

# Infected Bronchogenic Cyst With Left Atrial, Pulmonary Artery, and Esophageal Compression

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Ultrasound-guided transbronchial needle aspiration was used successfully for both diagnosis and treatment of a rare bronchogenic cyst caused by an *Actinomyces* infection that was compressing mediastinal structures.

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**B**ronchogenic cyst is a rare foregut malformation that typically presents during the second decade of life that arises due to aberrant development from the tracheobronchial tree.<sup>1</sup> Mediastinal bronchogenic cyst is the most common primary cystic lesion of the mediastinum, and bronchogenic cysts of the mediastinum represent 18% of all primary mediastinal malformations.<sup>2</sup> Patients with mediastinal bronchogenic cysts may present with symptoms of cough, dyspnea, or wheezing if there is encroachment on surrounding structures.

Rarely, bronchogenic cysts can become infected. Definitive treatment of bronchogenic cysts is surgical excision; however, endobronchial ultrasound (EBUS)-guided drainage also can be employed. EBUS-guided drainage may be used when the cyst cannot be distinguished from solid mass on computed tomography (CT) images, to relieve symptomatic compression of surrounding structures, or to provide a histologic or microbial diagnosis in cases where surgical excision is not immediately available. We present the first-ever described case of bronchogenic cyst infected with *Actinomyces*, diagnosed by EBUS-guided drainage as well as a review of the literature regarding infected bronchogenic cysts and management of cysts affecting mediastinal structures.

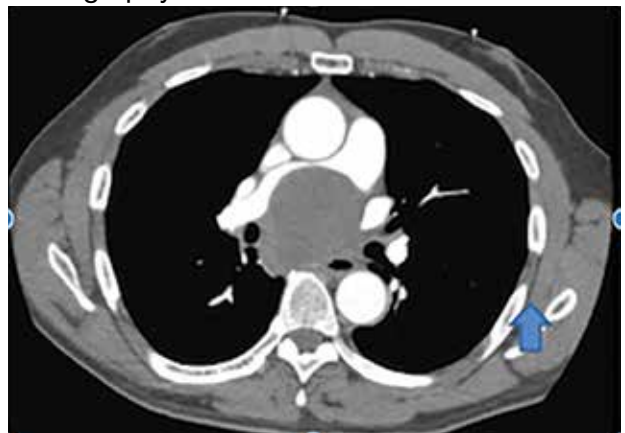
## CASE PRESENTATION

A 57-year-old African American male presented with a 4-day history of continuous, sharp, substernal chest pain accompanied by dyspnea. Additionally, the patient re-

ported progressive dysphagia to solids. The posteroanterior view of a chest X-ray showed a widened mediastinum with splaying of the carina. A contrast-enhanced CT of the chest showed a large, middle mediastinal mass of heterogeneous density measuring 7.3 × 7.0 × 6.0 cm with compression of the right pulmonary artery, left atria, superior vena cava and esophagus (Figure 1).

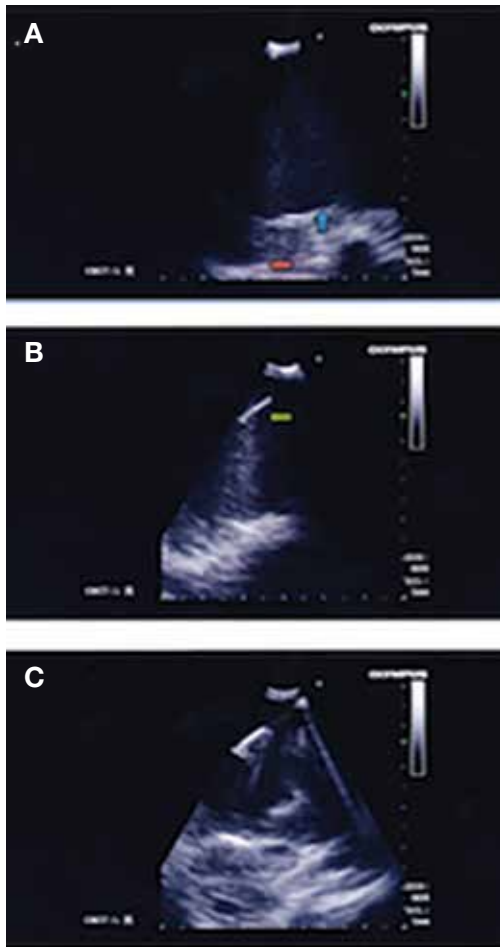
The mass demonstrated neither clear fluid-level nor rounded structure with a distinct wall and uniform attenuation consistent with pure cystic structure and, in fact, was concerning for malignant process, such as lymphoma. Due to the malignancy concern and the findings of significant compression of surrounding mediastinal structures, the decision was made to proceed with bronchoscopy and EBUS-guided transbronchial needle aspiration (EBUS-TBNA) to assist in diagnosis and potentially provide symptomatic relief.

