

# Extrapericardial bronchogenic cyst associated with atrial fibrillation

## Abstract

Bronchogenic cysts are rare and usually asymptomatic. There are very few reports of bronchogenic cysts producing atrial fibrillation in the literature. We present a rare case of atrial fibrillation as a complication of a mediastinal bronchogenic cyst. To our knowledge, there have been only 13 cases of atrial fibrillation caused by a bronchogenic cyst published in the literature. As demonstrated by prior studies, surgical resection has been a successful treatment of the atrial fibrillation in this population. Our case report presents additional evidence to support the effectiveness of surgical resection in the treatment of bronchogenic cyst induced atrial fibrillation.

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## Introduction

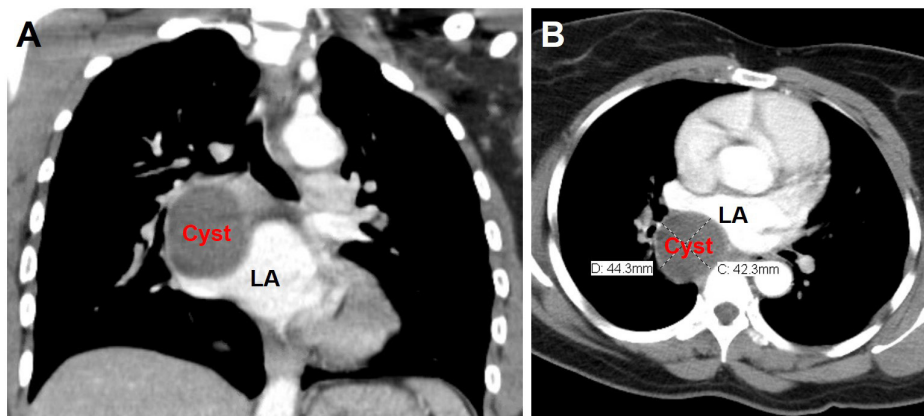
A Bronchogenic cyst is a rare congenital intrathoracic lesion that accounts for approximately 5%–20% of primary mediastinal masses.<sup>1-3</sup> The majority of cysts are asymptomatic or have a nonspecific clinical presentation due to the compression of surrounding structures.<sup>4</sup> Cardiovascular complications have rarely been reported and are usually due to an intrapericardial location of the cystic mass.<sup>3</sup> In this article, we present a patient with an extrapericardial bronchogenic cyst that manifested with new-onset paroxysmal atrial fibrillation with rapid ventricular response (RVR). The arrhythmia was caused by direct compression on the left atrium and pulmonary vein and was managed by performing a surgical cyst excision. Patient has remained asymptomatic for 15 months postoperatively.

## Case report

A 43-year-old African American female presented with progressively worsening chronic nonproductive cough, shortness of breath and palpitation without chest pain, lightheadedness or syncopal episodes. She was recently diagnosed with new-onset paroxysmal atrial fibrillation (AF) three months prior to presentation and was prescribed Eliquis for thrombosis prophylaxis. On presentation, an ECG revealed atrial fibrillation with RVR with a rate of 150bpm

and prolonged QT interval. A Chest CT scan demonstrated a large 43x44x50 mm posterior mediastinal cystic lesion in the right infrahilum region causing a mass effect on the bronchus intermedius as well as the left atrium. The appearance of the mass on CT was consistent with a bronchogenic cyst (Figure 1). The patient's new-onset AF was attributed to the compression of left atrium and pulmonary vein. Patient was prescribed flecainide and subsequently pharmacologically converted to sinus rhythm.

After rhythm stabilization, stress test and cardiology clearance, patient was taken to the operating room for a bronchogenic cyst excision. During bronchoscopy, no endobronchial lesions were observed; however, the right bronchus intermedius was almost completely collapsed due to external compression. A VATS procedure was then attempted. Findings demonstrated a large firm cystic structure in the posterior mediastinum that was compressing and intimately adherent to the subcarinal region, right lower lobe and extended to the pulmonary vein with extensive pericystic inflammation and fibrosis. Due to dense adhesions and inability to safely isolate the cystic lesion, the procedure was converted to a thoracotomy. Dissection proceeded and the cyst wall was freed from surrounding structures employing extreme caution to prevent damage of adherent anatomy and avoiding cautery adjacent to the airway. The entire cyst was excised and sent to pathology.



**Figure 1** Chest CT images showing large homogeneous, low-density, unilocular, well-circumscribed mass 43mmx44mmx50mm (17 to 20 Hounsfield units), with direct extrinsic compression of the left atrium. (A) coronal and (B) axial views.

