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Recurrent Bronchogenic Cyst After Surgical Resection

Abdul Hamid Alraiyes, MD,[1] Khaldoon Shaheen, MD,[2] Jordan Reynolds, MD,[3] Michael Machuzak, MD[4]

[1]Interventional Pulmonology, Department of Medicine, Roswell Park Cancer Institute and University at Buffalo, State University of New York, Buffalo, NY [2]Department of Hospital Medicine, Cleveland Clinic Foundation, Cleveland, OH [3]Pathology and Laboratory Medicine Institute, Cleveland Clinic Foundation, Cleveland, OH [4]Department of Pulmonary, Allergy, and Critical Care Medicine, Cleveland Clinic Foundation, Cleveland, OH

Background: Bronchogenic cysts are rare congenital anomalies that are often solitary and rarely multiple. Most bronchogenic cysts are asymptomatic, and symptoms when present are usually the result of compression by the cyst on the surrounding structures.

Case Report: We report a case of recurrent bronchogenic cyst following a partial resection treated with endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA).

Conclusion: EBUS-TBNA can provide instant decompression of the cyst and relieves the pressure on the surrounding structures.

Keywords: *Bronchogenic cyst, bronchoscopy, endoscopic ultrasound-guided fine needle aspiration*

Address correspondence to Abdul Hamid Alraiyes, MD, Assistant Professor of Oncology, Interventional Pulmonology, Department of Medicine, Roswell Park Cancer Institute, Elm and Carlton Streets, Buffalo, NY 14263. Tel: (716) 845-8153. Email: abdul.alraiyes@roswellpark.org

INTRODUCTION

Bronchogenic cysts are rare congenital anomalies that are often solitary and rarely multiple. Most bronchogenic cysts are asymptomatic, and symptoms when present are usually the result of compression by the cyst on the surrounding structures. The standard approach to symptomatic bronchogenic cysts has been surgical resection, but endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA) may provide an alternate treatment strategy.

CASE REPORT

A 67-year-old man presented with a 1-month history of chest pain and dyspnea on exertion. The pain was in the middle of the chest and dull in nature without radiation or aggravating or relieving factors. Symptoms progressively worsened, limiting his daily activities. Following the progression of these symptoms, the patient reported a sudden episode of dizziness with sharp chest pain and palpitation while playing golf. He presented to the emergency department with stable atrial fibrillation and was admitted to the intensive care unit for rate control. He denied fever, night sweats, cough, or hemoptysis. He had never smoked tobacco and denied alcohol or illicit drug abuse. His history was significant for hypertension, atrial fibrillation, and a bronchogenic cyst. Prior to admission, he was taking valsartan, hydrochlorothiazide, and aspirin. Eight years prior, he had reported similar symptoms that were ultimately attributed to a bronchogenic cyst that was compressing the right pulmonary artery. The cyst was partially resected at that time. Since then, it had been followed by periodic surveillance computed tomography (CT) imaging of the chest. A recurrence of the bronchogenic cyst was identified 1 year prior to this presentation (7 years postresection) that was growing in size but had no symptoms until the current presentation.

During examination, the patient was not in respiratory distress. His vital signs were significant for an irregularly irregular heart rate with 85 bpm, and the rest of his vital signs were unremarkable with a respiratory rate of 16 breaths per minute and 95% oxygen saturation on room air. He had good air entry into both lungs with no added sounds. The rest of the examination was unremarkable. Laboratory findings revealed a normal complete blood count and basic metabolic profile. Electrocardiogram (EKG) confirmed the atrial fibrillation with a rate of 85 bpm. Chest x-ray revealed no acute cardiopulmonary process and no infiltrates. Chest CT scan was significant for a large, homogeneous, low-density lesion consistent with recurrent bronchogenic cyst. Compared to the CT scan done 6 months prior, the cyst had grown in size (Figure 1) and was large enough to cause severe narrowing of the right main pulmonary artery. A repeat surgical procedure was considered to be complicated, so the interventional pulmonary team was consulted to evaluate the bronchogenic cyst for possible transbronchial drainage.

A bronchoscopy was performed, and the cyst was identified using EBUS. Cyst drainage was achieved with EBUS-TBNA (Figure 2), and 56 mL of a white cloudy fluid was drained from the cyst. The cyst wall was then sampled. Rapid onsite cytology evaluation demonstrated benign epithelial cells, blood, and foamy macrophages, suggestive of cyst contents. No malignant cells were identified (Figure

Figure 1. Chest computed tomography shows a large, homogeneous, relatively low-density lesion consistent with recurrent bronchogenic cyst. We observed mass effect from the lesion and very severe narrowing of the right pulmonary artery (arrow).