**FIGURE 1** Preprocedure Axial View Computed Tomography of Chest With Contrast



A 7.3 × 7.0 × 6.0-cm heterogeneous mass compressing the right pulmonary artery is indicated by the blue arrow.

**FIGURE 2** Endobronchial Ultrasound Demonstrating Cystic Structure

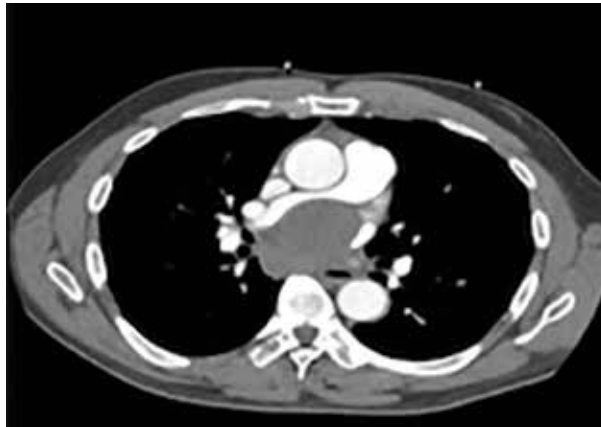


A, Cystic structure is indicated by blue arrow next to the pulmonary artery (red arrow). B, Insertion of needle is indicated by green arrow. C, Subsequent decompression is visible.

Under general anesthesia a P160 Olympus bronchoscope was advanced into the tracheobronchial tree; bronchoscopy with airway inspection revealed splayed carina with obtuse angle but was otherwise unremarkable. Next, an EBUS P160 fiber optic Olympus bronchoscope was advanced; ultrasound demonstrated a cystic structure. The EBUS-TBNA of cystic structure yielded 20 mL of brown, purulent fluid with decompression bringing pulmonary artery in ultrasound field (Figure 2). Rapid on-site cytology was performed with no preliminary findings of malignancy. The fluid was then sent for cytology and microbiologic evaluation.

Following EBUS-guided aspiration, the

**FIGURE 3** Postprocedure Axial View Computed Tomography of Chest With Contrast

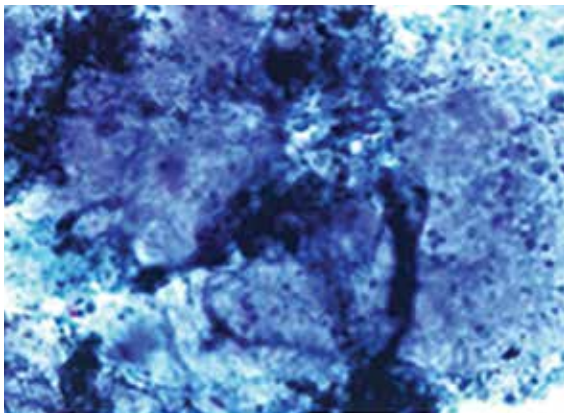


Cyst decreased in size to 5.9 × 5.5 × 4.6 cm with relief in compression of pulmonary artery.

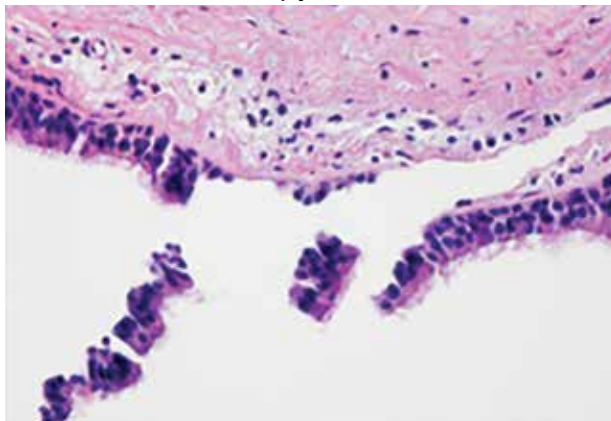
patient reported significant improvement in chest pain, dyspnea, and dysphagia. A repeat chest CT demonstrated decrease in mass size to 5.9 × 5.5 × 4.6 cm with relief of the compression of the right pulmonary artery and decreased mass effect on the carina (Figure 3). Pathology ultimately demonstrated no evidence of malignancy but did demonstrate filamentous material with sulfur granules and anthracotic pigment suggestive of *Actinomyces* infection (Figure 4).

