CASE REPORT

Broncholithiasis presenting as bronchiectasis and recurrent pneumonias

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SUMMARY

A broncholith is defined as the presence of calcified material within a bronchus or within a cavity communicating with a bronchus. It is most frequently caused by *Histoplasmosis* or tuberculosis (TB) spp. Bronchial distortion, irritation and erosion by broncholiths can cause bronchiectasis, recurrent pneumonias and haemoptysis. We present a case of recurrent pneumonia due to a broncholith, which resolved conservatively with antibiotics. Owing to recurrent fevers and post obstructive pneumonias, a lobectomy or rigid bronchoscopic removal were considered but the patient was deemed not to be a candidate for general anaesthesia due to her comorbidities. Broncholiths are an uncommon cause of bronchiectasis and recurrent pneumonias. However, the wide range of symptoms and low clinical suspicion are the main reasons why a diagnosis can be delayed. Various treatment options are available and the choice of therapy should be made depending on the broncholith's size, mobility, location and local surgical expertise.

BACKGROUND

A broncholith is defined as the presence of calcified material within a bronchus or within a cavity communicating with a bronchus.^{1 2} More specifically, they are calcified peribronchial lymph nodes that erode into the adjacent airway subsequent to an inflammatory process, most frequently *Histoplasmosis* or tuberculosis (TB) spp.^{1 3} Patients become symptomatic when the broncholith impinges on or erodes into the lumen of the airway. Bronchial distortion, irritation and erosion by broncholiths can cause bronchiectasis, cough, recurrent pneumonias, haemoptysis and dyspnoea. Life-threatening complications, such as massive haemoptysis or broncho-oesophageal fistulas, can also occur.^{1 4} Given the wide range of symptoms, broncholiths may not be discovered for a prolonged period of time. We present a case of broncholithiasis presenting as fever of unknown origin, bronchiectasis and recurrent pneumonia. This case is important because it serves to raise the awareness of the multiple complications of broncholithiasis, in particular, recurrent pneumonia.

CASE PRESENTATION

A 61-year-old Hispanic woman with a medical history of hypertension and an implantable cardiac defibrillator (ICD) for hypertrophic obstructive cardiomyopathy, and a social history negative for smoking, came to the emergency department with a 1-day history of fever, shortness of breath and pleuritic chest pain as well as one episode of cough with blood streaked sputum. The patient

had prior admissions within the last 18 months for fever of unknown origin. The last admission was 8 weeks prior, during which time she had persistent fevers with maximal temperature (Tmax) of 101.7F. An extensive infectious evaluation was performed including respiratory cultures, blood cultures, stool cultures, TB and fungal cultures, which were all negative. A plain chest radiograph (CXR) did show a left upper lobe (LUL) homogenous opacity but no chest CT was performed during that admission. However, CT of the abdomen and pelvis with contrast did reveal ground glass opacities of the lung bases with a small left pleural effusion. The patient was discharged after 7 days with broad-spectrum antibiotic coverage for a diagnosis of healthcare-associated pneumonia based on abdominal CT findings. On this admission, her vital signs revealed Tmax 100.7F, blood pressure 172/75 mm Hg, pulse 101 bpm, oxygen saturation of 94% on room air and respiratory rate of 16 breaths/min. The rest of the physical examination was normal except for bibasilar crackles. CXR posteroanterior and lateral views showed cardiomegaly, bilateral hilar calcifications and an increased LUL density partially obscured by the patient's ICD ([figure 1](#)). Owing to haemoptysis, a CT angiogram (CTA) of the chest was performed for further evaluation. CTA of the chest showed a collapse of a segment of the LUL with distal bronchiectatic changes as well as proximal calcified lymph nodes in the superior aspect of the left hilum ([figure 2](#)). Owing to these findings, a fiberoptic bronchoscopy was performed and confirmed a partially mobile endobronchial broncholith with complete occlusion of the lumen of the LUL bronchus on expiration but subtotal occlusion on end inspiration. The extraction of the broncholith was not performed because it was felt that attempted removal without airway control incurred too much risk of bleeding due to the broncholith's association with the LUL pulmonary artery. Bronchial alveolar lavage (BAL) and washings were performed, and showed reactive epithelial and squamous cells. BAL and respiratory cultures were negative including for Acid-fast bacilli. The patient was started on treatment for post obstructive pneumonia with vancomycin and piperacillin–tazobactam for a 21-day course. Her fevers and shortness of breath improved.

