

# Giant Intrapericardial Bronchogenic Cyst Associated with Congestive Heart Failure and Atrial Fibrillation: A Case Report

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## Case report

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# Abstract

**Background:** Large intracardiac bronchogenic cysts are rare mediastinal masses, however they must always be considered in the differential diagnosis of heart failure.

**Case Presentation:** We present a 60-year-old female patient with de novo atrial fibrillation and heart failure, resulting from an incidental large intrapericardial mass. The patient underwent successful surgical resection, with pathological findings confirming a bronchogenic cyst.

**Conclusions:** Large bronchogenic cysts located intrapericardially are very rare, however they should be included in the differential diagnosis of patients presenting with atrial fibrillation and heart failure.

## Background

A bronchogenic cyst is a remnant of the primitive gut tube occurring as a consequence of abnormal embryonic development in the tracheobronchial tree, between the 5th and 16th gestational week<sup>1-3</sup>. It is a rare congenital defect, with an incidence of 1/50000 people<sup>4</sup>, representing 14–22% of all pulmonary congenital defects<sup>2</sup> and 10–15% of all primary mediastinal masses<sup>1</sup>. The average diameter ranges from 2-4cm<sup>2</sup> and is classified based on the localization as parenchymal or mediastinal, the latter being the most common in 86% of cases. Intrapericardial and intracardiac locations are not common in the literature<sup>5</sup>. We present a case of a patient whom underwent successful resection of a giant intrapericardial bronchogenic cyst of 8 × 6 cm, written according to surgical case report (SCARE) criteria<sup>6</sup>.

## Case Presentation

A 66-year-old female patient, with no previous cardiovascular diseases, presented to the emergency department (ED) with severe chest pain, worse on exertion and associated with fatigue. Patient described multiple similar episodes in the preceding months, with increasing frequency and pain intensity prompting presentation to ED. Upon arrival, the electrocardiogram showed a de novo atrial fibrillation with a rapid ventricular response. The transthoracic echocardiogram revealed a non-obstructive concentric hypertrophic left ventricle with moderate systolic dysfunction – left ventricular ejection fraction (LVEF) of 35–40%, sclerosis of the mitral and aortic valve without hemodynamic compromise, mild tricuspid insufficiency, and moderate pericardial fluid. These findings correlated with signs and symptoms of congestive heart failure, prompted diuretic management with furosemide and rate control with beta-adrenergic blocking agents, which failed to control the heart rate and intravenous amiodarone was commenced.

A coronary arteriography ruled out coronary disease. A transesophageal echocardiogram revealed a LVEF of 51%, grade 1 tricuspid insufficiency, moderate pulmonary hypertension, moderate pericardial effusion, and multiple pericardial masses. The pericardial masses were further investigated with a chest contrast computed tomography (CT) scan revealing a 10 cm compressive mass in the postero-medial

mediastinum suggestive of a pericardial cyst. Cardiac magnetic resonance image (MRI) defined a 96 × 88 × 77 mm mass in its transverse, antero-posterior and cephalo-caudal diameters, respectively. The mass was located in the superior pericardial ridge, in contact superiorly with the pulmonary artery, posteriorly with the superior vena cava, anteriorly with the vertebral bodies, and cephalically with the left atrium producing partial compression. The mass had a homogeneous high signal intensity in T2, without any changes in the opposite phase and did not highlight with intravenous contrast [Figure 1].

The patient underwent surgical resection via a conventional sternotomy, however due to firm adhesions between the pericardium and epicardium, it was impossible to reach the mass. Extracorporeal circulation with bicaval aortic cannulation was therefore commenced and adhesiolysis completed to provide an accessible approach to the 8 × 6 cm cyst. A complete division and extraction of the cyst wall from the left atrium, superior and inferior pulmonary veins, the right pulmonary artery and the superior vena cava, successfully achieved its resection [Figure 2]. Specimen and pericardial biopsy were sent to pathology.

After a 4-day intensive care unit (ICU) stay, the patient was moved to a general ward. A post-operative transthoracic echocardiogram showed an LVEF of 55%, without any pericardial effusion or evidence of heart failure. Given the favorable outcome and evolution, the patient was discharged after 3 days later, with a 7-day total in-hospital stay. Two weeks later, during out-patient follow-up, the pathological report described an 8 × 6 cm mass of 0.3 cm wall thickness of fibroelastic consistency, with histological findings suggestive of a benign cystic lesion made up of fibromuscular wall and mature cartilage, outlined by ciliated columnar epithelium, consistent with a bronchogenic cyst. Patient follow-up at 1, 3, and 6-months later reported a Short Form 36 (SF-36) quality of life scale of 95% (minimal disability).

