

## Surgical Treatment of a Bronchial Cyst Causing Compression to the Left Atrium and Pulmonary Artery

Cenk Eray Yildiz, MD,<sup>1</sup> Sina Ercan, MD,<sup>2</sup> Mehmet Umit Ergenoglu, MD,<sup>1</sup>  
Halit Yerebakan, MD,<sup>1</sup> Ferda Ozkan, MD,<sup>3</sup> Suha Kucukaksu, MD<sup>1</sup>

Divisions of <sup>1</sup>Cardiovascular Surgery, <sup>2</sup>Thoracic Surgery, and <sup>3</sup>Pathology, Yeditepe University Hospital, Istanbul, Turkey



Dr. Yildiz

### ABSTRACT

Bronchogenic cysts that formed during the development of the tracheobronchial tree in the gestational period are mostly asymptomatic until adulthood. Cysts localized in the middle mediastinum, specifically in the subcarinal region, on the other hand, may cause serious symptoms by compressing the heart and major vessels due to their close proximity. In this report it is suggested that surgical resection of a giant bronchogenic cyst that compresses the heart and great vessels should be securely performed in the presence of a readily available cardiopulmonary bypass capability.

### INTRODUCTION

Bronchogenic cysts are tracheobronchial system anomalies caused by abnormal budding of the tracheal diverticulum at the time of gestational development of the tracheobronchial tree. If this bud's connection with the tracheobronchial tree is interrupted, it develops as an enlarging cyst due to internal secretions. If its connection remains patent, then it will contain air as well besides the secretions [Rogers 1964]. While bronchogenic cysts are usually localized in the mediastinum as they are formed in earlier stages of gestational period, they may also locate within lung parenchyma when they are formed in later periods. Within the mediastinum, the subcarinal and paratracheal regions are more common locations [St-Georges 1991]. The cyst wall contains cartilage and smooth muscle and is internally covered with ciliary epithelium and bronchial glands secreting a mucoid fluid.

Bronchogenic cysts can be symptomatic according to their localization, size, infectious state, the age of the patient; they may stay totally asymptomatic as well [Kanemitsu 1999]. Though compressive symptoms of alimentary and respiratory

tract may be seen in neonates and infants, adults may present with hemoptysis or infectious symptoms. Still, most patients are diagnosed coincidentally. Draining of the infected mediastinal cyst into to the pleura, pericardium, and bronchus is rarely seen [Rimet 1996].

### CASE REPORT

A 22-year-old female patient presented to the emergency room with increasing respiratory distress and cough complaints for 24 hours. The patient also described night sweats, fever, and retrosternal pain for 3 weeks. On physical examination, blood pressure was 110/70 mmHg, and heart rate was 118 beats per minute as sinus tachycardia. There were coarse respiratory sounds with basilar rales. She had leucocytosis ( $11.73 \times 10^3/\mu\text{L}$ ) and elevated C reactive protein level (46 mg/L, Normal <5).

The patient's chest x-ray (Figure 1A) showed fullness in the right paracardiac region distal to carina, and thoracic computerized tomography (CT) delineated a subcarinal cystic mass in the middle mediastinum, significantly compressing the right pulmonary artery, and both compressing and posteriorly displacing the left main bronchus (Figure 1B-C). The mass was tightly saddled between the 2 right pulmonary veins.

Barium esophagography was normal with no luminal connection with the cyst. Surgical exploration was planned with a preliminary diagnosis of bronchogenic cyst due to its highly compressive nature. Following the induction for anesthesia in the operating room, the compressed left main bronchus was internally stented with the bronchial arm of the left sided double-lumen endotracheal tube. The computed tomography of the thorax also showed a suspicious tongue-shaped filling defect, indicative of a thrombus, protruding from the right inferior pulmonary vein into the left atrium. Therefore the right inguinal region was prepared and sterilely draped during right posterolateral thoracotomy for a possible cardiopulmonary bypass. The right hemithorax was entered through the sixth intercostal space. The lung was freed by cutting the inferior pulmonary ligament, and the inferior pulmonary vein was exposed. A mass was seen between the pulmonary vein and bronchi in the hilum of the right lung. A parallel incision to the phrenic nerve was performed to open the pericardium; it showed that the mass was compressing the

