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CASE REPORT
Other

Epicardial mesothelial cyst originating from the roof of the left atrium: a case report

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Background Epicardial mesothelial cysts are cysts that are attached to the epicardium within the pericardial cavity. Reports on epicardial mesothelial cysts are rare, and limited studies have investigated their surgical management. Here, we report the rare case of an epicardial cyst originating from the roof of the left atrium.

Case summary A 73-year-old man with dyspnoea on exertion and lower limb oedema was diagnosed with a giant cyst (diameter, 7 cm × 4.5 cm) in the pericardial cavity using computed tomography. Off-pump surgery was performed with successful resection of the cyst with a pedicle connected to the roof of the left atrium. Histological examination confirmed the mesothelial origin of the tumour cells.

Conclusion Cysts rarely develop within the pericardial cavity, especially an epicardial cyst. The few studies exploring this disease have suggested that patients with this condition may be asymptomatic or have mild breathlessness or cardiac tamponade, which might be occasionally or incidentally diagnosed. Sufficient preoperative evaluation, particularly involving the coronary artery, is essential, and a rational way of surgery should be planned considering all factors.

Keywords Epicardial cyst • Cardiac surgery • Off-pump technique • Case report

ESC Curriculum 2.4 Cardiac computed tomography • 6.6 Pericardial disease • 6.8 Cardiac tumours • 7.5 Cardiac surgery • 9.7 Adult congenital heart disease

Learning points

- We shared the management of the rare case of an epicardial cyst originating from the roof of the left atrium.
- Summary of the reported cases shows that symptoms vary from asymptomatic or mild breathlessness to cardiac tamponade, and they could be diagnosed incidentally or in emergency.
- Sufficient preoperative evaluations, especially of the coronary arteries, are needed, and a rational operation including on-pump, off-pump, or video-assisted thoracoscopic surgery should be based on an overall consideration of the patient.

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Introduction

Cysts rarely develop within the pericardial cavity. Those who do develop can be classified into pericardial and epicardial variants. The pericardial variant is a more common congenital abnormality (incidence of 1 in 100 000 individuals), accounting for 7% of mediastinal masses and 33% of mediastinal cysts.[1,2] Conversely, the epicardial variant—a mesothelial cyst attached to the epicardium surrounding the heart or great vessels within the pericardial sac[3]—is extremely rare. Data on its incidence are scarce. To date, only 16 patients have been reported to have epicardial mesothelial cysts.[3–17] In these reported cases, the cysts have been found to originate from the ventricle, right atrium, and great vessels at the bottom of the heart. Here we report the rare case of an epicardial cyst originating from the roof of the left atrium.

Timeline

Time	Events
6 June 2018	Cyst found in the pericardium on CT
12 November 2020	Symptoms appear, dyspnoea on exertion
11 December 2020	Hospitalization
23 December 2020	Epicardial cystectomy
3 January 2021	Hospital discharge
5 February 2021	First post-operative re-examination
6 April 2021	Second post-operative re-examination
9 July 2021	Third post-operative re-examination

Case presentation

A 73-year-old man was referred to our hospital with complaints of dyspnoea on exertion and lower limb swelling for >1 month. His cardiac functional capacity was classified as Class II as per the New York Heart Association classification. He was found to have a cyst (5 cm × 4 cm) inside his pericardial cavity near the right atrium on routine computed tomography (CT) 3.5 years before he presented to our hospital; the cyst was left untreated. The patient had a senile lacunar infarction with no obvious sequelae 4 years before presenting to the hospital. No other anamnesis was noted.

Physical examination revealed slightly dilated neck veins and mildly positive Kussmaul's sign. Blood test results revealed increased levels of high-sensitivity troponin-T at 48.52 (normal range, 0–14) pg/mL and N-terminal pro b-type natriuretic peptide at 1184 (normal range, 0–125) pg/mL. Other routine blood test and biochemistry results were within normal ranges. Electrocardiography indicated an atrial flutter and right bundle branch block.

Enhanced cardiac CT with electrocardiographic gating was performed on the day after admission to our hospital, which revealed that the cyst had grown in size (7 cm × 4.5 cm; *Figure 1A*). Transthoracic echocardiography revealed a large elliptical cystic dark area compressing the superior vena cava and right atrium (*Figure 2A*). No regional wall motion abnormalities were noted and left ventricular systolic function was preserved. Coronary angiography performed as a routine preoperative examination for older patients revealed diffuse slight stenosis (30%) and calcification of the left anterior descending branch as well as ostial stenosis (30%) of the left circumflex branch. No evidence of stenosis or compression was noted in the right coronary artery (*Figure 2B*). The patient was diagnosed as having a pericardial or epicardial cyst, which was believed to be the source of his symptoms. A traditional median sternotomy was planned with cardiopulmonary bypass (CPB) standby instead of video-assisted thoracoscopic surgery (VATS) because of the close association between the cyst and coronary artery.

