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Case Report

Total lobar collapse caused by *mycobacterium avium* in an immunocompetent adult: a case report and review of the literature

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Abstract: Although the prevalence of nontuberculous mycobacterial pulmonary disease is increasing worldwide, endobronchial lesions caused by nontuberculous mycobacterium (NTM) are very rare in immunocompetent adults. Herein, a unique case is presented showing total collapse of the upper lobe of the left lung due to endobronchial NTM disease caused by *Mycobacterium avium*, which was finally confirmed by surgery.

Keywords: NTM pulmonary disease, mycobacterium avium, endobronchial disease

Introduction

The prevalence of pulmonary disease (PD) caused by nontuberculous mycobacteria (NTM) has been increasing worldwide [1]. NTM-PD has many clinical/radiological similarities to pulmonary tuberculosis (TB), although the bronchiectatic form of NTM-PD has a relatively distinct phenotype [2]. It is not easy to differentiate NTM-PD from pulmonary TB without microbiological examinations, especially when NTM-PD manifests in the fibrocavitary form, a solitary nodule, or accompanies a pleural effusion. Endobronchial lesions caused by *Mycobacterium tuberculosis* (M.TB) are common. Bronchoscopists, especially those who reside in countries with a high prevalence of TB, already have considerable experience and knowledge on endobronchial TB [3]. However, NTM with endobronchial involvement is extremely rare in immunocompetent hosts. This is a case report in which an immunocompetent healthy adult presented with total lobar collapse caused by *Mycobacterium avium (M. avium)* and underwent surgery with medical treatment. In addition, previous case reports are summarized.

Case presentation

A 49-year-old man visited our hospital because of an abnormality in his chest radiography that was incidentally detected during a routine medical check-up. He was an ex-smoker (40 pack-years) and had a history of treatment for pulmonary tuberculosis (TB) four years prior to presentation, which was finally cured with standard anti-TB medication. He did not have any medical diseases and was negative for human immunodeficiency virus infection. Chronic cough was his only complaint.

He had visited our clinic 15 months previously due to suspicious recurrence of TB, and chest radiography at that time showed fibronodular opacities in both upper lobes along with linear atelectasis in the left middle lung field (**Figure 1A**), which was aggravated when compared with the previous chest x-ray. Despite the abnormalities on chest radiography and history of TB, the patient refused chest computed tomography (CT) at that time. Additionally, sputum acid-fast bacilli (AFB) stain and culture were negative. In spite of the negative sputum results, his attending physician asked him to follow up for