3). The procedure was completed without complications. Fluid microbiologic culture revealed no growth, and the final cytology report was negative for malignancy. The patient's symptoms improved after the procedure, the follow-up EKG showed sinus rhythm, and he was discharged home. At 3-month follow-up, surveillance CT scan of the chest showed significant shrinking of the bronchogenic cyst with relief of the right pulmonary artery compression (Figure 4).

DISCUSSION

Bronchogenic cysts are rare congenital anomalies that arise in early gestation as a result of abnormal budding of the developing respiratory system.[1] Abnormal bronchi or bronchioles may form large saccular structures that may later form cystic lesions. Bronchogenic cysts predominantly occur in males and are often solitary and rarely multiple. Typically, bronchogenic cysts have ciliated epithelium with cartilage, smooth muscles, and mucus-producing bronchial glands similar to the bronchial walls. Bronchogenic cysts do not have alveolar structures because the abnormal budding occurs before the formation of the alveoli. Their location varies and depends on the embryonic stage of development at which the abnormality happens. Intrathoracic bronchogenic cysts have 2 radiographic classifications based on location: mediastinal and intrapulmonary. Mediastinal bronchogenic cysts account for approximately 20% of all primary mediastinal masses.[2] Mediastinal bronchogenic cysts appear as smooth, rounded, thin-walled, unilocular, sharply circumscribed structural opacities in the mediastinum adjacent to the tracheobronchial tree, often below the carina. Their location is in the middle mediastinum in 80% of cases, but they are also found in the posterior and anterior mediastinum in 17% and 3% of cases, respectively.[3] Intrapulmonary bronchogenic cysts are predominantly found in the lower lobes in the medial third of the parenchyma and are variable in size.[3] Bronchogenic cysts are also seen subcutaneously[4] and in the pericardium,[5] thymus, cervical region,[6] diaphragm, retroperitoneum, abdomen,[7] esophagus, and sternum.

Approximately 75% of bronchogenic cysts are asymptomatic and are found incidentally; symptoms such as cough, chest pain, dyspnea, or fever develop occasionally.[8] The size and location of the cyst are important factors in producing symptoms because the majority of symptoms are related to compression of the surrounding structures.[3] Compression of adjacent structures may lead to recurrent respiratory infections if the bronchi are affected; superior vena cava syndrome if the superior vena cava is affected; and hypoxemia, pulmonary hypertension, or atrial fibrillation if the bronchogenic cyst is located near the pulmonary

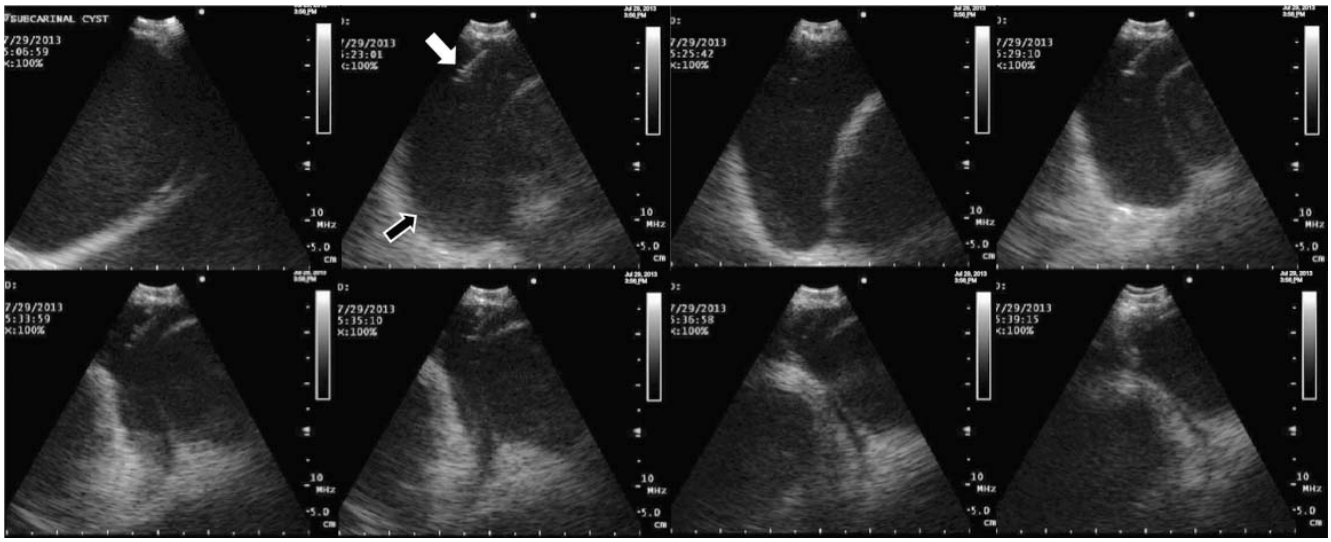

Figure 2. Sequential imaging of endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA) demonstrates a 5 cm subcarinal cystic lesion. Transbronchial needle aspiration (white arrow) was performed, and gradual reduction in the size of bronchogenic cyst (black arrow) was observed.

