

Multiple lipomata of the tricuspid valve and papillary muscle: case report

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Received 4 December 2020; first decision 8 January 2021; accepted 8 June 2021

Background

Cardiac lipomas are rare benign primary tumours of the heart. Due to the nature of these tumours, they are often asymptomatic and diagnosed incidentally. Whether asymptomatic patients with cardiac lipomas should perform surgery still remains controversial.

Case summary

A 34-year-old Asian male who was incidentally found hyperechoic masses in the right ventricle (RV) on the transthoracic echocardiogram by annually routine physical examination was admitted to our cardiology department. His medical history was unremarkable. The repeated transthoracic and transoesophageal echocardiogram showed multiple solitary and well-demarcated masses in the RV. On the cardiac magnetic resonance imaging, four discrete masses (considering the possibility of it being a lipoma) partially occluding the right ventricular outflow tract (RVOT) were observed. During the open-heart resection surgery, it was found that the tricuspid valve and papillary muscle were covered by multiple adipose masses in the RV that arose from the interventricular septum and the free wall, resulting in partial RVOT obstruction. These excised masses were histopathologically confirmed as lipomata characterized by the mature adipocytes with entrapped myocardial cells. The patient had no cardiac abnormality in the 1-month follow-up after the surgery.

Discussion

This rare clinical case of multiple lipomata of the tricuspid valve and papillary muscle acknowledges that multimodality imaging is the cornerstone for the assessment and diagnosis. Surgery should be performed in cases of symptomatic or large lipomas as well as when a lipoma is considered to be high risk because of RVOT obstruction.

Keywords

Multiple lipomata • Cardiac magnetic resonance • Echocardiogram • Case report

Learning points

- Multimodality imaging is the cornerstone for the assessment and diagnosis of cardiac multiple lipomata.
- Surgery should be performed in cases of symptomatic or large lipomas as well as when a lipoma shows signs of right ventricular outflow tract obstruction and when there is felt to be high risk of thromboembolic sequelae.

Handling Editor: Mark Abela

Peer-reviewers: Luca Arcari and Stefano Albani Compliance Editor: Brett Sydney Bernstein Supplementary Material Editor: Katharine Kott

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Introduction

Primary cardiac neoplasms in the heart are rare. A meta-analysis of 731 309 autopsies from 22 studies showed the incidence was only 0.02%. Adult cardiac benign tumours are mainly myxomas, followed by lipomas. The sporadic cases of lipomas account for about 2.4-8.4% of patients with cardiac benign tumours.² As with other cardiac tumours, whether presenting symptoms such as atrial and ventricular arrhythmias, atrioventricular block, and heart failure are associated with tumour location. More frequently, they are discovered incidentally on imaging, during cardiac surgery, or at autopsy. Multimodality imaging with transthoracic, transoesophageal echocardiography, myocardial contrast, three-dimensional echocardiography, computed tomography, and magnetic resonance imaging are of great help in the diagnosis of cardiac tumours.⁴ Here, we report a rare case with multiple lipomata, located in the tricuspid valve and papillary muscle of the right ventricle (RV), which was successfully removed by surgery and the patient recovered well after surgery. Through transoesophageal echocardiogram and cardiac magnetic resonance (CMR) imaging, four discrete masses (considering the possibility of it being a lipoma) were observed. By reporting this case, we aimed to highlight the diagnostic role of echocardiography and CMR imaging in patients with multiple lipomata, and promote the management of this disease.

Timeline

Time	Event
1 day	Chief complaint of finding hyperechoic masses in the right ventricle (RV) incidentally on routine transthoracic echocardiogram a month ago.
3 days	The repeated transthoracic and transoesophageal echo- cardiogram showed multiple solitary and well-demar- cated masses in the RV.
4 days	On cardiac magnetic resonance imaging, four discrete masses (considering the possibility of lipomata) were observed. During the open-heart surgery, it was found that the tricuspid valve and papillary muscle was covered by multiple adipose masses in the RV that arose from the interventricular septum and the free wall, partially occluding the right ventricular outflow tract (RVOT). Surgical resection was performed to obtain a suitable route to the RVOT though its potential for malignancy was low. These excised masses were histo pathologically confirmed as lipomata characterized by the mature adipocytes with entrapped myocardial
1 month	cells. He had no complaints and his echocardiogram was normal.

Case presentation

A 34-year-old Asian male with hyperechoic masses in the RV found incidentally on the transthoracic echocardiogram during a routine



Video I The transthoracic echocardiography before surgery.

physical examination was admitted to our cardiology department. There was no past medical history of coronary artery disease, cardiomyopathy, heart failure, dysrhythmia, sudden death, or autoimmune disease. On admission, the physical examination, vital signs, and laboratory profile were normal, and the patient had a normal sinus rhythm.