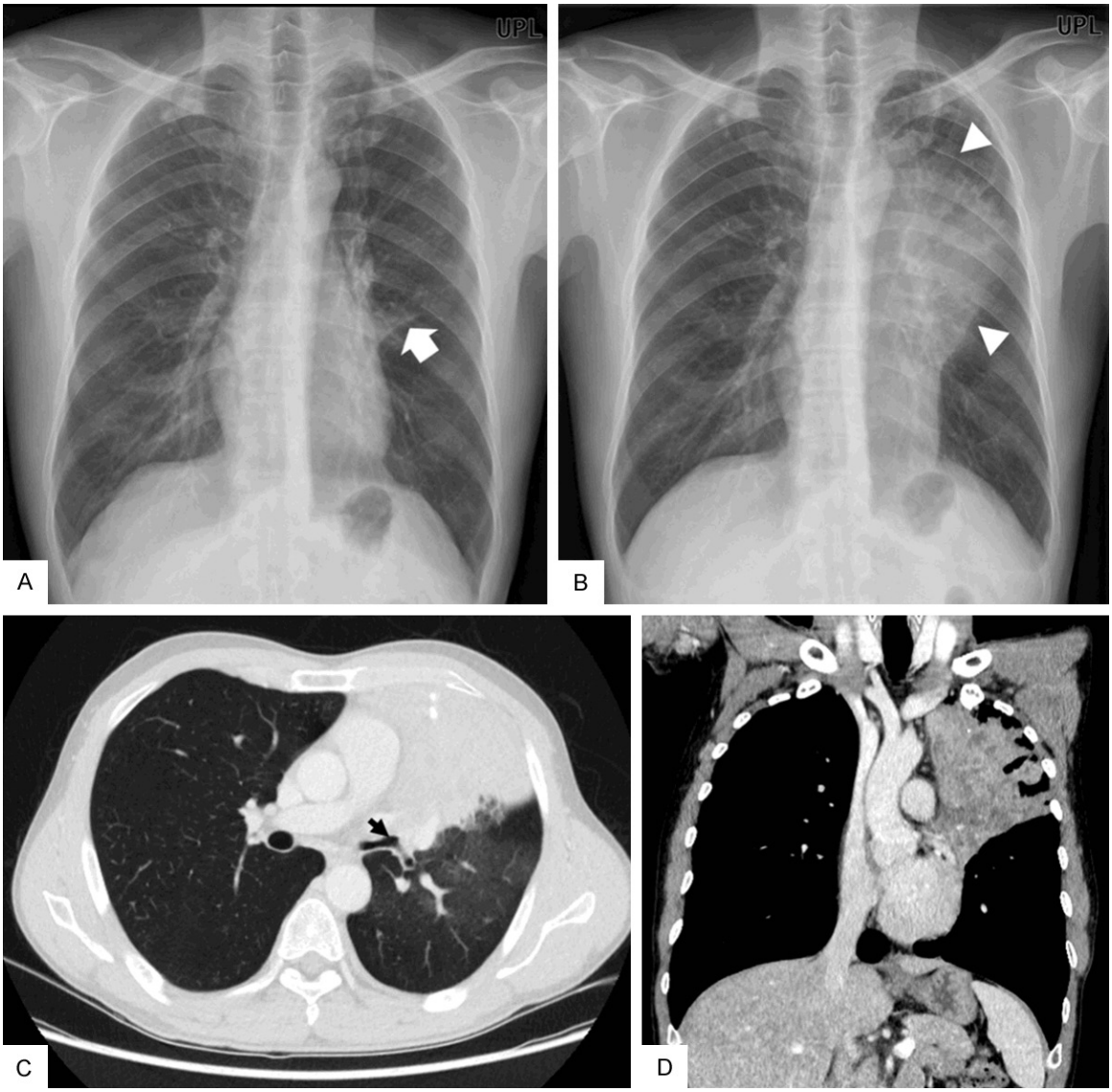

Figure 1. (A) Chest radiography obtained on the initial hospital visit revealed linear atelectasis around the left middle lung zones (white thick arrow). (B) Chest radiography and (C and D) computed tomography 15 months after the initial hospital visit demonstrated total collapse of the left upper lobe (white triangle in B) and obstruction of the left upper lobar bronchus (black thin arrow in C).

several months, but he did not visit our hospital again until the abnormal lesion was detected on his routine check-up.

His chest radiography at the present visit revealed a huge irregular consolidation or mass at the left upper lobe (**Figure 1B**). Chest CT demonstrated total obstruction of the left upper lobar bronchus with resultant atelectasis (**Figure 1C** and **1D**). Additionally, ill-defined centrilobular nodules and ground glass opacities were observed in the apicoposterior segment of the left upper lobe along with fibrotic bands with calcified nodules and cicatricial emphysema. The chest CT scans led to a suspicion of endobronchial TB or central lung cancer.

On the following day, a bronchoscopy was performed for the microbiological or histological confirmation, and total left upper lobar closure due to post-infectious sequelae was observed (**Figure 2A** and **2B**). A mucosal biopsy with bronchial washing was blindly conducted around the distal left main bronchus. AFB stain and polymerase chain reaction (PCR) for M.TB of the bronchial washing fluid were negative. The