The pericardial sac was opened carefully, revealing a giant elliptic cyst at the right atrioventricular sulcus compressing the right atrium and superior vena cava (*Figure 3A*). The cyst was well wrapped and did not adhere to the pericardium; thus, it was intraoperatively confirmed to be an epicardial cyst. After careful dissection, the cyst was found to be connected to the roof of the left atrium through a pedicle between the right auricle and the ascending aorta (*Figure 3B*). Approximately 60 mL of turbid yellowish fluid was drained from the cyst for further microbiological and cytological examinations. We then excised the shrunken cyst via sharp and blunt dissections without any complications. Macroscopic analyses indicated that the cyst was thin-walled and unilocular with an eroded inner face and no solid portion (*Figure 4*). The surgery was completed as per the usual protocol. The patient suffered from post-operative pneumonia, but his condition improved after antibiotic administration. He was discharged from the hospital on post-operative Day 10. He was followed-up three times post-operatively at 1, 3, and 6 months, and no abnormalities were found.

Pathological examination confirmed that the mass was an epicardial cyst. The cyst lumen was found to be lined by a flattened mesothelial cell monolayer (*Figure 4*). Microbial culture analysis of the pericardial effusion was negative, and no evidence of malignancy was noted.

Discussion

Pericardial cysts, which are similar to the epicardial cysts, have been reported in various studies since being first reported by Leroux in 1959.[18] Patients with pericardial cysts are mostly asymptomatic and incidentally diagnosed. In most cases, the mass was detected at the right or left cardiophrenic angle during radiological investigations as part of routine assessments for other illnesses.[19,20] With an increase in cyst size, symptoms may appear due to its compression against nearby structures such as the heart, great vessels, oesophagus, and trachea.[21] Different theories have been proposed for the aetiology of pericardial cysts. Lambert[22] suggested that such cysts embryologically originate from disconnected mesenchymal lacunae, which normally unite to form the pericardial coelom. However, Lillie *et al.*[23] determined that the origin of the pericardial cysts was based on the concept of differential persistence and graded the constriction of the ventral recess of the pericardial coelom.

Epicardial cysts are rarer than pericardial cysts. Differentially diagnosing these two variants from each other is difficult as they cannot be distinguished using CT or magnetic resonance imaging (MRI).

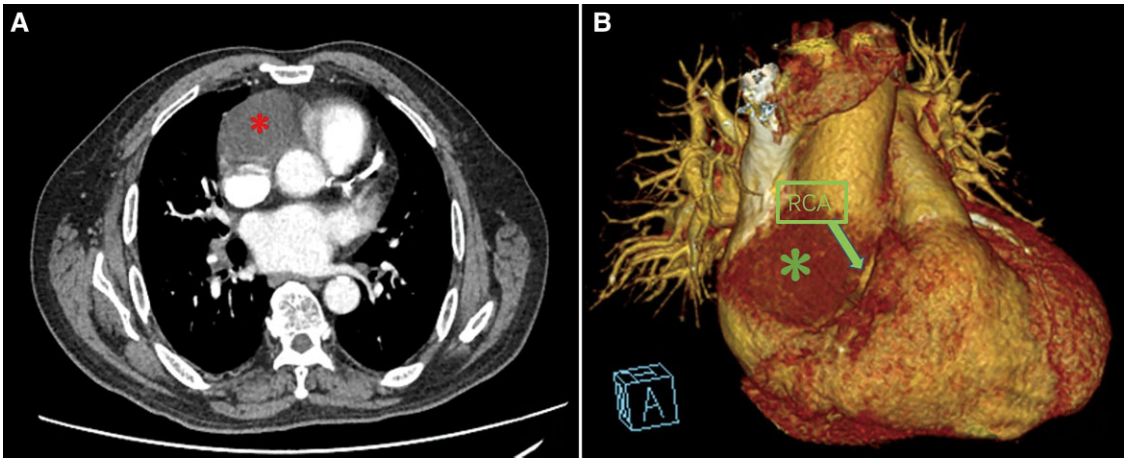


Figure 1 (*A*) Computed tomography of the chest showing cystic lesions (asterisk) and compression of the superior vena cava and right atrium; (*B*) the cyst located near the right coronary artery (asterisk).