The repeated transthoracic echocardiogram showed three solitary and well-demarcated masses in the RV (Supplementary material online, $Table\ S1,\ Video\ 1$). The size of these three lumps of poplar homogeneous isoechoic shadows were about $23\ \text{mm}\times 17\ \text{mm}$, $12\ \text{mm}\times 33\ \text{mm}$, and $21\ \text{mm}\times 25\ \text{mm}$, respectively. The boundaries were clear and their internal echo was similar to that of point-like particles. The residual cardiac structure (including pericardial thickness), intracardiac flow, and left ventricular function (ejection fraction 65%) were unremarkable. The preliminary diagnosis was multiple solitary and well-demarcated masses in the pericardium (considering the possibility of lipoma or fibrinous cystadenoma).

The computed tomography also demonstrated multiple circular low-density shadows near the interventricular septum in the RV with a mean value of -56 HU (*Figure 1*). The preliminary consideration was lipomas. It was not possible to identify the degree of subvalvular apparatus involvement.

Given these findings, CMR was performed for better tissue characterization. CMR demonstrated four irregular but well-circumscribed masses (the largest size being $20.5\,\mathrm{mm}\times13.6\mathrm{m}\times15.7\,\mathrm{mm}$). Two bases were attached to the interventricular septum and arranged up and down, the other two were attached to the lower wall of the RV (Figure 1). The CMR diagnosis was intracardiac partially occluding the right ventricular outflow tract (RVOT) (Figure 2, Video 2), and similar findings were found on the echocardiography (Figure 2). The T1-weighted image revealed a hyperintense signal and was uniformly suppressed in the fat-suppression mode, but an unusual malignant tumour with high-fat content could not be excluded (Figure 3, Video 3).

Considering the age of the patient and the concrete possibility of the future development of significant RVOT obstruction, the surgical treatment was recommended. Surgical excision was performed to avoid potentially fatal embolism and obtain a definitive diagnosis,

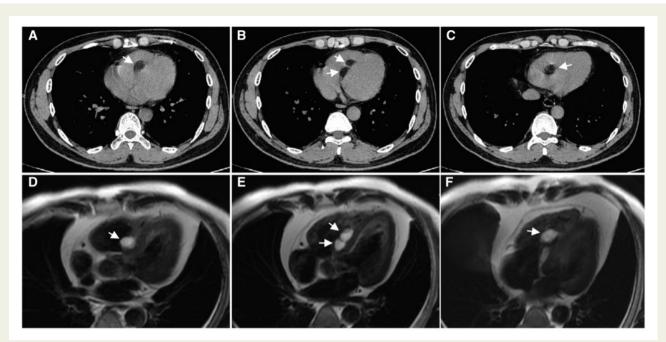


Figure 1 Four circular low-density shadows (A–C, white arrows) near the interventricular septum in the right ventricle with the mean value of -56 HU, which indicate fat density. Four irregular, well-circumscribed masses (D–F, white arrows), the largest size being 20.5 mm × 13.6 mm × 15.7 mm.

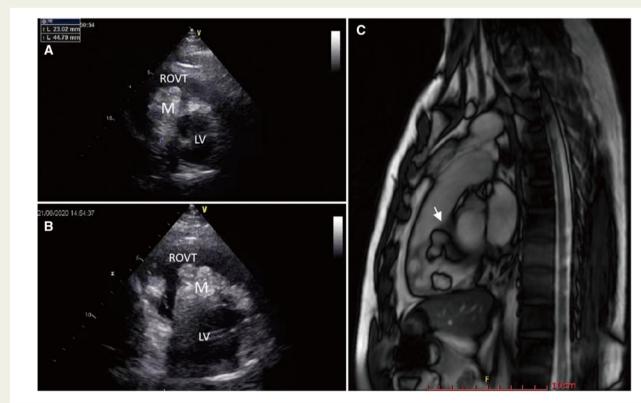


Figure 2 Both echocardiography (A and B) and cardiac magnetic resonance (C, white arrows) showed that the mass partially occluded the right ventricular outflow tract. LV, left ventricular; M, masses; RVOT, right ventricular outflow tract.

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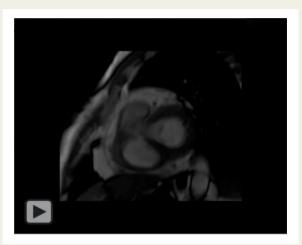
excluding the small possibility of malignancy. Thus, an operation involving tricuspid valve repair and resection of the tumour was electively planned.