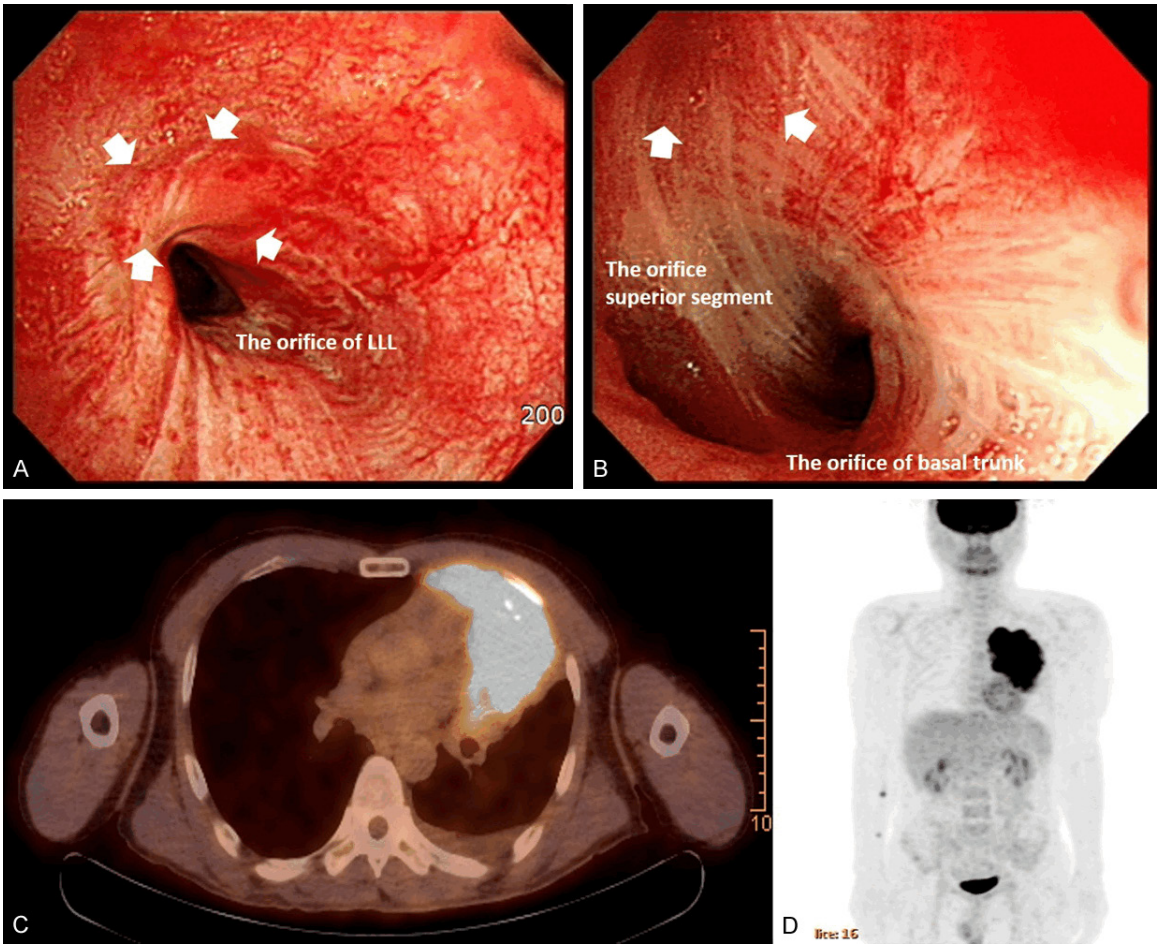

Figure 2. Bronchoscopy showed disappearance of the orifice of the left upper lobar bronchus due to post-infectious fibrosis (white arrows in A and B). Positron emission tomography demonstrated a relatively mild hypermetabolic mass (SUVmax = 3.3) in LUL central portion of the lung with heterogeneous hypermetabolic lesion (SUVmax = 20.2) involving almost the entire left upper lobe (C and D). This suggested the possibility of a low grade tumor, such as bronchial carcinoid, with total obstructive pneumonitis. Additionally, a focal hypermetabolic lymph node in the aortic-pulmonary window was observed, so the possibility of metastasis could not be excluded.

biopsy did not reveal any significant findings except for a fibrotic granulation tissue formation. PCR of the mucosal tissue for M.TB was also negative. Positron emission tomography (PET) demonstrated low grade tumor, such as bronchial carcinoid, with total obstructive pneumonitis (**Figure 2C** and **2D**). Percutaneous transthoracic needle biopsy demonstrated chronic granulation inflammation without necrosis, and the AFB stain and PCR for M.TB from the biopsy were negative. It was not possible to exclude recurrent pulmonary TB, tumors (including lung cancer), or any fungal infection.

Because the collapsed left upper lobe could no longer function properly, the left upper lobe was resected in collaboration with thoracic surgeons for the confirmative diagnosis. Fortunately, the postoperative forced expiratory volume in one second and diffusion capacity were expected to be 58% (2.06 L) and 65% (15.03 mL/mm Hg/min) of predicted, respectively, although he showed an obstructive pattern on pulmonary function tests.

