#### CASE REPORT

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# A huge right ventricular hydatid cyst related to tricuspid valve causing severe pulmonary hypertension

# Lubna Bakr 🕩 🕴 Somar Alnajar

Department of Cardiac Surgery, Faculty of Medicine, Damascus University, Damascus, Syria

#### Correspondence

Lubna Bakr, Department of Cardiac Surgery, Faculty of Medicine, Damascus University, Damascus, Syria. Email: lubna-bakr@hotmail.com

## Abstract

Preserving tricuspid valve's function is the cornerstone in surgical excision of cardiac hydatid cysts present in the right side of the heart. The perfect surgical technique may not exist. However, further studies are needed to ensure the clear path.

#### **KEYWORDS**

cardiac surgery, hydatid cyst, right ventricle, severe pulmonary hypertension, tricuspid regurgitation

# **1** | INTRODUCTION

Preserving the valve's function is the cornerstone in surgical excision of cardiac hydatid cysts. The perfect surgical technique may not exist. This report highlights the need for further studies to ensure the clear path in approaching such cases.

Cardiac hydatid cysts are rare even in endemic countries. They pose a therapeutic challenge due to varying presentation and unpredictable complications.<sup>1</sup> Echinococcosis is a parasitic infection caused by Echinococcus granulosus.<sup>2</sup> In humans, the disease is characterized by development of three-layered cysts. The cysts develop primarily in the liver and the lungs. Cardiac involvement is very uncommon, only about 0.01%-2% of all cases.<sup>3</sup> Although it might be asymptomatic, it warrants early surgical repair because cyst rupture is potentially fatal.<sup>4</sup> Excision of cardiac cysts under cardiopulmonary bypass (CPB) is the treatment of choice for cardiac hydatid disease.<sup>5</sup> In our case, the hydatid cyst was related to the tricuspid valve in a way that required tricuspid valve repair. It also caused severe pulmonary hypertension that was relieved soon after the surgery.

# 2 | CASE REPORT

A 37-year-old housewife presented to our hospital with chest pain that started two months previously, along with having a nonspecific productive cough.

The contrast-enhanced computed tomography (CT) scan showed multiple small hydatid cysts in both lungs, along with a large hydatid cyst within the right ventricle (Figure 1). The transthoracic echocardiography revealed a large cystic mass measuring 4.3 cm × 2.9 cm occupying 40%-50% of the right ventricle cavity and the outflow tract, fixed to the interventricular septum. Multiple daughter cysts could be seen within it, suggesting a hydatid cyst. The systolic pulmonary artery pressure (SPAP) was 60 mm Hg, tricuspid regurgitation velocity was 3.5 m/s. The diagnosis of right ventricular hydatid cyst was established. No liver cysts were seen on the CT. The head CT scan showed no brain abnormality. The patient was started on albendazole a month before the surgery, and it was continued intraoperatively and postoperatively.

The surgery was performed under CPB (Video S1). The surgical field was walled off with sponges soaked in hypertonic saline. After the heart was arrested, the right ventricle was opened through a vertical right ventriculotomy.

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FIGURE 1 The preoperative chest CT images showing the hydatid cyst within the right ventricle

A trans-atrial approach was not used as it would not provide adequate exposure. Additionally, the anterior wall of the right ventricle was thin to a degree that the cyst was almost visible. A huge cyst of about  $4.5 \times 3$  cm was seen strongly attached to the right ventricular free wall (Figure 2A), and the anterior papillary muscle (Figure 2B). With no aspiration, the cyst was completely excised (Figure 3). However, the chordae tendineae of the anterior tricuspid leaflet became unattached. They were reattached with the papillary muscle using polyester sutures supported with pledgets of PTFE felt (Figure 2C). The cyst was in the anterior papillary muscle, and was far from the pulmonary valve. The right ventricular outflow tract (RVOT) was intact, and so was the pulmonary valve. The right ventriculotomy was closed with continuous stitches of 3-0 polypropylene over two strips of PTFE felt. A right atriotomy was carried, and the DeVega repair technique was performed. After separation from CPB, sinus rhythm was automatically regained without pacing or vasoactive drug support.

The patient's postoperative course was uneventful. She underwent transthoracic echocardiography four days after surgery, which revealed mild tricuspid valve regurgitation, and SPAP of 35 mm Hg. She was discharged home and followed up on albendazole therapy. The postoperative histopathology confirmed the diagnosis.



**FIGURE 2** The intraoperative view. (A) Right ventricle was opened, a huge cyst attached to the right ventricular free wall. (B) The cyst in the anterior papillary muscle. (C) Chordae tendineae reattached using polyester sutures

**FIGURE 3** The macroscopic aspect of the cardiac hydatid cyst (A) exteriorly, (B) interiorly, showing multiple daughter small cysts



# **3** | **DISCUSSION**

Among hydatid disease cases of cardiac involvement, the left ventricle is the most frequently involved (55%-60%).<sup>5</sup> In our case, the hydatid cyst was of remarkable size, seldom occupying the right ventricle cavity and outflow tract. This involvement is unusual since the patient did not undergo any previous surgeries.

Although pulmonary hypertension could be thromboembolic due to the presence of the cyst within the heart,<sup>5</sup> no thrombi were identified in our case. Such significant pulmonary hypertension was reported in another case with a much smaller hydatid cyst measuring 2 cm  $\times$  2 cm with no detected pulmonary thromboembolism.<sup>6</sup> Pulmonary embolism due to hydatid cysts is a very rare clinical entity.<sup>7</sup> However, it was reported previously in the literature.<sup>7,8</sup> The patient had neither dyspnea nor syncope, and her vital signs were normal. Therefore, no additional investigations were carried out.

