



Case report

A case of intrathoracic desmoid tumor with pulmonary *Mycobacterium abscessus* diseaseTakuma Katano^{a,*}, Eri Hagiwara^a, Hiromasa Arai^b, Midori Sato^a, Takafumi Yamaya^a, Michihiko Tajiri^b, Takashi Ogura^a^a Department of Respiratory Medicine, Kanagawa Cardiovascular and Respiratory Center, Yokohama, Japan^b Department of General Thoracic Surgery, Kanagawa Cardiovascular and Respiratory Center, Yokohama, Japan

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ABSTRACT

A 68-year-old man who was on treatment for pulmonary *Mycobacterium avium* complex complained a worsening of sputum. Although he archived negative sputum culture two months ago, sputum culture tests revealed the newly isolation of *Mycobacterium abscessus* repeatedly. Chest computed tomography showed newly-appeared extra-pulmonary mass lesion in contact with a cyst at the bottom of his right lung. From the results of contrast-enhanced magnetic resonance imaging, we first suspected loculated pleural effusion due to *Mycobacterium abscessus* infection. A thoracoscopic examination was performed as the right pneumothorax developed, and the pleural lesion was successfully resected and diagnosed as an intrathoracic desmoid tumor. Intrathoracic desmoid tumor is very rare, and this is the first report of a case with pulmonary *Mycobacterium abscessus* disease.

1. Introduction

Desmoid tumors are rare tumors accounting for less than 0.03% of all neoplasms and less than 3% of all soft tissue tumors [1]. More than half of desmoid tumors are intraabdominal, and the most frequent extra-abdominal sites include chest wall, shoulder girdle, inguinal region, and head/neck [2–4]. Intrathoracic desmoid tumors not extended from chest wall are extremely rare [5]. A case of intrathoracic desmoid tumor complicated with pulmonary *Mycobacterium abscessus* disease is here reported.

2. Case

A 68-year-old man with cough and sputum was referred to our hospital. He had 35 pack-year smoking history and a history of thoracoscopic surgery for pneumothorax of his right lung three years ago. We diagnosed him as having pulmonary *Mycobacterium avium* complex (MAC) disease with pneumothorax of his right lung. He underwent chest-tube thoracostomy and initiated chemotherapy with a three-drug regimen containing clarithromycin, rifampin and ethambutol. After two

months from initiation of three-drug regimen, his condition improved and he archived negative sputum culture. Five month later, his respiratory symptoms worsened with increased sputum during chemotherapy. Twice sputum culture tests revealed *M. abscessus* infection with high bacterial burden of 3+ smear. Chest computed tomography showed the newly-appeared extra-pulmonary mass lesion in contact with a cyst at the bottom of his right lung (Fig. 1). Contrast-enhanced magnetic resonance imaging (MRI) showed high intensity on short tau inversion recover (STIR) and diffusion weight image (DWI), slightly high intensity on T1-weight images (T1WI) (Fig. 2). From these findings and the clinical course, we assumed the newly appeared mass as loculated pleural effusion due to *M. abscessus* infection. After seven months after his first visit, he was admitted to our hospital because of a recurrence of pneumothorax of his right lung. As chest-tube thoracostomy and chemotherapy for *M. abscessus* failed to improve pneumothorax, we conducted thoracoscopy to close pulmonary fistula and to investigate the pleural mass lesions. Because leak points were not identified due to the high degree of adhesion, we performed reinforcement with polyglycolic acid felt and confirmed the cessation of air leakage. The appearance of the mass lesion was more likely a tumorous lesion rather

Abbreviations: MAC, *Mycobacterium avium* complex; MRI, magnetic resonance imaging; STIR, short tau inversion recover; DWI, diffusion weight image; T1WI, T1-weight images.

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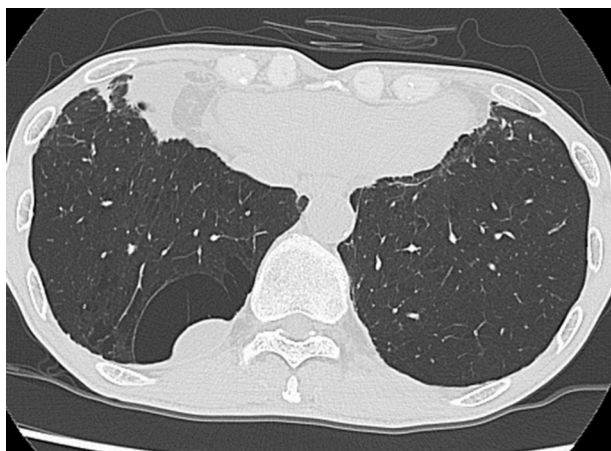


Fig. 1. Chest computed tomography imaging. Extra pulmonary mass lesion in contact with a cyst at the bottom of the right lung.

than loculated pleural effusion, and we consequently performed chest wall tumor resection at the same time (Fig. 3). The pleural mass lesion was diagnosed as desmoid tumor through histopathological examination, and the resected margin was negative. He continued to receive treatment for *M. abscessus*, and no recurrence of desmoid tumor was observed.