A sleeve lobectomy for the upper lobe of the left lung was performed. The left upper lobar bronchus was obstructed, and multiple abscess pockets and multifocal pleural adhesions were seen, as well as segmental consolidation with fibrosis and multifocal caseous necrosis (**Figure 3A**). Microscopic findings from the resected left bronchus showed chronic granulomatous inflammation with caseous necrosis, and the tis-

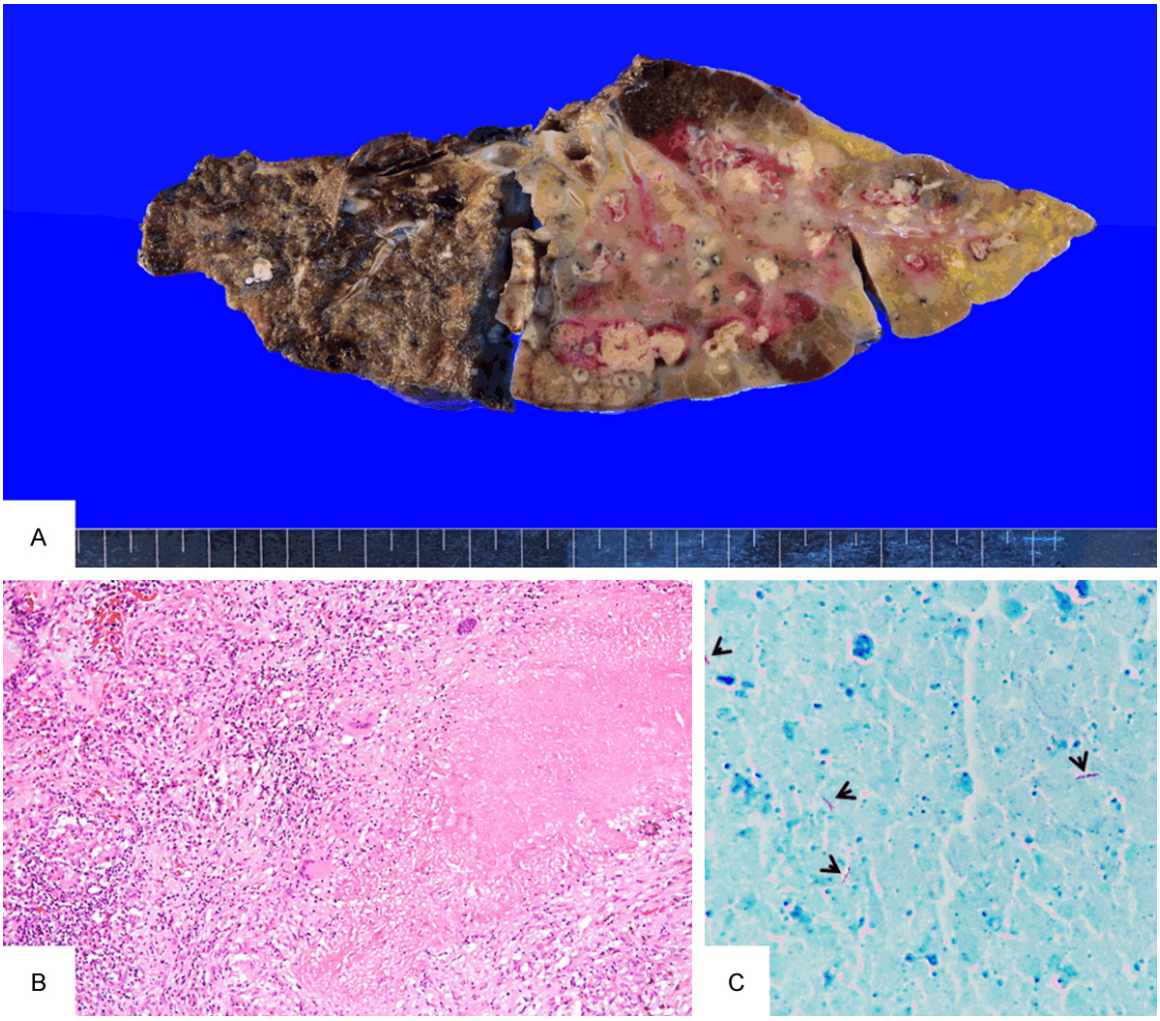

Figure 3. (A) The gross specimen showed segmental consolidation with fibrosis and multifocal caseous necrosis. (B) Microscopically, chronic granulomatous inflammation with necrosis was observed and (C) acid-fast bacilli were frequently identified (black arrow). (B, H-E, ×40; C, Ziehl-Neelsen, ×1000).

sue tested positive for AFB stain (**Figure 3B** and **3C**). However, PCR for M.TB on the surgical specimens was negative. AFB cultures in both liquid and solid media of the surgical specimens were positive, which was proved to be nontuberculous mycobacteria (NTM).