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Correspondence: Cenk Eray Yildiz, MD, Assistant Professor at Yeditepe University, School of Medicine, T.C. Yeditepe University Hospital, Division of Cardiovascular Surgery, Devlet Yolu Ankara Cad. No: 102/104, 34752 Kozyatagi/Istanbul, Turkey; +90-216-578-4815; fax: +90-216-578-4939 (e-mail: [ceyildiz@yeditepe.edu.tr](mailto:ceyildiz@yeditepe.edu.tr) or [ceyildiz@hotmail.com](mailto:ceyildiz@hotmail.com)).

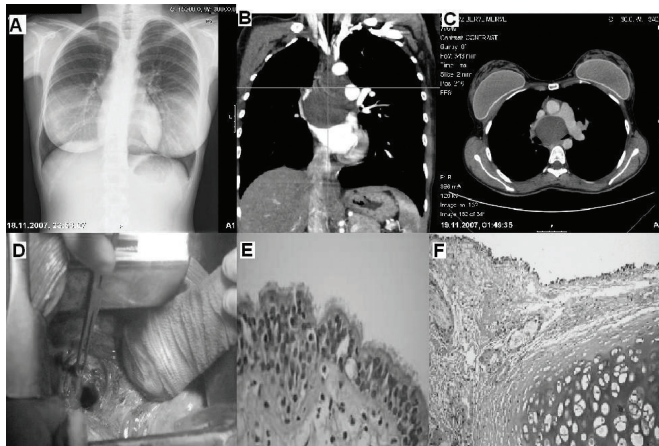


Figure 1. A, Preoperative posterior to anterior chest x-ray. B, Preoperative thorax computed tomography (CT) with contrast, coronal sections. C, Preoperative thorax CT with contrast, axial sections. D, Aspirated and opened subcarinal cyst at the level of posterior hilum. E, Ciliated columnar epithelium with inflammation, hematoxylin & eosin stain (H&E)  $\times 400$ . F, Cartilage tissue in the wall covered with ciliated epithelium, H&E stain  $\times 100$ .

superior vena cava, the left atrium, both pulmonary veins, and the aorta. A cyst ( $10 \times 12$  cm) was posteriorly located behind the left atrium, reaching up to the left atrial appendix contralaterally. Intraoperative ultrasonography was performed by gentle manipulation to visualize the right inferior pulmonary vein and left atrial lumen. There was no thrombus seen within the right inferior pulmonary vein, confirming the results of preoperative transthoracic echocardiography. While contrasting the preoperative thoracic CT scan findings, this negated the need for cardiopulmonary bypass. The esophagus was localized by nasogastric catheter placement. Dissection was started by detaching the mass from the esophageal wall. The cyst wall was exposed in the subcarinal region. Keeping the possibility of a hydatid cyst in mind, the dome of the cyst was encircled with sponges soaked with hypertonic saline, and the thick yellow cystic fluid was aspirated with a needle connected to the surgical suction device with utmost attention to prevent any spillage. The fluid was cultured with no later growth. Then the cyst was opened showing thick septations and a dense and fibrinous material with some organized thrombus (Figure 1D). Then the cyst was completely cleaned in pieces all the way to the contralateral side behind the left atrium without opening the left pleura. The operation was ended once the hemostasis was completed, and air leaks were checked. There was no perioperative or postoperative complication, and the patient was dismissed on postoperative day 5.

