



Case Report

Cardiac hydatid cyst in the right ventricle: An unusual case at a rare site

Nouradden N. Aljaber, MD^a, Sultan A. Alshoabi, MD^{b,*}, Abdulaziz A. Qurashi, PhD^b
and Tareef S. Daqqaq, MD^c

^a Department of Cardiology, Faculty of Medicine, Sana'a University, Sana'a, Republic of Yemen

^b Department of Diagnostic Radiology Technology, College of Applied Medical Sciences, Taibah University, Almadinah Almunawwarah, KSA

^c Department of Radiology, Faculty of Medicine, Taibah University, Almadinah Almunawwarah, KSA

Received 14 December 2019; revised 14 February 2020; accepted 19 February 2020; Available online 25 March 2020



المخلص

الكيسة العدارية هي عدوى طفيلية بالطور اليرقي للديدان الشريطية المشوكة الحبيبية. عادة تصيب الكبد والرئة ونادرا ما تصيب أعضاء أخرى في الجسم. التصوير الطبي هو الأساس لتشخيص الكيسة العدارية. في هذا التقرير نسجل موقعا نادرا جدا للكيسة العدارية في البطين الأيمن للقلب في مريضة عمرها ٢٣ سنة، قدمت تعاني من ضيق في التنفس مع سعال وبلغم. كشفت الفحوصات المخبرية عن كثرة اليوزينيات مع فقر الدم. تصوير الصدر بالأشعة وتصوير البطن بالموجات فوق الصوتية كانت غير ملفتة. تصوير القلب بالأشعة المقطعية كشف عن وجود كيسين داخل البطين الأيمن. تخطيط صدى القلب عبر الصدر أكد وجود كيسين داخل البطين الأيمن ملتصقين بالحاجز البطيني. الكيسة العدارية كانت هي التشخيص الأول. تم إجراء عملية استئصال للكيسين وكشف تحليل العينات التي تم استئصالها بعد العملية وجود عدة أكياس تحتوي على الرؤيسات الحية للمشوكة الحبيبية. تعافى المريض وخرج من المستشفى واستمر بالعلاج بـ "البندازول" عن طريق الفم.

الكلمات المفتاحية: الكيسة العدارية؛ البطين الأيمن؛ استئصال كيسة قلبية؛ المشوكة الحبيبية

Abstract

Hydatid disease is a parasitic infection by the larval stage of the tapeworm *Echinococcus granulosus*. It affects liver, lungs and rarely other organs. Medical imaging provide the basis for diagnosis. This case report describes an

extremely rare location of cardiac hydatid cyst in the right ventricle of the heart. We describe a 23-year-old woman who presented with shortness of breath and productive cough. Laboratory investigations showed marked eosinophilia and anemia. Chest radiography and abdominal ultrasonography were unremarkable. Cardiac computed tomography (CT) identified two well-defined fluid densities in the right ventricle without contrast enhancement. A transthoracic echocardiography (TTE) showed two cystic lesions in the right ventricular cavity that was attached to the interventricular septum. Hydatid cyst was the most likely diagnosis followed by the possibility of a congenital cardiac cyst. An open-heart surgery with cardiac cystectomy was performed. Post-operative analysis of the resected specimens showed multiple hydatid cysts with living scolices of *Echinococcus granulosus*. The patient recovered uneventfully and was discharged on oral albendazole.

Keywords: Cardiac cystectomy; *Echinococcus granulosus*; Hydatid cyst; Right ventricle

© 2020 The Authors.

Production and hosting by Elsevier Ltd on behalf of Taibah University. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

* Corresponding address: Department of Diagnostic Radiology Technology, College of Applied Medical Sciences, Taibah University, Almadinah Almunawwarah, 42312, KSA.

E-mail: alshoabisultan@yahoo.com (S.A. Alshoabi)

Peer review under responsibility of Taibah University.



Introduction

Hydatid disease is a parasitic infection caused by the larval stage of the tapeworm *Echinococcus granulosus*. Humans are infected following ingestion of food or water contaminated with tapeworm eggs or larvae or after contact

with contaminated soil. After ingestion, the larvae penetrate the intestinal wall and travel via the blood or lymph to the liver, lungs, or other organs where they mature into a hydatid cyst.¹ According to World Health Organization estimates, more than 1 million individuals are affected by echinococcosis each year. This infection commonly occurs in the liver and lungs and rarely affects other organs.² Most patients are diagnosed based on medical imaging findings, and ultrasonography is the gold standard for screening and postoperative monitoring of these patients.^{1–3}

To date, few studies have reported cardiac hydatidosis.^{4–9} We report an extremely rare case of cardiac hydatid cysts detected in the right ventricle (RV). In this report, we describe the clinical and medical imaging findings in a patient with cardiac hydatid cysts and the outcomes of surgical treatment. Our study emphasises the role of medical imaging in the diagnosis of patients with an ambiguous clinical presentation.