Cardiac hydatid cysts with intracavitary expansion should be treated surgically without delay. A computerized review of cardiac hydatid cysts published in the literature in 1990, revealed mortality of 23.47%.9 Right ventricular hydatid cysts are different from left ventricular cysts. They tend to expand inside the right ventricular cavity and in the subendocardial space, and are more prone to rupture which can lead to pulmonary embolism, anaphylaxis and sudden death. Left ventricular hydatid cysts grow as subepicardial masses, and rarely rupture in the pericardial space.<sup>10</sup> Gentle handling of the heart during cardiopulmonary bypass minimizes operative risk. All patients should be investigated for systemic cysts,<sup>9</sup> and this was done in our case. The surgical technique includes cavity cleaning and closure using multiple purse string sutures, or partial resection and ventricular patch plasty after removal of the cystic material.<sup>9</sup> In our case, complete resection was performed.

Many surgeons conduct aspiration of the cyst content. However, we avoided that due to the fear of spillage and subsequent reimplantation, the deadly anaphylactic shock which was reported in the literature previously, and the uncertain effect of hypertonic saline on cardiac tissue. As aspiration was not performed, preserving the cardiac muscle and the valve's function was a true challenge.

We used DeVega repair technique, as we were operating on what was left of the tricuspid valve. It was reported that DeVega annuloplasty successfully treated tricuspid regurgitation, and preserved normal tricuspid annular dynamics and geometry.<sup>11</sup>

The perfect surgical technique for excising cardiac hydatid cysts may not exist. However, further studies are needed to ensure the clear path.

#### ACKNOWLEDGEMENTS

Informed consent was obtained from the participant included in the study.

# **CONFLICT OF INTEREST**

None declared.

## AUTHOR CONTRIBUTIONS

LB: involved in conceptualization, data curation, formal analysis, investigation, project administration, resources, software, supervision, writing the original draft, review and editing, and was the first assistant in the surgery. SA: involved in conceptualization, supervision, and performed the surgery.

## ETHICAL APPROVAL

All procedures performed in this study were in accordance with the ethical standards of Damascus University Research Ethics Committee and with the 1964 Helsinki Declaration and its later amendments.

## DATA AVAILABILITY STATEMENT

The authors declare that the data supporting the findings of this study are available within the article and its supplementary information files (Video S1).

# ORCID

1856

Lubna Bakr D https://orcid.org/0000-0003-4766-7913

# REFERENCES

- Kothari J, Lakhia K, Solanki P, et al. Invasive pericardial hydatid cyst: Excision of multiple huge cysts. *J Saudi Heart Assoc*. 2017;29(1):53-56. https://doi.org/10.1016/j.jsha.2016.06.005
- Huerta-Obando AV, Olivera-Baca EY, Silva-Díaz J, Salazar-Díaz A. Cardiac hydatid cyst in a child: a case report. *Rev Peru Med Exp Salud Publica*. 2018;35(2):338-343. https://doi.org/10.17843/ rpmesp.2018.352.3258
- Gencheva DG, Menchev DN, Penchev DK, Tokmakova MP. An incidental finding of heart echinococcosis in a patient with infective endocarditis: a case report. *Folia Med (Plovdiv)*. 2017;59(1):110-113. https://doi.org/10.1515/folmed-2017-0017
- Kumar A, Ballal P, Nagamani AC, Sheriff SA. Surgical excision of an epicardial ventricular hydatid cyst. *Asian Cardiovasc Thorac Ann.* 2020;28(5):273-275. https://doi.org/10.1177/0218492320 927200
- Orhan G, Bastopcu M, Aydemir B, Ersoz MS. Intracardiac and pulmonary artery hydatidosis causing thromboembolic pulmonary hypertension. *Eur J Cardio-Thorac Surg.* 2018;53(3):689-690. https://doi.org/10.1093/ejcts/ezx330
- Kurdal AT, Kahraman N, Iskesen I, Sirin BH. Unusual location of hydatid cyst: the posterior leaflet of tricuspid valve. *Ann Ital Chir*. 2010;81(3):211-214.
- 7. Poyraz N, Demirbaş S, Korkmaz C, Uzun K. Pulmonary embolism originating from a hepatic hydatid cyst ruptured into the inferior

vena cava: CT and MRI findings. *Case Rep Radiol*. 2016;2016:1-4. https://doi.org/10.1155/2016/3589812

- Leila A, Laroussi L, Abdennadher M, Msaad S, Frikha I, Kammoun S. A cardiac hydatid cyst underlying pulmonary embolism: a case report. *Pan Afr Med J.* 2011;8:12.
- Younis SN, Faraj AA. Cardiac hydatid disease, case report, and review of literature. *Acta Clin Belg*. 2014;69(1):66-68. https://doi. org/10.1179/0001551213Z.0000000003
- Kankilic N, Aydin MS, Günendi T, Göz M. Unusual hydatid cysts: cardiac and pelvic-ilio femoral hydatid cyst case reports and literature review. *Braz J Cardiovasc Surg.* 2020;35(4):565-572. https:// doi.org/10.21470/1678-9741-2019-0153
- Malinowski M, Schubert H, Wodarek J, et al. Tricuspid annular geometry and strain after suture annuloplasty in acute ovine right heart failure. *Ann Thorac Surg.* 2018;106(6):1804-1811. https:// doi.org/10.1016/j.athoracsur.2018.05.057

# SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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