INVESTIGATIONS

CTA of the chest showed collapse of a segment of the LUL with distal bronchiectatic changes as well

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Figure 1 Chest radiograph posteroanterior and lateral views showing cardiomegaly, bilateral hilar calcifications and an increased left upper lobe density partially obscured by the patient's implantable cardiac defibrillator.

as proximal calcified lymph nodes in the superior aspect of the left hilum.

Fibreoptic bronchoscopy confirmed a partially mobile endobronchial broncholith with complete occlusion of the lumen of the LUL bronchus on expiration but subtotal occlusion on end inspiration. BAL and washings showed reactive epithelial and squamous cells.

BAL and respiratory cultures, and Gram stain, were negative including for Acid-fast bacilli.

DIFFERENTIAL DIAGNOSIS

- ▶ Malignancy
- ▶ TB
- ▶ Fungal pathogens
- ▶ Foreign body aspiration
- ▶ Bronchial stenosis or atresia
- ▶ Localised bronchiectasis

Figure 2 CT angiography of the chest (axial view) showing collapse of a segment of the left upper lobe with distal bronchiectatic changes (top row) as well as proximal calcified lymph nodes in the superior aspect of the left hilum (bottom row).

TREATMENT

- ▶ Antibiotic treatment with vancomycin and piperacillin–tazobactam for 21 days
- ▶ Assessment for rigid bronchoscopy as a therapeutic option

OUTCOME AND FOLLOW-UP

The patient was seen 2 months later in the outpatient pulmonary clinic and resolution of symptoms were noted. Owing to recurrent fevers and recurrent post obstructive pneumonias, a lobectomy or rigid bronchoscopic removal were considered but the patient was deemed not to be a candidate for general anaesthesia for the removal of the broncholith due to the severity of her hypertrophic obstructive cardiomyopathy. The patient continued to be asymptomatic 7 months later.

DISCUSSION

Symptoms caused by broncholithiasis can be very difficult to diagnose given the wide variety of symptoms and because of

low clinical suspicion. Furthermore, broncholiths may not always be seen on CXR. However, in patients who present with recurrent pneumonias, further investigation should be performed to rule out post obstructive pneumonia secondary to other causes and to confirm the obstruction is due to the broncholith.

There are two types of broncholiths, fixed and mobile. Mobile broncholiths are either loose in the airway or move when probed with bronchoscopic instruments during bronchoscopy. Fixed broncholiths do not exhibit any movement when probed during bronchoscopy.⁵

Broncholiths most commonly develop secondary to TB and *Histoplasmosis* infections.⁶ TB is most commonly seen in Korea and Europe, and *Histoplasmosis* in the USA. Other possible aetiologies include other infectious aetiologies such as *Cryptococcus*, coccidioidomycosis, actinomycosis, aspergillosis, nocardiosis and fungal infections as well as lung abscess, pulmonary infarction, silicosis and inhalation of foreign materials.⁷ Clinical manifestations of broncholithiasis range from asymptomatic-to-mild coughing, sputum production, haemoptysis, lithoptysis, chest pain, fever, bronchiectasis, atelectasis and broncho-oesophageal fistula.^{7,8}

Three mechanisms have been described in the formation of a broncholith. First, inhaled materials in the bronchus mix with calcium and develop into a broncholith. Second, bronchial cartilage is either calcified or ossified, which then breaks apart in the endobronchial space. In the third and most common aetiology, a lung parenchymal infection causes lymphadenitis leading to calcium deposition in the lymph nodes under an alkaline environment.⁸