Case report

A 23-year-old woman presented to the emergency department with shortness of breath (SOB) and productive cough. Her illness began with fever, sweating, and weight loss observed 3 months prior to presentation, and she was treated with antibiotics and antitussive drugs for a diagnosis of chest infection. She did not improve despite receiving treatment for 2 weeks and developed haemoptysis. Chest radiograph was unremarkable; however, the Mantoux test showed a strongly positive result. The patient was referred to the Tuberculosis (TB) National Center in Sana'a where three

sputum samples were collected for acid-fast bacilli testing; however, all specimens yielded negative results. Considering her family history of TB, an antituberculous drug regimen was initiated in this patient. Following 2 months of antituberculous therapy, the patient developed SOB with massive haemoptysis, necessitating resuscitation with 1 unit of blood transfusion. Physical examination showed that the patient was conscious, oriented, and cooperative with the examination. We observed mild pallor with normal vital signs (blood pressure = 90/60 mmHg, heart rate = 112 beats/min, and respiratory rate = 28/min). Chest auscultation revealed scattered crepitations in the right lung field. Auscultation of the heart revealed tachycardia with normal S1 and S2. Laboratory investigations revealed significant eosinophilia but a normal leucocyte count, serum haemoglobin level = 10 mg/dL, total leucocyte count = 4200 cells/mm³ (normal range 4000–11,000 cells/mm³), neutrophils = 50% (normal range 60–70%), lymphocytes = 28% (normal range 32–40%), eosinophils = 12.6% (normal range 4–5%), and erythrocyte sedimentation rate = 60 mm/h (normal range 0–20 mm/h). Blood chemistry tests showed no abnormalities, and chest radiography and abdominal ultrasonography findings were unremarkable.

Chest computed tomography (CT) revealed two well-defined defects (measuring 36 × 25 mm and 20 × 20 mm, density 10–15 Hounsfield units) located in the RV (Figure 1a). These lesions showed no enhancement after contrast agent administration, suggesting their cystic nature (Figure 1b). Nonspecific electrocardiogram revealed mild ischaemic changes (Figure 1c). Transthoracic echocardiography (TTE) revealed two cystic lesions

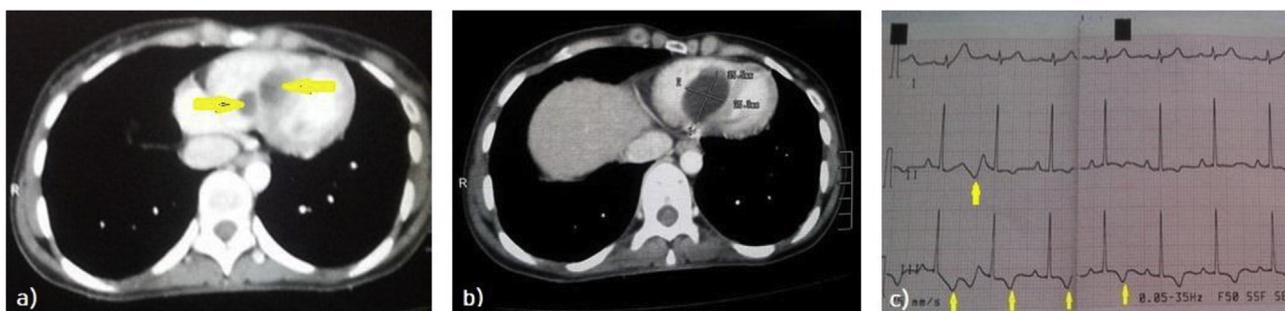


Figure 1: Contrast-enhanced computed tomography images showing a) two well-defined filling defects (arrows) in the right ventricle with no enhancement after contrast administration and, b) dimensions of the larger filling defect (cyst). c) Electrocardiogram (ECG) showing T-wave inversion in the inferior leads (arrows) indicating ischaemic changes.



Figure 2: Transthoracic echocardiography images showing a) two cystic lesions in the right ventricle, b) dimensions of the two cysts and, c) mild preoperative tricuspid regurgitation (right ventricular pressure 23 mmHg).