Once the patient was intubated, a transoesophageal echocardiography was performed (Supplementary material online, Video S1). The operation was carried out through a median sternotomy with standard hypothermic cardiopulmonary bypass. After opening the pericardium, the masses were seen. Through the right atrium incision, the tricuspid valve and papillary muscle were covered by four adipose masses in the RV that arose from the interventricular septum and the

tricuspid chordae tendineae, which partially occluding the RVOT (Figure 4, white asterisks). These multiple yellowish masses partly merged with each other, integrated with packs of chordae and papillary muscle of the tricuspid septal leaflet, as well as the right interventricular septum. While trying to maintain the integrity of papillary muscles and tricuspid valve chords, a part of the chordae tendineae was severed and four masses were fully exfoliated (Figure 5). After that, saline was injected into the RV and severe tricuspid valve insufficiency was found. The severed chordae tendineae were sutured to the papillary muscle using a prolene 4–0 suture for two stitches.



Video 2 The CMR diagnosis was intracardiac partially occluding the RVOT.



Video 3 The T1-weighted image and fat-suppression mode.

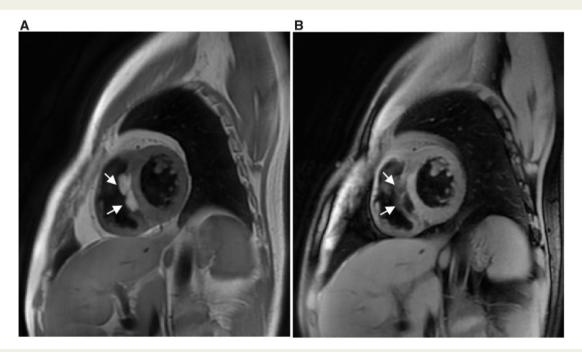


Figure 3 The masses revealed a hyperintense signal in the T1-weighted image and was uniformly suppressed in the fat-suppression mode (A and B, white arrows).

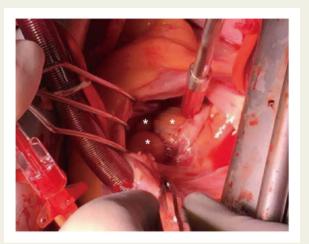


Figure 4 Surgical resection showed that the tricuspid valve and papillary muscle were covered by four adipose masses (white asterisks) in the right ventricle.



Figure 5 Four masses were surgically removed.

Again, it was found that the tricuspid closure improved and only with a small amount of reflux. After an aortic cross-clamp time of 3 h, the patient was weaned from cardiopulmonary bypass. A transthoracic echocardiogram was performed 1 day after surgery (Supplementary material online, *Table S1*). The S', peak systolic tissue velocity at the tricuspid annulus was 8.6 cm/s, and there was no sign of tricuspid regurgitation or RVOT obstruction, which indicated that the patient had an uneventful in-hospital recovery without any signs of right-sided cardiac failure. The macroscopic appearance of the four ovoid fat masses was histopathologically confirmed as lipomas, characterized by the mature adipocytes with entrapped myocardial cells. No atypical features or malignancy were identified (*Figure 6*).

The patient had no complaints and his echocardiogram was unremarkable in the scheduled 1-month follow-up after the surgery.

Discussion

The rare clinical case of the multiple lipomata of the RV is given. Hyperechoic masses in the RV were found accidentally on the transthoracic echocardiogram during an annual routine physical examination. This case is special because there are rarely previous reports describing multiple cardiac lipomata of the RV of the heart.

Cardiac lipomas with well-encapsulated tumours composed of mature fat cells are rare, accounting for 2–8% of all benign cardiac tumours, most commonly found in the subendocardial region (approximately 50%). They show a predilection for the right atrium and left ventricle.³ The aetiology and natural pathogenetic process of cardiac lipomas remain undetermined. There is no difference in the distribution of cardiac lipoma between genders, but it may be more common between 40 and 70 years old.⁵ In fact, Lang-Lazdunski et al.⁶ reported a huge epicardial lipoma weighing 4850 g that was diagnosed only at autopsy. In the present case, approximately four multiple adipose masses covered the tricuspid valve and papillary muscle

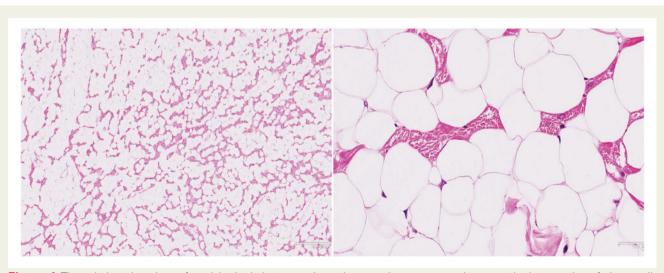


Figure 6 The pathological results confirmed that both the intracardiac and extracardiac masses were lipomas with a large number of adipose cells and little fibrous tissue.

