal of a broncholith might cause bleeding, bronchial tearing, or a bronchial fistula, albeit rarely. The complication and mortality rates resulting from endoscopic broncholith removal are lower than those following surgical removal.[15] Therefore, in the case of non-synechia or partial non-synechia, endoscopic broncholith removal can be considered. With hemoptysis, strong synechia, failed endoscopic removal, or non-exclusion of cancer and to make an accurate diagnosis, surgical removal can be considered. Administration of antibiotics is necessary even following surgical removal. In our case, we attempted to remove the broncholith with forceps through a flexible endoscope, but the potential for bleeding due to a partial synechia did not allow this. We succeeded in removing it by cryotherapy without complications. This is the first published case of removal of a broncholith using cryotherapy in Korea. If removal of a broncholith through a flexible bronchoscope is difficult due to adhesions or the risk of bleeding, cryotherapy is a good alternative.

We successfully treated a case of endobronchial actinomycosis with a broncholith by administering antibiotics for a short period after broncholithectomy via cryotherapy through a flexible bronchoscope.

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