Bronchogenic Cysts Presenting as Thymic Cysts

HARRY W. DONIAS, MD; QUYEN D. CHU, MD; TIMOTHY M. ANDERSON, MD;
RAFFY L. KARAMANOUKIAN, MD; WILLIAM J. GIBBONS, MD;
HRATCH L. KARAMANOUKIAN, MD

abstract Although relatively rare, bronchogenic cysts are the most common of the cystic mediastinal masses. Because the majority will become symptomatic or complicated, bronchogenic cysts should be considered in differential diagnoses of mediastinal masses. Bronchogenic cysts most commonly occur in the middle or posterior mediastinum but have been reported throughout the thorax as well as in other atypical locations. We report the diagnosis and treatment of two extremely rare cases of bronchogenic cysts occurring within the thymus.

Bronchogenic cysts are relatively uncommon congenital anomalies of lung development. They arise when a group of cells break off from the developing lung bud and differentiate on their own.\(^1\) They are, however, the most common cystic mediastinal masses, representing 5%–15% of all primary mediastinal tumors, so should be included in the differential diagnosis of any cystic mass appearing in the mediastinum.\(^1-4\) Bronchogenic cysts usually represent incidental findings on radiographs, but can produce compression or irritation of adjacent structures, causing symptoms that most commonly include chest pain, cough, and dyspnea.\(^1-3\)

While bronchogenic cysts typically occur in the middle or posterior mediastinum in the paratracheal or subcarinal region, they are not infrequently encountered throughout the mediastinum or thorax. They have also been reported to occur in atypical locations including the neck, intradural positions, within the pericardium, or subdiaphragmatic space.\(^1,2,5\)

Most authors recommend that all presumed bronchogenic cysts seen in the adult population be completely resected because the majority will ultimately become symptomatic or complicated, and because definitive diagnosis can only be established based on the surgical specimen.\(^1,5\) We present two cases of bronchogenic cysts occurring within the thymus, an extremely rare presentation.\(^1,6\)

Embryology

The respiratory system and the esophagus have a common embryologic origin. During development, the primitive respiratory system develops as a ventral diverticulum located in the floor of the foregut, which will become the primitive bronchial tree.\(^2\) The respiratory budding arises between days 20 and 40 of gestation. The cleavage between the respiratory and the digestive tube occurs on the 28th day of gestation,\(^3\) and the lobar bronchi develop by the 35th day.

Bronchogenic cysts are an anomaly of bronchial development from the primitive ventral foregut, and

From the Departments of Surgery, *Pulmonary Medicine, and Cardiothoracic Surgery*, State University of New York at Buffalo and Kaleida Health at Buffalo General Hospital; the Departments of Surgical Oncology and Thoracic Surgery, Roswell Park Cancer Institute, Buffalo, NY; and the Department of Surgery, University of California, San Diego.

Correspondence: Hratch L. Karamanoukian, MD; Center for Less Invasive Cardiac Surgery and Robotic Heart Surgery, Kaleida Health at Buffalo General Hospital, 100 High Street, Buffalo, NY 14203; telephone (716) 859-1080; fax (716) 859-4697 (e-mail: Lisbon5@yahoo.com).
arise from cells that are isolated from the main pulmonary branching when the lung bud separates from the primitive gut. When this abnormal budding occurs early in gestation, the cysts tend to be located along the tracheobronchial tree, usually in the middle or posterior mediastinum, and they seldom communicate with the trachea or bronchi. Cysts arising later in gestation are more peripheral and may be located within the lung parenchyma, and often have a patent bronchial communication. This explains why cysts lined with respiratory-type mucosa are apt to develop not only along the trachea, along the bronchi, and within the lung depending on the moment of their formation, but also along the esophagus, including below the diaphragm as the esophagus grows longer caudally during the second month of gestation.

Development of the thymus typically begins after the majority of bronchogenic cysts have already formed. Paired thymic primordia first appear as buds on the ventral aspect of the third pharyngeal pouches late in the sixth week of development. During the eighth week, the caudad ends enlarge, unite superficially, and attach to the anterior pericardium, which enhances the descent of the thymus into the thorax.

We believe that the bronchogenic cysts seen in our two patients developed late in gestation and were engulfed by the developing thymus.

Histology
Typical histological features of bronchogenic cysts include a ciliated columnar epithelial lining and a connective tissue wall. Unless infected, they contain thick mucus with focal or extensive areas of squamous metaplasia and chronic inflammatory infiltrate. Their walls may also contain one or more of the tissues normally found in the tracheobronchial tree such as hyaline cartilage plates, smooth muscle, bronchial mucus glands, and nerve trunks.