## Discussion

Bronchogenic cysts are rare congenital malformations of the anterior intestine<sup>5</sup>. This malformation can occur during the 3rd and 4th gestational week, when the primitive gut tube divides dorsally into the eventual esophagus, and ventrally into the eventual pulmonary yolk and tracheobronchial tree<sup>2</sup>. During development of an intrapericardial bronchogenic cyst, the heart and lungs share a common celomic cavity, thus the pulmonary yolk separation and its inclusion in the pericardial cavity can occur simultaneously<sup>5</sup>. Based on the location, they can be classified as parenchymal or mediastinal, the latter being more common in 86% of cases<sup>2</sup>. Additionally, they may also be classified as precarinal (52%), paratracheal (19%), paraesophageal (14%), and retrocardiac (9%)<sup>2</sup>. Occasionally, they may be present in other locations such as subcutaneous, cutaneous, neck and diaphragm, however the intrapericardial and intracardiac are extremely rare in the literature<sup>5</sup>. Their size often varies on average from 2–4 cm<sup>2</sup> [Table 1].

Table 1  
Comparison of Reports of Bronchogenic Cysts in the Literature.

Authors	Age (years)	Gender	Size (cm) L x W x H	Location	Treatment	Extracorporeal Circulation
Maldonado J et al.	66	Female	8 × 6 × 0.3	Pericardial	Resection	Yes
Qu X et al. <sup>5</sup>	1.6	Female	5.3 × 3.6 × 2.8	Pericardial	Resection	Not described
Li Z et al. <sup>9</sup>	17	Male	9 × 8.4	Left atrium	Resection	Yes
Olsen M et al. <sup>10</sup>	50	Female	3.4 × 3.3 × 4.1	Interatrial septum	Resection	Yes
Wang J et al. <sup>11</sup>	41	Female	2.5 × 1.5 × 2	Left ventricle	Resection	Yes
Borges AC et al. <sup>12</sup>	43	Female	4.4 × 3.4	Interatrial septum	Resection	Not described
Forcillo J et al. <sup>13</sup>	41	Female	1 × 1.7 × 1.2	Left ventricle	Resection	Yes
Nishida N et al. <sup>14</sup>	73	Male	0.5	Interventricular septum	Death	N/A

Often asymptomatic, in 19% of cases, the cysts are usually diagnosed as incidental findings on chest x-rays<sup>2,3</sup>. Typical symptoms may include retrosternal chest pain, dyspnea, coughing, stridor, fatigue, weakness, anorexia and fever<sup>2,5,3</sup>. Clinical manifestations depend on the localization of the mass, its size and whether it exerts any compression on adjacent structures<sup>5</sup>. If the cyst is small enough, avoiding any adjacent structures, it is usually asymptomatic. When the cyst is large, it usually produces coughing, chest pain, respiratory difficulty or even dysphagia. In adults, most cysts are very small reducing the chance of any ongoing symptoms<sup>1</sup>, however in this case report we show a large cyst generating important clinical manifestations, including heart failure and a cardiac arrhythmia.

A CT scan is used to identify location, size, shape, and its relation with adjacent structures, however it only accurately identifies these characteristics in 10–40% of patients<sup>7</sup>. Other patients often require additional studies such as an MRI, which has shown a higher diagnostic use given its high signal intensity in T2 images<sup>8</sup>. Additionally, the MRI can be useful for determining the origin of the cyst lesion, its relation with other structures and can even determine the best surgical approach<sup>7</sup>. In this case report, the chest CT scan was not enough to provide an adequate characterization of its walls and relation to adjacent structures, hence warranting a cardiac MRI.

The definitive diagnosis of a bronchogenic cyst is histopathological, once surgical resection is completed<sup>3</sup>. The typical findings are cysts with ciliated columnar epithelial cells along the inner lining<sup>8</sup>. The internal wall can also contain cartilage and smooth-muscle tissue, however these are not essential for the diagnosis. In this case, the cyst had ciliated columnar epithelial cells with fibromuscular tissue and mature cartilage of the inner wall, confirming the bronchogenic cyst diagnosis.

The definite treatment is surgical resection; this includes asymptomatic patients so as to avoid later complications such as infection, rupture or malignity<sup>2,5</sup>. Prognosis without treatment has a 100% mortality rate, which is reduced to 0–14% in surgical management<sup>2</sup>. In this case report, given the intrapericardial location, an extracorporeal circulation with bicaval aortic cannulation was required to ensure an adequate visualization and subsequent resection. In previous intracardiac and intrapericardial case report series, results have shown most of these cysts require extracorporeal circulation to undergo successful resection.