The presence of a ciliated columnar epithelium and cartilage tissue with active chronic infiltrative inflammation, hyperemia, and bleeding in the histopathologic evaluation confirmed the diagnosis of a bronchogenic cyst (Figure 1E-F). In direct microscopy of the cystic fluid, there was lymphocytic predominance, and no microorganism was seen. Following an uneventful hospital course and dismissal, the patient returned

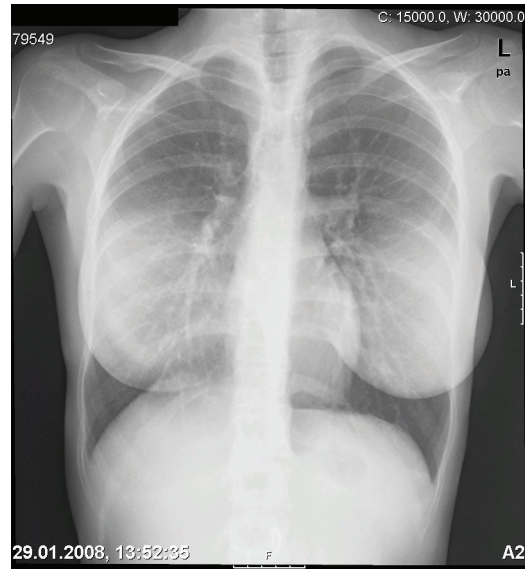


Figure 2. Chest x-ray at 2 months postoperative.

to an outpatient clinic at the end of the second postoperative month. On the control chest x-ray at that time, the fullness at the right para-mediastinal region was cleared (Figure 2). Currently, 18 months after surgery, the patient is doing well with no sign of recurrence.

## DISCUSSION

Bronchogenic cysts cause symptoms when they compress adjacent airways or become infected if open into bronchi. Mediastinal bronchogenic cysts can be classified in 5 groups: paratracheal, carinal, hilar, paraesophageal, and mixed type [Di Lorenzo 1989]. Occasionally they can be found in the presternal or supraclavicular region, intrapericardially, or under the diaphragm as well [Amendola 1982]. In our case the  $10 \times 12$  cm cyst was subcarinally located, compressing the superior vena cava, left atrium, both right pulmonary veins, and the aorta. To our knowledge this is one of the largest reported bronchogenic cysts in the posterior mediastinum. It also considerably decreased the diameters of the left main bronchus and right pulmonary artery (Figure 2). Carinal type bronchogenic cysts especially may cause signs of left atrial overload due to its close proximity. The patient had been having night sweats, high fever, and intermittent angina for 3 weeks, with gradually increasing dyspnea and cough for the last 24 hours.

Although the mediastinal or hilar bronchogenic cysts are well bordered in radiologic studies, it may still be hard to differentiate between solid or cystic nature. They may have a high protein or calcium content, or the presence of infection may yield a high density. This may leave surgical exploration as the only way of reaching a definitive diagnosis in some cases.

Some clinicians, unless the cyst becomes symptomatic or gets complicated, prefer conservative therapy due to its high probability of benign nature. On the other hand, others

prefer surgical excision due to the risk of fistulization, bronchial ulceration, bleeding, or infection [Bolton 1992].

Subcarinal and retro cardiac cysts especially, either diagnosed incidentally or owing to their symptoms, should be surgically resected by protecting the surrounding vital organs. These patients should be scanned periodically if there is the slightest doubt of incomplete resection, because relapsing cysts and malignant degeneration have been reported [Okada 1996].

The complete surgical resection of bronchogenic cysts is recommended to obtain a definitive diagnosis, relieve symptoms, and eliminate the risks of infection and malignant degeneration. It is advisable that when operating, particularly on central cysts compressing the heart and great vessels, cardiopulmonary bypass should be readily available for minimizing potential cardiac or vascular injuries and other surgical risks such as air emboli.

Despite all efforts made for a complete resection, this may not be possible with a large subcarinal cyst in close proximity with the left atrium and heart as reported by Mawatari et al [Mawatari 2003]. It was also proposed by the same authors that the use of video-assisted thoracoscopic surgery (VATS) for the removal of bronchogenic cysts should be considered particularly in cases for which partial resection and ablation are planned in advance or when relapse occurs. Due to well documented risk of local recurrence and malignant degeneration, it is our belief that a complete resection should be the goal in every case. If a complete resection was not achievable in the first surgery,

VATS resection would likely not be feasible for resection in local recurrence either.

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