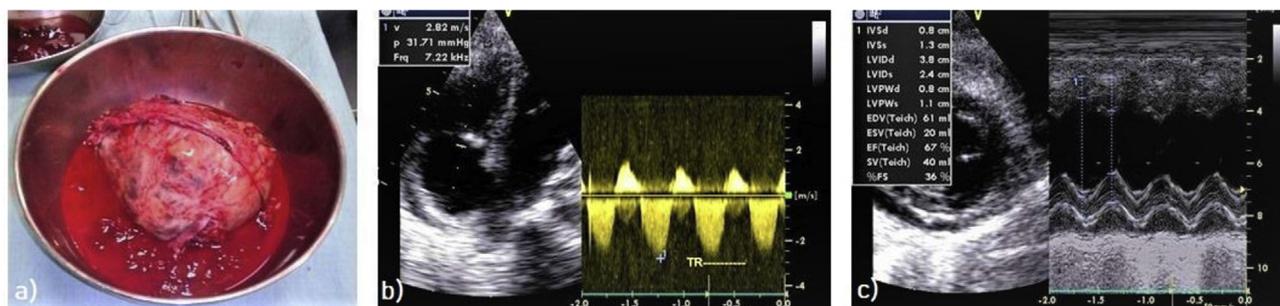


Figure 3: Postoperative images showing a) macroscopic view of the excised hydatid cyst, b) transthoracic echocardiography image showing mild postoperative tricuspid regurgitation (right ventricular pressure 31 mmHg) and, c) M-mode transthoracic echocardiography image showing a normal size and good contractility of the left ventricle.

(measuring 30×27 mm and 20×18 mm) attached to the interventricular septum within the RV cavity (Figure 2a & b). The cysts were directed toward the pulmonary artery but did not involve either the pulmonary or the tricuspid valve. Colour flow mapping revealed mild tricuspid regurgitation (Grade 1) (Figure 2c). Two-dimensional Doppler echocardiography and colour flow mapping revealed that all other cardiac valves were normal. All cardiac chambers appeared normal without any mural thrombi or vegetations. Based on the aforementioned findings, cardiac hydatidosis was considered the most likely diagnosis, followed by congenital cysts included in the differential diagnosis. The patient was admitted for open-heart surgery for hydatid cystectomy, and we performed surgical excision under cardiopulmonary bypass. Post-operative histopathological examination of the resected specimens (Figure 3a) revealed multiple hydatid cysts containing viable protoscolices of *E. granulosus*. Postcystectomy TTE revealed persistent mild tricuspid regurgitation (Figure 3b) with a normal left atrium (Figure 3c). The patient remained well and continued medical treatment with oral albendazole.

Discussion

Cardiac hydatid cysts are extremely rare owing to the continuous rhythmic contractile activity of the heart, which prevents lodgement of eggs or cysts within cardiac tissue. Our patient presented with hydatid cysts in the RV (an extremely rare site of involvement of the heart) with ambiguous clinical findings and an insidious onset of disease, resulting in misdiagnosis as TB and incorrect treatment for 2 months. Firouzi et al.⁷ reported that the free wall of the left ventricle (LV), the interventricular septum, and the atria are sites at which cardiac hydatid cysts are known to occur. We report a rare case of hydatid cysts detected in the RV; our findings are similar to those observed by L'aarje et al.,¹⁰ who reported a similar case of hydatid cysts detected in the RV. Cardiac hydatid cysts are rare and occur less commonly in the RV than in the LV,^{5,10} which is attributed to the larger muscle mass and greater vascular supply of the LV.¹⁰ Our patient presented as a diagnostic dilemma and was misdiagnosed and incorrectly treated initially. CT followed by echocardiography later established the correct diagnosis. Despite advances in

diagnostic imaging modalities, cardiac hydatid cysts are often indistinguishable from other cardiac masses, and accurate diagnosis remains challenging. Many previous studies have reported that echocardiography is the most effective modality to diagnose cardiac hydatid cysts. Cardiac CT, magnetic resonance imaging, and serological tests are other diagnostic modalities used in clinical practice.^{6–12} Our patient was effectively treated with surgical excision of the cyst and oral albendazole therapy. Several studies have reported that surgical excision combined with albendazole therapy remains the gold standard for treatment of cardiac hydatid cysts.^{4–11} Puncture, aspiration, injection of a scolicedal agent, and re-aspiration (PAIR) is a new minimally invasive method to treat hydatid cysts and is associated with a lesser risk of serious complications.¹³ Chemotherapy and PAIR followed by surgical excision of cysts is a new therapeutic strategy for hydatid cysts of the liver and the posterior mediastinum.¹⁴ However, the effectiveness of PAIR in the management of cardiac hydatid cysts remains unclear. The role of other novel therapeutic techniques such as radiofrequency ablation in the treatment of hydatid cysts in humans is questionable.¹⁵

In conclusion, cardiac hydatid cysts involving the RV are extremely rare and present a diagnostic dilemma. Medical imaging is the only effective method to diagnose cardiac hydatid cysts. Open-heart surgery remains the gold standard for the treatment of cardiac hydatid cysts and is associated with good outcomes.