3. Discussion

We here reported a rare intrathoracic desmoid tumor incidentally found when the isolated bacilli were converted to *M. abscessus* during the treatment for MAC. Radiological studies alone were not enough to diagnose the tumor. Desmoid tumors do not distantly metastasize but show strong local invasiveness, and complete resection with negative margins is recommended as an appropriate therapy. In the present case, we successfully performed completely resection and no recurrence was observed. This case provides two important clinical implications. First, a rare intrathoracic desmoid tumor was incidentally found when the isolated bacilli were converted to *M. abscessus* during the treatment for MAC. Second, it was difficult to diagnose desmoid tumor from radiographic studies.

The etiology of desmoid tumors is unclear. However, association with previous trauma or surgery and molecular factors such as familial

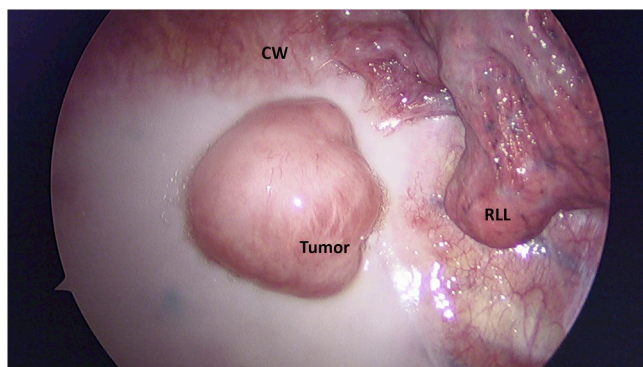


Fig. 3. Thoracoscopic imaging. Abbreviation: CW: chest wall, RLL: right lower lobe.

adenomatous polyposis have been reported [6–8]. In the present case, there was no history of trauma or familial adenomatous polyposis. However, he had a history of thoracoscopic surgery. Although the location of surgical scars was different from that of tumor, the possibility of association with thoracoscopic surgery three years ago remains. There have been no reports of intrathoracic desmoid tumor complicated with *M. abscessus*. In the present case, no pleural nor extra-pulmonary lesion was observed when MAC disease was diagnosed even with retrospective examination on CT images at his first visit. Although there was no evidence of associations between *M. abscessus* and desmoid tumor, this is the first report of desmoid tumor complicated with nontuberculous mycobacterium disease.

Most desmoid tumors are homogeneously isointense on T1WI, and heterogeneously hyperintense on T2WI or STIR images in MRI. However, MRI characteristics of desmoid tumors vary widely depending on their cellularity and fibrous content [9]. In the present case, loculated pleural effusion due to *M. abscessus* was primarily suspected because of the difficulty of diagnosing desmoid tumor by image. This case showed us again the risk of diagnosing the disease on the basis of images alone and necessity of biopsy.

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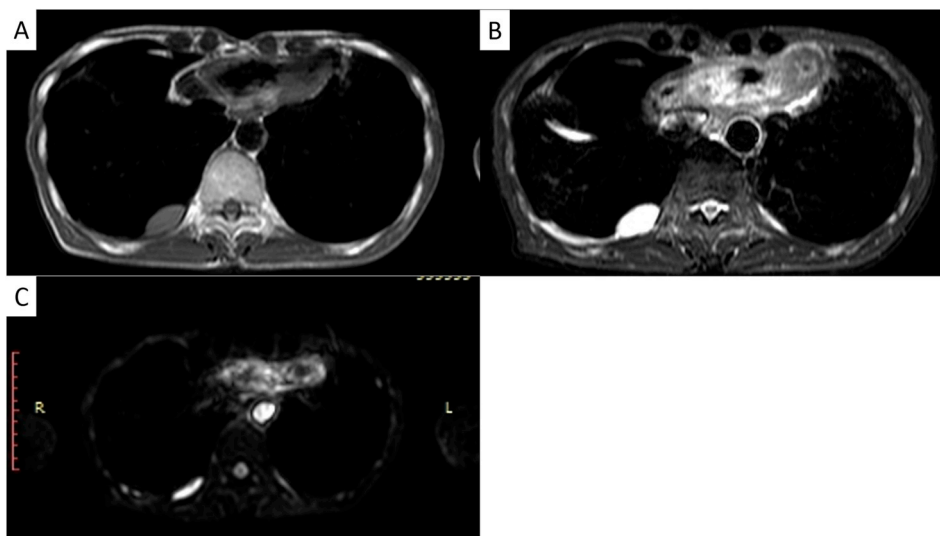


Fig. 2. Contrast-enhanced chest magnetic resonance imaging. A: slightly high intensity on T1WI, B: high intensity on STIR, C: high intensity on DWI.

Declaration of competing interest

The authors declare no Conflicts of Interest (COI) in association with this article.

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