A diagnosis of broncholithiasis is suggested strongly by the following CT findings: a calcified lymph node that is either endobronchial or peribronchial, the presence of bronchiectasis and the absence of an associated soft tissue mass.⁹ Most cases of bronchiectasis from broncholithiasis are acquired from either endobronchial obstruction or external compression and may result in recurrent bacterial bronchopneumonia. The specific nature of the infection is not as important as the bronchial obstruction leading to prolonged infection and delayed resolution. Isolated bronchiectasis, defined as permanent dilation of the bronchi with destruction of the bronchial wall, is generally associated with prior bronchial inflammation or infections. However, bronchiectasis can occur by bronchial obstruction from a variety of sources including a broncholith. Gross examination of bronchiectatic lungs show dilation of the bronchi, which are filled with suppurative yellow-green secretions. The bronchial mucosa is often oedematous, ulcerated or necrotic. Microscopically, bronchiectasis is characterised by the loss of ciliated epithelium, mucoelastic tissue and, occasionally, cartilage. The scarring of the bronchial wall will result in contraction of the peribronchial tissues, creating further bronchial dilation.¹⁰

Although central airway obstruction is most commonly due to malignancy, it is also a complication of benign obstructive lesions such as broncholiths leading to post obstructive pneumonia. As seen our case, bronchial obstruction secondary to broncholithiasis was the cause of recurrent pneumonias despite appropriate antibiotic treatment. Most post obstructive pneumonias are polymicrobial in nature. In fact, as much as 30–55% of the aspirates obtained are found to be polymicrobial, which explains why our patient's respiratory and BAL cultures were not diagnostic. The most common organisms isolated are *Haemophilus influenza*, *Klebsiella pneumonia*, *Enterobacter cloacae*, *Acinetobacter* spp, *Pseudomonas aeruginosa* and *Staphylococcus aureus*.¹⁰

When broncholithiasis is suspected, CT is the diagnostic modality of choice. The advantage of CT is that it not only detects the existence of a broncholith but it also identifies its location in relation to the bronchial wall.⁷ Bronchoscopy should be performed thereafter to exclude the presence of an endobronchial neoplasm and to confirm the presence of a broncholith.^{11–12} Direct bronchoscopic visualisation is diagnostic. However, a broncholith is only visualised in 28–44% of cases.⁷

Therapeutic options should be determined by mass size, mobility, location and patient's symptoms. Conservative treatment is adequate in the majority of cases.⁸ The conventional method to remove a broncholith is performed with the use of forceps during bronchoscopy.

However, this is only applicable for broncholiths that are not firmly attached to the bronchial wall.⁸ Hesitation in removing a broncholith is secondary to fear of massive haemorrhage and bronchial injury.^{5,13} When a broncholith is not amenable to removal by bronchoscopy alone, laser therapy has been used to fragment mobile broncholiths that would otherwise be too hard or large to be removed by bronchoscopy.⁶ This method can also induce life-threatening haemoptysis if the broncholith is adjacent to a vessel.⁸ Lastly, surgical interventions, such as lobectomy, segmentectomy or pneumonectomy, can be performed but are rarely needed.⁸ Recurrent and persistent haemoptysis, bronchiectasis, bronchial stenosis, development of broncho-oesophageal or aortotracheal fistula and obstructive pneumonia are indications for surgery. In addition, if the broncholith is firmly attached to the bronchial wall or if bronchoscopic removal is not possible, surgical intervention is indicated.^{1,7,8} Although our patient was not a surgical candidate, conservative management has been successful. She has been asymptomatic for 1 year.

In conclusion, broncholiths are an uncommon cause of bronchiectasis and recurrent pneumonias. However, the wide range of symptoms and the low clinical suspicion are the main reasons why a diagnosis can be delayed. Various treatment options are available and the choice of therapy should be made depending on the expertise of the physicians in the available facility.

Learning points

- ▶ Persistent or recurrent pneumonia should prompt more invasive investigation.
- ▶ Presence of lobar bronchiectasis on CT is concerning for bronchial obstruction.
- ▶ Broncholiths may present with a wide variety of symptoms and signs including bronchiectasis, which may delay the diagnosis.
- ▶ Therapeutic options are determined by mass size, mobility and location of the broncholith.
- ▶ Referral to expert cardiothoracic surgeons must be made for endobronchial manipulation of complicated broncholiths.

Competing interests None declared.

Patient consent Obtained.

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Reminder of important clinical lesson

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