According to these findings, identification of the NTM species and drug susceptibility was expected and no anti-TB medications were given. Finally, *M. avium* was isolated. Based on this report, a triple regimen comprising oral clarithromycin (1 g per day), rifampin (600 mg per day), and ethambutol (800 mg per day) was started. Six months later, the patient continues on medication without reversion of sputum AFB stain/culture, and there has been no clinical evidence of deterioration.

Discussion

The prevalence of NTM-PD has been increasing in South Korea and all over the world. *M. avium* complex (MAC) is the most commonly identified NTM species [1, 4]. In general, patients with NTM-PD present as one of two distinct phenotypes: fibrocavitary form or nodular bronchiectatic form. These phenotypes involve primarily lung parenchyma with multiple centrilobular nodules and/or cavities. Endobronchial involvement, resulting in wheezing or lung collapse, is rare, whereas endobronchial TB is combined with other bacteria in up to 20% of patients with parenchymal involvement [5]. Cases involving immunocompetent adults seem to be even rarer. The reasons for this scarcity and difference have not been determined.

Table 1. Cases of nontuberculous mycobacteria with endobronchial involvement in immunocompetent adults

Author	Region	Age/Sex	Species	Bronchoscopy findings	Treatment	Outcome
M. Connolly et al. (1993) [11]	USA	52/F	*M. kansasii*	Polypoid lesion arising from posterior segment of right upper lobe causing complete obstruction	Rifampin + isoniazid + ethambutol (poorly compliant with medication)	Small endobronchial lesion obstructing posterior segment of right upper lobe at time of follow-up bronchoscopy 4 year later, chronic right upper lobe cavity with retraction of the right hilum
K. Fukuka et al. (2003) [12]	Japan	57/M	*M. avium*	Occlusion of the apical segmental bronchus of the left upper lobe by a white-coated polypoid lesion	Rifampin + isoniazid + ethambutol + sparfloxacin + clarithromycin	Resolution of endobronchial polypoid lesions after 1 year of medical treatment
E. Manali et al. (2005) [13]	USA	58/M	*M. kansasii*	Bilateral concentric narrowing of the main stem bronchi and truncus intermedius with ulceration	Rifampin + isoniazid + ethambutol laser resection and balloon bronchoplasty	No reversion of bronchial cultures for AFB 1 year after treatment completion
S-H Kang et al. (2013) [14]	South Korea	59/F	*M. avium*	Whitish- and yellow-colored irregular mucosal plaques in the lingular division	Clarithromycin + ethambutol + rifampin	Resolution of respiratory symptoms and decreased infiltration on the left lower lobe after two months of treatment
H. Kim et al. (2015) [15]	South Korea	37/F	*M. avium*	Ulcerative lesion at the medial side of the right main bronchus, bronchial stricture of the right intermedius bronchus and lower lobar bronchus	Azithromycin + ethambutol + rifampin	Improved atelectasis of right lower lobe and no recurrence 12 months after treatment completion
Present case (2019)	South Korea	49/M	*M. avium*	Fibrinous scar change and loss of the opening of the left upper bronchus with total collapse of LUL	Surgical resection of left upper lobe followed by clarithromycin + ethambutol + rifampin	No reversion of sputum culture 6 months after treatment

A literature search revealed that most cases of NTM with endobronchial involvement develop in immunocompromised adults (28 cases) [6, 7] or very young pediatric populations (eight cases less than five years old) [8-10]. There have been only five cases of endobronchial NTM disease in immunocompetent adults [11-15]. **Table 1** shows the previous cases and the present one in summary. Interestingly, only *M. avium* among MAC has been identified from endobronchial lesions.

In general, MAC lung disease with the nodular bronchiectatic form, not the fibrocavitary form, does not required prompt treatment in immunocompetent hosts. Physicians need to primarily follow the course of clinical manifestation, bacterial burden, and radiological changes prior to combination antimicrobial therapy. This case presented initially as linear atelectasis of the left upper lobe when he first visited our clinic. Afterwards, he was lost to follow-up without further evaluation or treatment. When he finally revisited our clinic, his chest radiography showed total collapse of the left upper lung. Bronchoscopy showed only fibrosis of the orifice in the left upper lobe, and we could not find the left upper lobar bronchus. This indicates that endobronchial NTM disease can cause bronchial stricture or total obstruction if there is no treatment or intervention. Therefore, if NTM disease has endobronchial involvement, rapid treatment should be administered irrespective of the degree or pattern of parenchymal involvement. Delay in diagnosis and treatment can progress to bronchial stenosis or obstruction. Most previous cases reported a polypoid or ulcerative lesion with varying degrees of bronchus obstruction that improved with treatment.

