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A Rare Cause of Solitary Pulmonary Nodule: Broncholithiasis: Case Report

Soliter Pulmoner Nodülün Nadir Bir Nedeni: Bronkolitiyaz

ABSTRACT Broncholithiasis is defined as a condition in which calcified or ossified lesion is present within the bronchial lumen. The most common causes of broncholithiasis are tuberculosis or fungal infections. A 60 year-old male patient with solitary pulmonary nodule and chronic obstructive pulmonary disease was admitted to our clinic due to dyspnea and non-productive cough complaints. Here we report this case because of a very rare lesion broncholith was found in tracheobronchial tree and removed with forceps using flexible bronchoscopy without any complication.

Key Words: Bronchoscopy; tuberculosis, lymph node

ÖZET Bronkolitiyazis bronşiyal lümen içinde kalsifiye ya da kemiksi lezyonun görülmesine neden olan bir durum olarak tanımlanır. En sık neden tüberküloz ya da fungal enfeksiyonlardır. Soliter pulmoner nodül ve kronik obstrüktif akciğer hastalığı tanıları olan 60 yaşında erkek hasta kliniğimize kuru öksürük ve nefes darlığı şikayetleri ile başvurdu. Bu olguyu nadir bir lezyon olan bronkolitin trakeobronşiyal ağaçta bulunması ve fleksibl bronkoskopi aracılığıyla forseps kullanılarak komplikasyonsuz çıkartılması nedeniyle sunuyoruz.

Anahtar Kelimeler: Bronkoskopi; tüberküloz, lenf düğümü

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Broncholithiasis is a rarely seen entity characterized by the presence of calcified or ossified material in the lumen of the tracheobronchial tree.¹ In general, it is formed as a result of calcified lymph node eroding the bronchial wall and falling in the lumen.² The most common cause of broncholithiasis is erosion by and extrusion of a calcified adjacent lymph node into the bronchial lumen. Disease is usually associated with mycobacterial and fungal infections. Aspiration of bone tissue, in situ calcification of aspirated foreign material and erosion by and extrusion of calcified or ossified bronchial cartilage plates are the responsible mechanism of broncholith formation.¹⁻⁴ Bronchoscopy is the most important method in the diagnosis and treatment of this disease.⁵ We present a case with broncholithiasis which was diagnosed and treated by flexible bronchoscopy.

CASE REPORT

A 60 year-old man was admitted to our clinic due to dyspnea and non-productive cough for three years. He has been followed due to solitary pulmonary nodule in his thorax by computerized tomography (CT) and the first tomography was performed three months ago. His thorax CT showed partly emphysema areas in the upper lobes including calcified granular fibrotic changes in the anterior segment of the left upper lobe and tubular bronchiectatic changes in the lower lobes as well as 13 mm solitary pulmonary nodule in the right upper lobe (Figure 1a and 1b). Positron emission tomography indicated no 18-fluorodeoxyglucose uptake. He had 40 packs-years smoking history and no history of contact with an active tuberculous patient. He also had no history of a foreign body or food aspiration. Physical examination was normal except for a decrease in the sound of breaths in both hemithorax. Control thorax CT findings were similar with prior CT. His laboratory findings were as follows: white blood cell count of 6.31 K/µL, haemoglobin count of 13.6 g/dL, platelet count of 246 K/uL. Erythrocyte sedimentation rate was 44 mm/hour and the C-reactive protein was 0.927 mg/dL. Biochemical examinations were: blood urea nitrogen 26 mg/dL, creatinine 0.62 mg/dL, glucose 93 mg/dl, aspartate aminotransferase 14 U/L, alanine aminotransferase 10 U/L. Pulmonary function test revealed FVC 88%, FEV₁ 71%, ratio of FEV₁ to FVC 65. Bronchoscopic evaluation revealed nearly 5 mm in size, white-colored, rough lesion located in the anterior segment of the left upper lobe (Figure 2a). Additionally, changes in bronchial mucosa was compatible with chronic bronchitis. A small amount of serous secretion was aspirated. The changes in bronchoscopic evaluation suggested that they were compatible with previous tuberculosis disease. The lesion was removed with crocodile forceps via flexible bronchoscopy (Figure 2b). Pathologic examination displayed calcified necrotic material and psammoma body like structures (Figure 3). An informed consent form was obtained from the patient.