Anterior mediastinal cystic lesions of the thymus represent approximately 1% of all mediastinal masses. Bronchogenic cysts occurring in the thymus are extremely rare. However, location makes no practical difference in the management of bronchogenic cysts, as it is usually recommended that all bronchogenic cysts in the adult be resected. Work by St-Georges et al, who found that bronchogenic cysts in adults become symptomatic or complicated in 72% of cases, supports this theory.

Surgical Indications
The most common, serious complication of bronchogenic cysts is infection. This is especially common in those with bronchial communication. Early resection prevents a difficult or even hazardous operation once the cyst becomes symptomatic or complicated. In addition, early surgical intervention is important to obtain a tissue diagnosis.

Although rare, malignant degeneration of bronchogenic cysts has also been reported. Therefore, any cystic lesion that does not have the classic roentgenographic appearance of a mediastinal bronchogenic cyst, especially those that are intraparenchymal, should be removed surgically to exclude the possibility of a malignant or infectious lesion. In addition, any suspicious lesion of the thymus should be treated with total thymectomy.

Various surgical treatments are available for excision. A median or partial split sternotomy was used in the two cases presented here. Mediastinoscopy with piecemeal removal with biopsy forceps has been described; however, caution should be exercised in using this method for infected cysts or those suspicious for malignancy. Both left and right thoracotomy have also been used depending of the location of the cyst, and more recently excision has been described using video-assisted thoracoscopic surgery (VATS). When the lesion is confined to the thymus, thymectomy may be performed via sternotomy, transversely, or with VATS. If complete excision is not possible due to involvement of adjacent organs, partial excision with “de-epithelialization” of the residua can be performed, and if the patient is not a surgical candidate, bronchoscopic or thoracoscopic needle drainage is an alternative.

Conclusion
Although bronchogenic cysts are the most common cystic mediastinal mass, they are relatively uncommon in the adult population. Furthermore, bronchogenic cysts presenting within the thymus are extremely rare. However, the two cases presented here serve to demonstrate that bronchogenic cysts should be included in the differential diagnosis of any cystic mediastinal mass, regardless of its location.
**CASE REPORT**

**Thymectomy via median sternotomy for bronchogenic cysts in the thymus**

**Case 1.** A 73-year-old woman with a medical history significant for hypertension, asthma, and bladder cancer presented to her primary care physician with complaints of progressive shortness of breath and paroxysmal coughing. The only significant finding on physical examination was that of an expiratory wheeze noted throughout all lung fields. Work-up of her symptoms included a chest radiograph, which showed a soft-tissue density adjacent to the paratracheal stripe in the right superior mediastinum. Chest computed tomographic (CT) scans showed a soft-tissue mass in the anterior mediastinum measuring 1.5 cm in diameter. The patient was referred to the cardiothoracic service, where exploration and excision of the mediastinal mass was planned. At operation, a 1.5-cm, firm, well-encapsulated mass was palpated in the right lobe of the thymus, and a total thymectomy was performed via median sternotomy. The pathological specimen revealed a unilocular, smooth-walled cyst measuring 1.5 x 1.2 x 1.0 cm lined with respiratory-type epithelium containing tan viscous fluid. The mass was identified as a bronchogenic cyst with focal-reactive inflammation (Figures 1, 2).

The patient made an unremarkable recovery with the exception of a short run of atrial fibrillation that rapidly responded to low-dose metoprolol. She was discharged home on the third postoperative day (POD 3) and was asymptomatic at the three-month follow-up.

**Case 2.** A 62-year-old male presented to his primary care physician with a chief complaint of left scapular pain. During the work-up, a chest radiograph demonstrated an abnormal lesion in the anterior mediastinum. A CT scan of the chest confirmed the mass, measuring 3.2 x 2.4 cm in maximal diameter. (Of note, the work-up for the left scapular pain, including a bone scan, was negative and the patient’s symptoms resolved.) The patient had a repeat CT scan of the chest at three months that demonstrated the same mass, now measuring 3.4 x 2.6 cm (Figure 3). The patient was otherwise asymptomatic. He denied any infectious exposure in the past or travel to any exotic places. He smoked a pack of cigarettes per day for 30 years and had no other significant comorbidities. He underwent a partial sternal split and thymectomy. He made an unremarkable recovery, was discharged on POD 4, and remained asymptomatic at 3 months.

*FIGURE 1. Low-power (2X objective) view of partially collapsed cyst within thymic tissue.*

*FIGURE 2. High-power (20X objective) view of respiratory-type epithelium of cyst lining.*

*FIGURE 3. Chest CT scan demonstrating the anterior mediastinal mass incidentally discovered during the work-up for an unrelated complaint.*
References

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