## Conclusions

Although numerous reports have been published regarding bronchogenic cysts, a large size such as the one reported in this case report, as well as its location, are extremely rare. In patients presenting with heart failure and atrial fibrillation, a mediastinal mass cannot be ruled out and should be considered within the differential diagnosis. The chest CT scan and cardiac MRI are excellent complementary images, however the definite diagnosis is histopathological examination upon surgical extraction.

## Abbreviations

Surgical Case Report Guidelines: SCARE

Emergency department: ED

Left ventricular ejection fraction: LVEF.

Computed tomography: CT.

Magnetic resonance image: MRI.

Intensive care unit: ICU.

## Declarations

**Ethics Approval and Consent to Participate by:** the Ethical and Research Committee of the Clínica Universitaria Colombia.

**Consent for Publication:** Written consent was obtained from the patient for publication of this report. Any details identifying the individuals to the clinical history and images associated were eliminated as to

remain anonymous.

**Availability of data and materials:** The dataset supporting the conclusions of this article is included within the article, any other inquiry is available from the corresponding author on reasonable request.

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**Author Contributions:** Maldonado Escalante JD designed the study and was the operating physician. Rincon FM, Molina G, Perez Rivera CJ and Acosta Buitrago LM collected and analyzed the data and wrote the paper. Maldonado Escalante JD, Rincon FM, and Molina G participated in the revision of the manuscript. All authors read and approved the final manuscript.