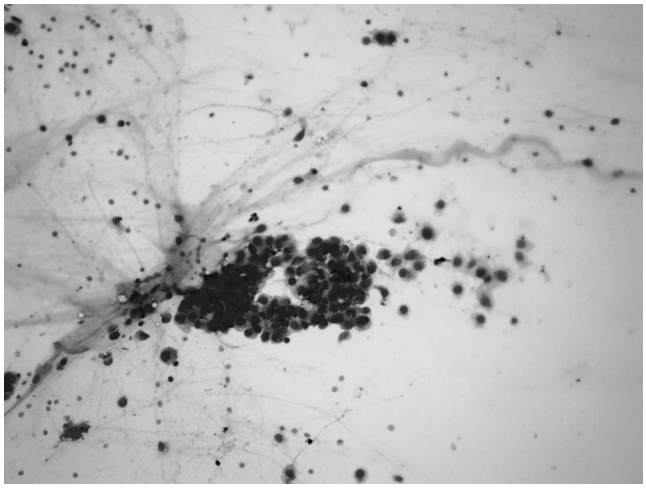

Figure 3. The slide shows abundant foamy macrophages with blood and lymphocytes (Diff-Quik stain, 20×). Extracellular matrix can be seen in the background, likely representing mucus from the respiratory tract. Although ciliated epithelial cells may be present, the sample consisted mainly of cyst contents.

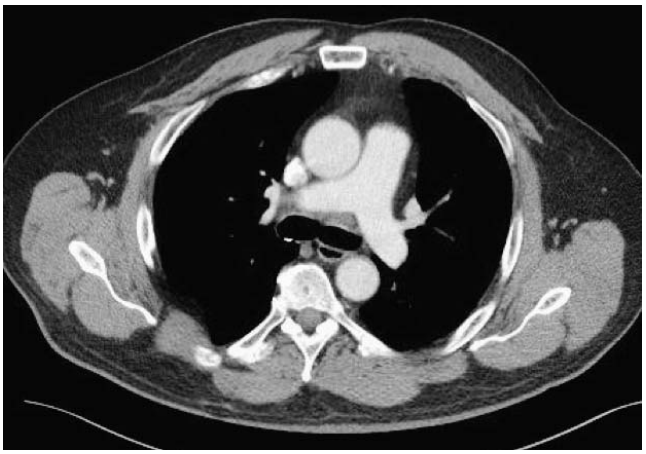

Figure 4. Chest computed tomography 3 months after cyst drainage shows a reduction in the size of the bronchogenic cyst with significant relief of the right pulmonary artery compression.

artery/atrium. Additional complications include infection of the cyst contents, fistulae to surrounding structures in the chest, cyst rupture, and hemorrhage into the cyst cavity. Cyst recurrence following incomplete excision as in our case has been reported, as well as malignant transformation into adenocarcinoma, squamous cell carcinoma, or carcinoid tumors.[7,9]

As mentioned above, several cases of bronchogenic cyst recurrence after incomplete surgical resection have been reported. Recurrence of the cyst usually takes years to appear.[10,11] Although primary bronchogenic cysts are commonly asymptomatic, recurrent bronchogenic cysts show symptoms more often. Bronchogenic cyst infection usually presents as areas of inhomogeneity with wall thickening on CT scan.[11]

Management of a bronchogenic cyst depends on symptoms and complications related to its location. Asymptomatic cysts in adults may be observed with periodic radiographic follow-up. For symptomatic cysts, complete surgical excision is usually the goal. Excision can be performed through a right or left thoracotomy or video-assisted thoracic surgery depending on cyst location.[9] Complete excision is imperative to reduce the risk of recurrence. If resection is incomplete, treatment with toxic agents such as ethanol injection into the cyst wall or electrocautery is required to destroy the epithelial layer.

TBNA has been suggested as an alternative method of decompression.[12] In our experience, as well as from review of the literature, EBUS-TBNA is a good diagnostic and therapeutic tool. When the cystic nature is confirmed by the EBUS, a 21- or 22-gauge EBUS needle can be inserted under direct EBUS vision, and TBNA can be performed by applying negative pressure. Cytologic analysis of the fluid should be performed to rule out any malignancy. EBUS-TBNA can provide instant decompression of the cyst and relieve the pressure on the surrounding structures.[13]

CONCLUSION

For symptomatic bronchogenic cysts, complete surgical excision is usually the goal, but TBNA is an alternate method of decompression that can instantly relieve the pressure on surrounding structures.

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