The patient was placed on amoxicillin/clavulanate 875 mg to 125 mg twice daily for 4 weeks based on antibiotic susceptibility testing to prevent progression to mediastinitis related to *Actinomyces* infection. The duration of therapy was extrapolated from treatment regimens described in case series of cervicofacial and abdominal *Actinomyces* infections.<sup>3</sup> Thoracic surgery evaluation for definitive excision of cyst was recommended after the patient completed his course of antibiotics.

The patient underwent dental evaluation to identify the source of *Actinomyces* infection but there appeared to be no odontogenic source. The patient also had extensive skin survey with no findings of overt source of *Actinomyces* and CT abdomen/pelvis also identified no abscess that could be a potential source. He subsequently underwent thoroscopic resection with pathology demonstrating a fibrous cyst wall lined with ciliated columnar epithelium consistent with diagnosis of bronchogenic cyst (Figure 5).

**FIGURE 4** Smear of Aspiration

Smear obtained during endobronchial ultrasound-guided transbronchial needle aspiration showing sulfur granule aggregates of *Actinomyces* and anthracotic pigment.

**FIGURE 5** Tissue Obtained During Video-Assisted Thoracoscopy

Tissue demonstrates fibrous cyst wall lined by ciliated columnar cells consistent with diagnosis of bronchogenic cyst.

### DISCUSSION

Bronchogenic cysts can present at birth or later in life; patients may be asymptomatic for decades prior to discovery.<sup>4</sup> Cysts located in the mediastinum can cause compression of the trachea and esophagus and cause cough, dyspnea, chest pain, and dysphagia.<sup>5</sup> More life-threatening complications include infection, tracheal compression, malignant transformation, superior vena cava syndrome, or spontaneous rupture into the airway.<sup>6,7</sup>

Infection can occasionally occur, and various bacterial etiologies have been described. Hernandez-Solis and colleagues describe 12 cases of superinfected bronchogenic cysts with *Staphylococcus aureus* and *Pseudomonas aeruginosa*, the most commonly described organisms.<sup>8</sup> Casal and colleagues describe a case of  $\alpha$ -hemolytic *Streptococci* treated with amoxicillin.<sup>9</sup> Liman and colleagues describe 2 cases of bronchogenic cyst infected with *Mycobacterium* and cite an additional case report by Lin and colleagues similarly infected by *Mycobacterium*.<sup>10,11</sup> Only 1 case was identified to have direct bronchial communication as a potential source of introduction of infection into bronchogenic cyst. In other cases, potential sources of infection were not identified, though it was postulated that direct ventilation could be a potential source of inoculation.

Surgical resection of mediastinal bronchogenic cysts has traditionally been considered the definitive treatment of choice.<sup>12,13</sup> How-

ever, bronchogenic cysts may sometimes be difficult to differentiate from soft tissue tumors by chest CT, especially in cases of cysts with nonserous fluid. In particular, cysts that are infected are likely to have increased density and high attenuation on imaging; therefore, surgical excision may be delayed until diagnosis is made.<sup>14</sup> Due to low complication rates, EBUS is increasingly used in the diagnosis and therapeutic management of bronchogenic cysts as an alternative to surgery, particularly for those who are symptomatic.<sup>15,16</sup> Ultrasound guidance can allow for complete aspiration of the cyst, causing complete collapse of the cystic space and can facilitate adhesion between the mucosal surfaces lining the cavity and reduce recurrence.<sup>17</sup> Nonetheless, bronchogenic cysts that are found to be infected, recur, or have a malignant component should be resected for definitive treatment.<sup>18</sup>

The mass discovered on our patient's imaging appeared to have heterogenous attenuation consistent with malignancy rather than homogenous attenuation surrounded by a clearly demarcated wall consistent with a cystic structure; therefore, EBUS-TBNA was initially pursued and yielded an expedited diagnosis of the first-ever described bronchogenic cyst with *Actinomyces* superinfection as well as dramatic symptomatic relief of compression of surrounding mediastinal structures, particularly of the right pulmonary artery. As this is a congenital malformation, the patient was likely asymptomatic

until the cyst became infected, after which he likely experience cyst growth with subsequent encroachment of surrounding mediastinal structures. Additionally, identification of pathogen by TBNA allowed for treatment before surgical excision, possibly avoiding accidental spread of pathogen intraoperatively.

## CONCLUSIONS

Our case adds to the literature on the use of EBUS-TBNA as a diagnostic and therapeutic modality for bronchogenic cyst. While cases of mediastinitis and pleural effusion following EBUS-guided aspiration of bronchogenic cysts have been reported, complications are extremely rare.<sup>19</sup> EBUS is increasingly favored as a means of immediate diagnosis and treatment in cases where CT imaging may not overtly suggest cystic structure and in patients experiencing compression of critical mediastinal structures.

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## Author disclosures

The authors report no actual or potential conflicts of interest with regard to this article.

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