Pathologic interpretation was confounded by cystic alterations noted by inflammation, hemorrhage and fibrosis. Secondary to these features, and lack of cartilage within the cystic wall, the cyst was initially thought to represent a thymic cyst. Additional features of the specimen demonstrated largely denuded ciliated columnar epithelial lining of the cyst. After further review, final pathology was determined to be consistent with inflamed and altered bronchogenic cyst.

Postoperatively patient begin ambulating on postoperative day 1 and chest tube was removed. The patient was discharged home on postoperative day 2 in normal sinus rhythm with a prescription of flecanide. Follow-up ECGs performed at 1, 5 and 12 months have demonstrated normal sinus rhythm and patient continued to be asymptomatic at 15 months.

## Discussion

A Bronchogenic cyst is a rare congenital benign neoplasm of primitive foregut origin that embryologically separates from a normal tracheobronchial tree during the third week of gestation. Bronchogenic cysts most commonly occur in the mediastinum adjacent to the major airways.<sup>3-6</sup> The classic histological findings associated with bronchogenic cysts are ciliated columnar epithelial cells that comprise the inner lining along with thick walls that typically contain cartilage and bronchial mucous glands, however these features are not essential for the diagnosis.<sup>1,7</sup>

Most cysts in adults are asymptomatic and incidentally identified on radiography; nevertheless, they can present when complications occur.<sup>4,8,9</sup> The clinical presentation depends on the location of the cyst and ranges from nonspecific symptoms such as cough,

chest pain and/or dyspnea, to severe life-threatening conditions secondary to infection, hemorrhage, or cystic rupture.<sup>5</sup> Enlarging lesions become symptomatic due to direct compression of adjacent anatomical structures. Cardiovascular complications are extremely rare and include various types of arrhythmias, sudden cardiac death, cardiac tamponade, as well as SVC syndrome.<sup>1,3,7,10</sup> Stretching of the pulmonary vein is known to trigger irregular neurotransmission of cardiomyocytes.<sup>5</sup> The insidious onset of atrial fibrillation in a young individual without underlying cardiac pathology should raise suspicion of a bronchogenic cyst. The standard diagnostic workup of a bronchogenic cyst should include plain radiography, echocardiography, and a CT scan of the chest. In the present case, the new onset of atrial fibrillation was due to direct compression of left atrium and pulmonary vein. Only 13 similar cases of atrial fibrillation secondary to a bronchogenic cystic have been described in the English literature (Table 1)<sup>1-13</sup> The gold standard for management of cardiac complications associated with cystic lesions is surgical resection. The most common approach for resection is thoracotomy which has been demonstrated by the 77% of the cases described in the literature. This approach is performed in the majority of cases due to extensive fibrosis and inflammation.<sup>2,4-11,13</sup> VATS or a robotic-assisted approach is an alternative surgical option for decompression or excision of a bronchogenic cyst.<sup>3,12</sup> Complete resection of the cystic wall is crucial because there is increased likelihood of cyst recurrence and malignant transformation following incomplete excision.<sup>1,12</sup> hence, complete cyst removal is recommended even if the patient is asymptomatic.<sup>6-8,10</sup> In poor surgical candidates, endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA), transthoracic CT-guided aspiration or mediastinoscopy may provide an alternate treatment strategy.<sup>1,8,13</sup>

**Table 1** Extrapericardial bronchogenic cysts associated with new-onset atrial fibrillation

Author	Year	Age	Sex	Treatment	Follow-up
Volpi et al. <sup>4</sup>	1988	36	M	Thoracotomy	-
Johnston et al. <sup>8</sup>	1992	44	F	CT-guided aspiration and thoracotomy	16 months
Mather et al. <sup>9</sup>	1995	50	F	Thoracotomy	-
Sick et al. <sup>11</sup>	2000	61	M	Thoracotomy	-
Parambil et al. <sup>5</sup>	2006	38	M	Thoracotomy	12 months
Marshall et al. <sup>7</sup>	2007	32	M	Thoracotomy	-
Fujino et al. <sup>6</sup>	2010	62	M	Thoracotomy	36 months
Narula et al. <sup>13</sup>	2013	53	M	EBUS-TBNA and thoracotomy	-
Alraiyes et al. <sup>1</sup>	2015	67	M	EBUS-TBNA	3 months
Smail et al. <sup>2</sup>	2015	66	M	Mediastinoscopy and thoracotomy with deroofting	24 months
Han et al. <sup>10</sup>	2018	49	F	Thoracotomy	60 months
Liu et al. <sup>3</sup>	2018	29	F	Robotic-assisted VATS	-
Nakamura et al. <sup>12</sup>	2019	83	F	Prone-position VATS with palliative fenestration	18 months

## Data availability

The data used to support the findings of this study are included within the article.

## Conflicts of interest

All authors have no financial disclosures or conflicts of interest to disclose.

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## Informed consent

Written/Verbal informed consent was obtained from all individual participants included in the study.

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