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in the RV but the patient was asymptomatic. The symptoms of cardiac lipomas depend on the tumour location and size.⁷ Improvements in cardiac imaging techniques have made an increased detection of even small tumour masses in both symptomatic and asymptomatic patients. Echocardiography is of great significance for preoperative evaluation, intraoperative assessment of aortic valve dysfunction, and postoperative follow-up.8 Both cardiac computed tomography and CMR can help identify fat with a high degree of specificity and therefore should be used to diagnose cardiac lipomas without equivocation. Typically in CMR, the lipoma has a homogenous high signal intensity in both T1- and T2-weighted images when compared with the myocardium. Of note, the black boundary sign in the cine sequence due to the chemical shift effect is of great help in diagnosing lipomas. 10 Although CMR cannot definitively exclude malignancy, this case had strong features of a benign aetiology (well-circumscribed mass; no invasion across tissue planes nor pericardial effusion and with typical features of a lipoma as outlined above) that made the diagnosis almost certain with imaging alone.

Due to the low prevalence, there is no randomized clinical trial to guide the treatment. A consensus about surgical treatment in symptomatic patients with cardiac lipoma is clear, but it is still a controversial issue whether to perform surgery in asymptomatic patients with cardiac lipoma is beneficial. In this case, we described multiple lipomata in the RV that arose from the interventricular septum and the tricuspid chordae tendineae partially occluding the RVOT. To our knowledge, this is much more multiple lipomata if not the most, compared to those previously reported. Due to the low prevalence of this unusual tumour, it is likely that few cardiologists can encounter this condition. By reporting this case, we aimed to highlight the diagnostic role of echocardiography and CMR imaging in patients with multiple lipomata and the indication of resection surgery, promoting the management of this disease.

Lead author biography



Yang Yan, MD, Professor, Chief of Cardiovascular Surgery. Engaged in cardiovascular surgery medical, teaching, and research work. His group has carried out a number of complicated congenital heart disease operations for infants and young children, minimally invasive cardiac surgery, and coronary artery bypass grafting under laparoscopy, and successfully imple-

mented cardiac mechanical auxiliary (VAD, ECMO) treatment for critically ill patients.

Supplementary material

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

Acknowledgements

We thank the Biobank of First Affiliated Hospital of Xi'an Jiaotong University for providing clinical data.

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

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: The authors declare that there is no conflict of interest.

Funding: This work was supported by the Key Research and Development Program of Shaanxi province (2020ZDLSF04-11).

Reference

- Reynen K. Frequency of primary tumors of the heart. Am J Cardiol 1996;77: 107–107
- Fang L, He L, Chen Y, Xie M, Wang J. Infiltrating lipoma of the right ventricle involving the interventricular septum and tricuspid valve. *Medicine* 2016;95: e2561.
- Maleszewski JJ, Bois MC, Bois JP, Young PM, Stulak JM, Klarich KW. Neoplasia and the heart pathological review of effects with clinical and radiological correlation. J Am Coll Cardiol 2018:72:202–227
- Cao S, Tan T, Zhou Y, Zhou Q. Giant left ventricular infiltrating lipoma one or two? Circ Cardiovasc Imaging 2019;12:e009361.
- Shu S, Wang J, Zheng C. From pathogenesis to treatment, a systemic review of cardiac lipoma. J Cardiothorac Surg 2021;16:1.
- Lang-Lazdunski L, Oroudji M, Pansard Y, Vissuzaine C, Hvass U. Successful resection of giant intrapericardial lipoma. Ann Thorac Surg 1994;58:238–240.
- Schrepfer S, Deuse T, Detter C, Treede H, Koops A, Boehm DH et al. Successful resection of a symptomatic right ventricular lipoma. *Ann Thorac Surg* 2003;76:1305–1307.
- Liu L, Zuo Y, Huang Y, Cao L. Echocardiographical findings of giant cardiac lipoma: a case report. Medicine 2019:98:e14456.
- Araoz PA, Mulvagh SL, Tazelaar HD, Julsrud PR, Breen JF. CT and MR imaging of benign primary cardiac neoplasms with echocardiographic correlation. Radiographics 2000;20:1303–1319.
- Wijesurendra RS, Sheppard KA, Westaby S, Ormerod O, Myerson SG. The many faces of cardiac lipoma-an egg in the heart. Eur Heart J Cardiovasc Imaging 2017;18:821–821.
- Wu S, Teng P, Zhou Y, Ni Y. A rare case report of giant epicardial lipoma compressing the right atrium with septal enhancement. J Cardiothorac Surg 2015;10:150.