DISCUSSION

We are reporting an uncomplicated case with posttuberculosis broncholith which is successfully removed by the flexible bronchoscopy. The most common cause of broncholithiasis is peribronchial calcified lymph nodes known to be a result of tuberculosis.^{1,3,4} In addition, bronchial obstruction due to aspiration of food particles may cause bronchial cartilage calcification and this may result in broncholithiasis. Rarely, silicosis and fungal infections such as histoplasmosis, nocardiosis, actinomycosis, coccidioidomycosis, cryptococcosis may also cause broncholithiasis.^{1,2} Chemical structure of broncholith is very similar to the bone and it contains 85-90% calcium phosphate and 10-15% calcium carbonate.⁶ Although the patient does not have tuberculosis in his medical history, his thorax CT findings were similar to tuberculosis sequelae.

Broncholithiasis is encountered in men and women in the same frequency, mostly in fifth or sixth decades and reported to occur generally in the right bronchus.⁷ In the case we report here, it was



FIGURE 1a: Peribronchial calcified lesion is seen in the left upper lobe (mediastinal window).



FIGURE 1b: Some fibrotic changes in the anterior segment of the left upper lobe the calcified lesion are seen in the left upper lobe (parenchyma window).



FIGURE 2a: A white-coloured and irregular shaped lesion is seen in the anterior segment of the left upper lobe.

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FIGURE 2b: The lesion is removing by crocodile forceps at the level of main carina.

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observed to be in the left bronchus. A broncholith seldom causes symptoms, as well as chronic nonproductive cough, dyspnea, fever, hemoptysis, chest pain, stone expectoration (lithoptysis). Lithoptysis is a pathognomonic symptom and rarely seen. Findings of physical or radiological examination are non-specific. Localized wheezing is reported in several cases.⁵ Radiological imaging techniques to diagnose bronholithiasis are commonly difficult. It is generally defined as a parenchymal or peribronchial calcified nodules or lymph nodes. Rarely, endobronchial obstruction findings such as air-trapping, atelectasis may be seen.^{1,8} An awareness of the typical imaging findings of broncholithiasis may help in establishing an accurate diagnosis to ensure proper case management.¹ In our case, the patient suffered from only dyspnea and cough. His symptoms, physical examination and some radiological findings were not attributable to a smoking related disease. Parenchymal calcification in the anterior segment of left upper lobe and solitary pulmonary nodule in the right upper lobe seen in his thorax CT may be related with healed primary tuberculosis.

Broncholithiasis can be diagnosed with thorax CT and bronchoscopic evaluation and should be considered for the diagnosis, if the appropriate clinical and radiological findings are obtained.^{1,6} In our case, bronchoscopic examination was done to investigate the etiology of SPN, but surprisingly a broncholith was seen.

In untreated patients some complications recurrent pneumonia, bronchial artery fistula, bronchoesophageal fistula and aortotracheal fistula may occur.^{9,10} In untreated patients, some complications may occur such as recurrent pneumonia, bronchial artery fistula, bronchoesophageal fistula and aortotracheal fistula.^{9,10} It is thought that development of hemoptysis is because of the damage of bronchial vessels with broncholith and sometimes it can be fatal.^{4,11}



FIGURE 3: Photograph of the removal lesion shows calcium crystalloid and psammoma body like structures as microscopically (May-Grünwald-Giemsa stain, x400).

(See color figure at http://akcigerarsivi.turkiyeklinikleri.com/)

Bronchoscopic techniques are recommended for the uncomplicated cases similar to our case. Especially loose and movable broncholiths are suitable for removal by bronchoscopic extraction.¹² Several authors suggested the removal of broncholith with rigid bronchoscopy. The success rate with flexible bronchoscopy is 30% and hemorrhage or central airway obstruction is rarely seen during the process. However, the success rate of rigid bronchoscopy is 67-87%. Nd YAG laser or holmium laser can be used to comminute larger broncholith.^{13,14} Surgical methods may be necessary in complicated cases which had hemoptysis arising from broncholithiasis-induced bronchial artery rupture or fistula or pulmonary artery fistula, recurrent obstructive pneumonia, secondary bronchiectasis or bronchoesophageal fistula.¹⁵⁻¹⁸ Proposed surgery methods are segmentectomy, lobectomy or pneumonectomy.^{4,11}

In the differential diagnosis, primary endobronchial infections with dystrophic calcification, calcified endobronchial tumors, tracheobronchial diseases with mural calcification, and hypertrophied bronchial artery with intramural protrusion should be kept in mind.

CONCLUSION

Broncholithiasis should be considered in the diagnosis of patients who had previous tuberculosis and/or calcified lesion adjacent to bronchus in thorax CT. Flexible bronchoscopy should be performed for diagnostic evaluation and may be worth to do bronchoscopic broncholith removal in uncomplicated cases. Rigid bronchoscopy or surgical methods may be required for complicated cases.

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