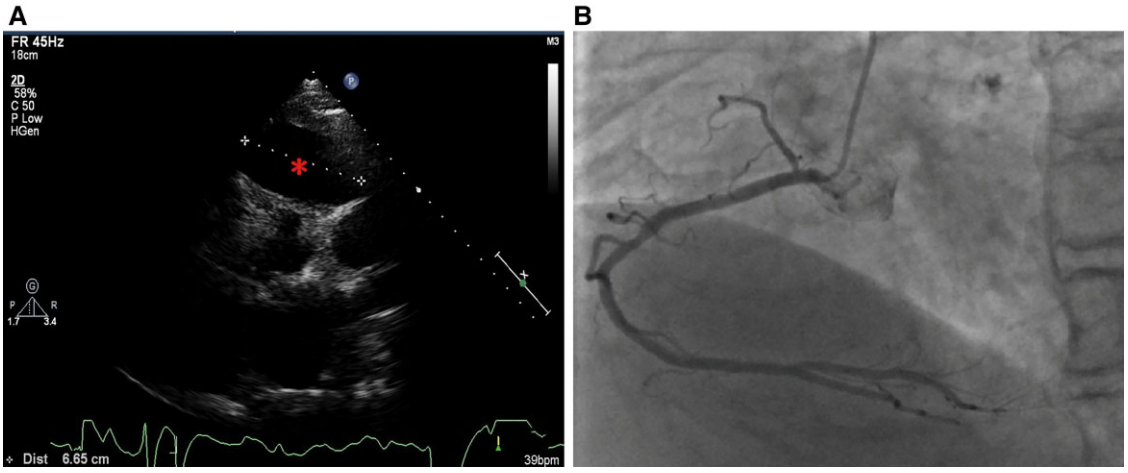


Figure 2 (*A*) Transthoracic echocardiography showing a large elliptic cystic dark area (asterisk) compressing the superior vena cava and right atrium; (*B*) coronary angiogram showing no evidence of stenosis or communication with the cyst on the right coronary artery.

A definite diagnosis of an epicardial cyst is always made intraoperatively. We have listed the data of all 16 cases of epicardial cysts reported to date in *Table 1*.

Symptoms of epicardial cysts range from no obvious symptoms, dyspnoea on exertion, shortness of breath, and fatigue to cardiac tamponade after chest trauma or infections such as acute abdomen and fever. In our case, the patient had dyspnoea on exertion and lower limb swelling, which might have been associated with an impaired diastolic function of the heart and elevated venous pressure owing to cyst compression. The epicardial cyst located at the right atrioventricular sulcus compressed the right atrium and superior vena cava, resulting in the congestion of the pericardial cavity. This restricted the diastolic function of the heart, which led to an increase in pressure in the vena cava with increased venous return volume, such as that observed after exercise or inhalation. This explains our patient's symptoms as well as the mild Kussmaul's sign noted on initial examination.

For pre-operative assessment, coronary CT angiography or coronary angiography should be considered based on the degree of adhesion and location of coronary vessels found on CT or MRI. Once the involvement of coronary vessels is confirmed, the need for coronary artery bypass grafting or CPB should be assessed intraoperatively. Of the 16 reported cases in the literature, CPB was performed intraoperatively for 6 patients. Of these patients, CPB was performed in two patients because of combined heart valve replacement. The same procedure was performed for the remaining