Although previous case reports and the present case in immunocompetent adults demonstrated radiological and bronchoscopic improvement as well as bacterial conversion with long-term combination antibiotics (**Table 1**), there is insufficient evidence about how endobronchial NTM disease should be managed (optimal treatment duration and methods of local intervention using bronchoscopy). Unfortunately, uncertainties regarding management are not easily answered because NTM with endobronchial involvement is extremely rare.

In conclusion, this is a report of a rare and unique case of endobronchial NTM disease due to *M. avium* in an immunocompetent healthy adult. Endobronchial NTM involvement should be considered when an endobronchial lesion is discovered during bronchoscopy, even in immunocompetent adult patients.

Disclosure of conflict of interest

None.

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References

[1] Kendall BA and Winthrop KL. Update on the epidemiology of pulmonary nontuberculous mycobacterial infections. Semin Respir Crit Care Med 2013; 34: 87-94.

[2] Goo JM and Im JG. CT of tuberculosis and nontuberculous mycobacterial infections. Radiol Clin North Am 2002; 40: 73-87, viii.

[3] Siow WT and Lee P. Tracheobronchial tuberculosis: a clinical review. J Thorac Dis 2017; 9: E71-E77.

[4] Yoon HJ, Choi HY and Ki M. Nontuberculosis mycobacterial infections at a specialized tuberculosis treatment centre in the Republic of Korea. BMC Infect Dis 2017; 17: 432.

[5] Lee JY, Yi CA, Kim TS, Kim H, Kim J, Han J, Kwon OJ, Lee KS and Chung MJ. CT scan features as predictors of patient outcome after bronchial intervention in endobronchial TB. Chest 2010; 138: 380-385.

[6] Salama C, Policar M and Venkataraman M. Isolated pulmonary mycobacterium avium complex infection in patients with human immunodeficiency virus infection: case reports and literature review. Clin Infect Dis 2003; 37: e35-40.

[7] Kim HC, Bae IG, Ma JE, Lee JS, Jeon KN, Lee JD and Hwang YS. Mycobacterium avium complex infection presenting as an endobronchial mass in a patient with acquired immune deficiency syndrome. Korean J Intern Med 2007; 22: 215-219.

[8] Kroner C, Griese M, Kappler M, Schoen C, Hoffmann F, Nicolai T and Reiter K. Endobronchial lesions caused by nontuberculous mycobacteria in apparently healthy pediatric patients. Pediatr Infect Dis J 2015; 34: 532-535.

[9] Naik V, Iroh Tam P, Gershan W, Colin AA and Demirel N. Pulmonary mycobacterium avium-intracellulare complex infection in an infant: a

silent and coincidental finding. Pediatr Allergy Immunol Pulmonol 2017; 30: 257-259.
[10] Cohen L, Guarner J and Hunt WR. A novel presentation of mycobacterium avium complex in a recipient of a lung transplant: a case report. J Med Case Rep 2017; 11: 240.
[11] Connolly MG Jr, Baughman RP and Dohn MN. Mycobacterium kansasii presenting as an endobronchial lesion. Am Rev Respir Dis 1993; 148: 1405-1407.
[12] Fukuoka K, Nakano Y, Nakajima A, Hontsu S and Kimura H. Endobronchial lesions involved in Mycobacterium avium infection. Respir Med 2003; 97: 1261-1264.
[13] Manali ED, Tomford WJ, Liao DW, Farver C and Mehta AC. Mycobacterium kansasii endobronchial ulcer in a nonimmunocompromised patient. Respiration 2005; 72: 305-308.
[14] Kang SH, Mun SK, Lee MJ, Kim SY, Choi HG, Byun J, Kim CH, Kim HR and Cho SY. Endobronchial mycobacterium avium infection in an immunocompetent patient. Infect Chemother 2013; 45: 99-104.
[15] Kim HI, Kim JW, Kim JY, Kim YN, Kim JH, Jeong BH, Chung MJ and Koh WJ. Isolated endobronchial mycobacterium avium disease associated with lobar atelectasis in an immunocompetent young adult: a case report and literature review. Tuberc Respir Dis (Seoul) 2015; 78: 412-415.