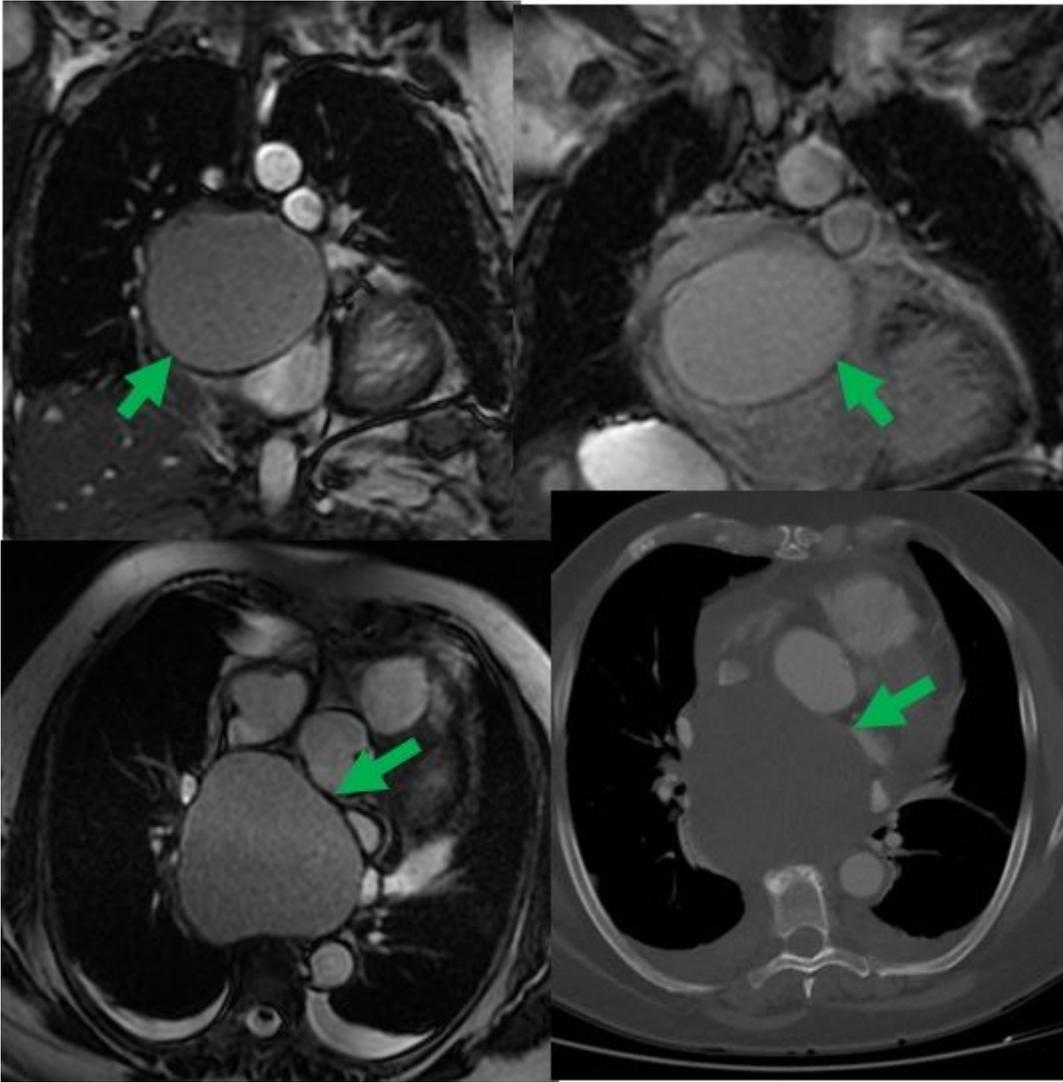
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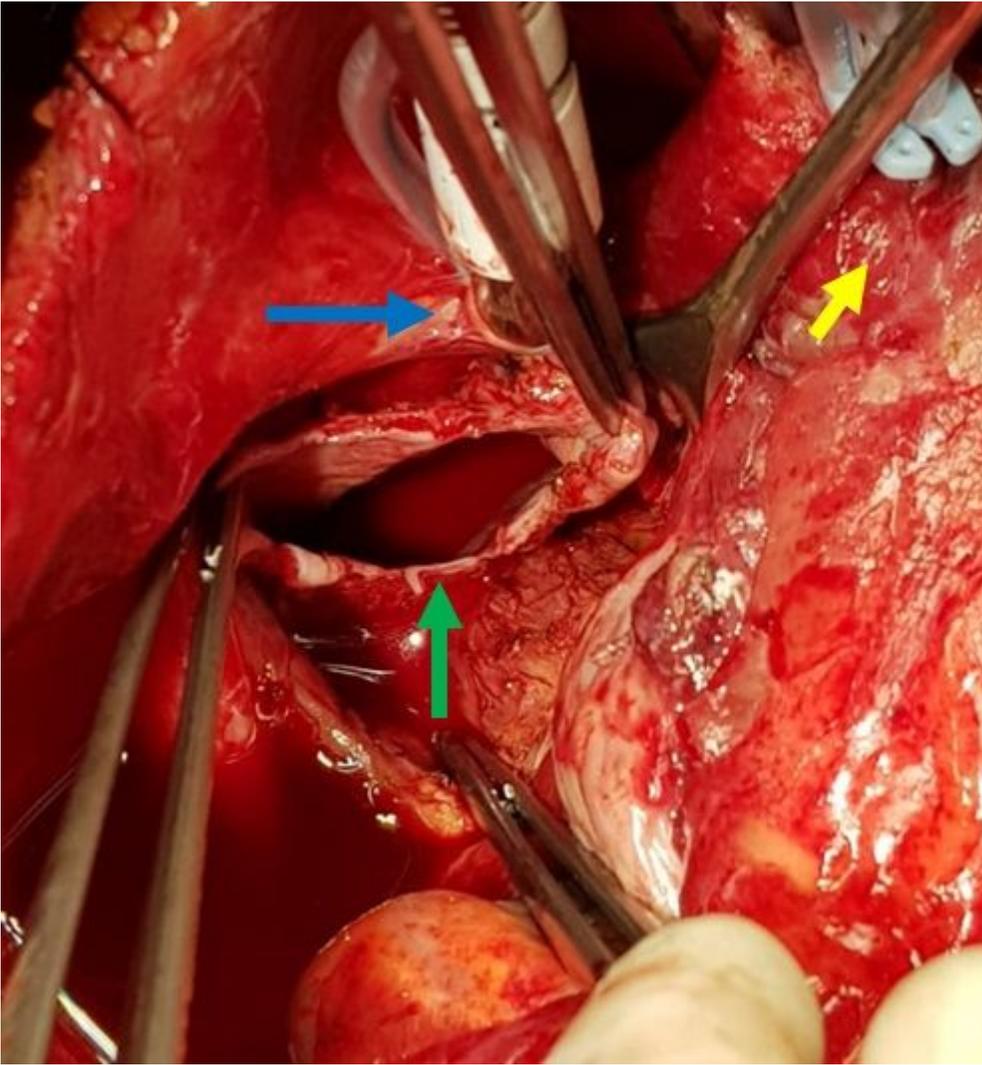
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## Figures



**Figure 1**

Cardiac MRI. Green arrows represent the dimensions and localization of the intrapericardial mass.



**Figure 2**

Intra-Operative Image. The open cyst (green arrow) showing the wall thickness, with the superior vena cava cannulation (blue arrow) and the aorta (yellow arrow).

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