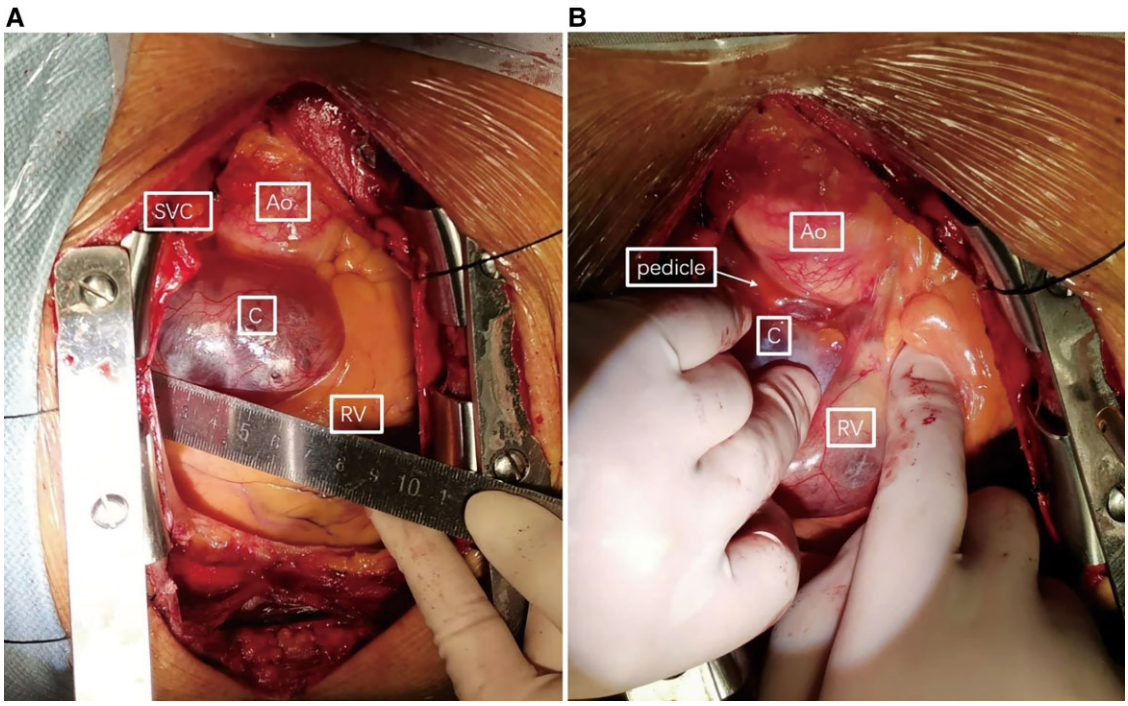


Figure 3 Intraoperative photograph after median sternotomy and opening the pericardium. (*A*) A giant elliptic cyst was observed at the right atrioventricular sulcus; it was not adhered to the pericardium. (*B*) The cyst was connected to the roof of the left atrium via a pedicle between the right auricle and ascending aorta. C, cyst; Ao, aortic artery; SVC, superior vena cava; RV, right ventricle.

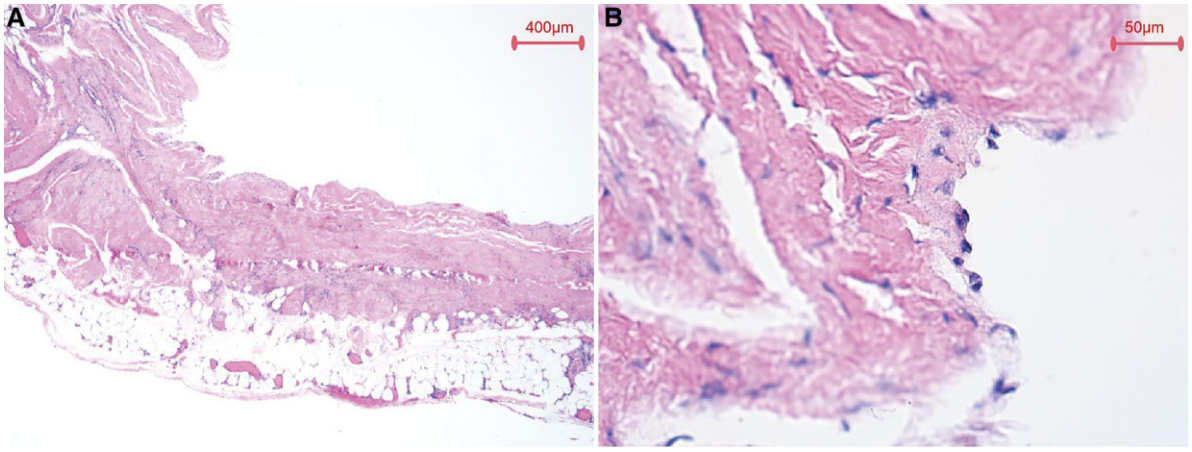


Figure 4 Pathological examination under low power field (*A*) and high power field (*B*) revealing that the cyst lumen was lined with a flattened mesothelial cell monolayer (haematoxylin–eosin staining).

four patients considering the close association between the cyst and coronary arteries or ventricles. The VATS has been reported as an alternative to median sternotomy for epicardial cysts because of its minimally invasive nature. However, when invasion into a vital structure is suspected on preoperative evaluation, median sternotomy should be performed without hesitation. Most of the patients in previously reported cases (13/16) underwent median sternotomy. Only one patient underwent left anterolateral thoracotomy, and two underwent VATS.

In our patient, preoperative CT revealed that the cyst was located above the right atrioventricular sulcus; therefore, median sternotomy with CPB standby was performed considering the patient's safety. The epicardial cyst was not adhered to the right coronary artery or ventricle wall. In hindsight, the cyst could have been resected

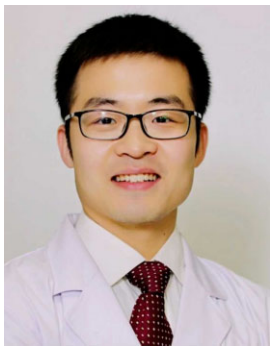
Table 1 Data of the 16 cases of epicardial cysts reported in the literature to date, including sizes, positions, and attachments of the cysts as well as the patient's symptoms and surgical methods use in the treatment

Report author/ year	Age at surgery	Gender	Symptoms	Position in the mediastinum	Attachment of the cyst	Size of the cyst (cm)	Surgery	Recurrence
Edwards/ 1972	43	M	Slight breathlessness for 2 years	Posterior	Posterior of left ventricle	Contain 500 mL fluid	Left anterolateral thoracotomy off-pump	No
Komeda/ 1985	7	F	No symptoms, found by radiography	Right anterolateral	Right atrium or ventricle	5.5 × 3.3	Median sternotomy off-pump	No
Ozasa/1991	64	F	No symptoms, found by echocardiography	Left anterolateral	Anterior of left ventricle	3 × 3	Median sternotomy off-pump	No
Debus/2001	8	F	Short syncope, impaired short-term memory	Posterior	Posterior of left ventricle	5 × 5	Median sternotomy on-pump	No
Scrofani/ 2002	60	M	Admitted for aortic valve insufficiency, slight dyspnoea	Right anterolateral	Upper portion of right atrium	7 × 5.5	Median sternotomy on-pump with aortic valve replacement	No
Omeroglu/ 2004	30	F	Admitted for severe aortic and mitral valve insufficiency, dyspnoea on exertion, fatigue, and palpitations	Left anterolateral	Anterior of left ventricle	5×4×2.5	Median sternotomy on-pump with aortic and mitral valve replacement	No
Buyukates/ 2008	20	M	Swelling in both legs for 1 year, dyspnoea on exertion	Right anterolateral	Anterior and inferior of right ventricle	6 × 2	Median sternotomy off-pump	No
Hatemi/ 2012	50	F	Dyspnoea and persistent cough for 1 week	Anterior, multiple lesions	Left ventricle over the LAD	2.6 × 2.5 × 2.8	Median sternotomy off-pump	No
	17	F	Emergency clinic for shortness of breath and fatigue	Left posterolateral	Left ventricle over the LCX	8.5 × 3.5	Median sternotomy on-pump	No
Joo Yeon Kim/2013	64	F	No symptoms, found by radiography	Left anterolateral	Anterior of right ventricle over the LAD	5.5 × 5 × 2.8	Median sternotomy on-pump	No
Suenaga/ 2015	76	M	Admitted for CABG, found by radiography	Anterosuperior	Ascending aorta	5.5 × 4	Median sternotomy on-pump with CABG	No
Masuoka/ 2015	2	M	Cardiac tamponade after chest trauma	Right anterolateral	Cephalic aspect of left atrium	8.9 × 7 × 4	Median sternotomy off-pump	No
Dribin/2016	3	M	Acute abdomen, fever	Right anterolateral	Acute margin of right ventricle	5 × 4	Median sternotomy off-pump	No
Yong Hwan Kim/2017	57	M	Progressive exertional dyspnoea	Left anterolateral	Left ventricle	8 × 4	VATS	No
Kaneyuki/ 2018	73	F	Dyspnoea on exertion	Left anterolateral	Pulmonary artery	12 × 10	Median sternotomy off-pump	No
Ding/2020	7	M	Increased heart rate and dyspnoea	Left posterolateral	Left ventricle	4.5 × 5.1 × 4.6	VATS	No

M, male; F, female; CABG, coronary artery bypass grafting; LAD, left anterior descending coronary artery; LCX, left circumflex coronary artery; VATS, video-assisted thoracoscopic surgery.

via VATS without any challenges if the surgery had been performed when the cyst was first detected 3.5 years ago. However, considering the disease progression, the less invasive nature of VATS was not considered sufficiently beneficial to outweigh the risks associated with increased surgical complexity. Ultimately, we found that the cyst was connected to the roof of the left atrium.

Lead author biography



Lei Xu, MD, graduated from the Cheeloo College of Medicine, Shandong University of China, in 2015, participated in a research programme on cardiology in the University of North Carolina at Chapel Hill during 2012–14, and now worked as an attending doctor at the Department of Cardiac Surgery, Shandong Provincial Hospital Affiliated to Shandong First Medical University, Shandong, China.

Supplementary material

Supplementary material is available at *European Heart Journal – Case Reports* online.

Slide set: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary material.

Consent: The authors confirm that written informed consent for submission and publication of this case report including images and associated text has been obtained from the patient in accordance with the Committee on Publication Ethics guidelines.

Conflict of interest: None declared.

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