Source of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors have no conflict of interest to declare.

Ethical approval

Consent was obtained from the patient to use radiographs and information. The patient understands that his name and initials will not be published, and due efforts will

be made to conceal his identity, but anonymity cannot be guaranteed.

Authors contributions

NNA working echocardiography of the patient and collected data, SAA designed and wrote the manuscript, AAQ reviewed the manuscript, and TSD provided logistic support. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

References

1. Fennira S, Kamoun S, Besbes B, Ben Mrad I, Zairi I, Ben Moussa F. Cardiac hydatid cyst in the interventricular septum: a literature review. *Int J Infect Dis* 2019 Nov; 88: 120–126. <https://doi.org/10.1016/j.ijid.2019.09.004>.
2. Mahmoudi S, Mamishi S, Banar M, Pourakbari B, Keshavarz H. Epidemiology of echinococcosis in Iran: a systematic review and meta-analysis. *BMC Infect Dis* 2019; 19(1): 929. <https://doi.org/10.1186/s12879-019-4458-5>. Published 2019 Nov 4.
3. Bhutani N, Kajal P. Hepatic echinococcosis: a review. *Ann Med Surg (Lond)* 2018; 36: 99–105. <https://doi.org/10.1016/j.amsu.2018.10.032>. Published 2018 Nov 2.
4. Bakkali A, Jaabari I, Bouhdadi H, Razine R, Bennani Mechita N, El Harrag J. [Cardiac hydatid cyst about 17 operated cases]. *Ann Cardiol Angeiol (Paris)* 2018; 67(2): 67–73. <https://doi.org/10.1016/j.ancard.2017.04.010>.
5. Noaman H, Rawaf S, Majeed A, Salmasi AM. Hydatid cyst of the heart. *Angiology* 2017 Oct; 68(9): 765–768. <https://doi.org/10.1177/0003319717690093>.
6. Separovic Hanzevacki J, Gasparovic H, Reskovic Luksic V, Ostojic Z, Biocina B. Staged management of a giant cardiac hydatid cyst: a case report. *BMC Infect Dis* 2018; 18(1): 694. <https://doi.org/10.1186/s12879-018-3599-2>. Published 2018 Dec 27.
7. Firouzi A, Neshati Pir Borj M, Alizadeh Ghavidel A. Cardiac hydatid cyst: a rare presentation of echinococcal infection. *J Cardiovasc Thorac Res* 2019; 11(1): 75–77. <https://doi.org/10.15171/jcvtr.2019.13>.
8. Su L, Yu J, Dai C, Liu Y, Peng L. Echinococcosis in left ventricle: a case report. *Medicine (Baltim)* 2019; 98(16):e15267. <https://doi.org/10.1097/MD.00000000000015267>.
9. Balani A, Kumar AD, Dey AK. Intramyocardial hydatid cyst: a case report. *Eur Heart J Case Rep* 2018; 2(2):yty053. <https://doi.org/10.1093/ehjcr/tyty053>. Published 2018 Jun 4.
10. L'aarje A, Lyazidi S, Kitane Y, Alami A, Habbal R. Cardiac hydatid cyst of the right ventricle: severe localization. *J Cardiol Cases* 2017; 16(4): 138–140. <https://doi.org/10.1016/j.jccase.2017.06.009>. Published 2017 Aug 2.
11. Seth HS, Mishra P, Khandekar JV, Raut C, Mohapatra CKR, Ammannaya GKK. A concomitant intramyocardial and pulmonary hydatid cyst: a rare case report. *Braz J Cardiovasc Surg* 2017; 32(2): 138–140. <https://doi.org/10.21470/1678-9741-2016-0046>.
12. Orah AY, Faqe DA, Kadoura M, Kakamad FH, Yaldo FF, Aziz SQ. Cardiac Hydatid cysts; presentation and management. A case series. *Ann Med Surg (Lond)* 2018; 30: 18–21. <https://doi.org/10.1016/j.amsu.2018.04.001>. Published 2018 Apr 7.
13. Richter J, Profis E, Holtfreter MC, Orhun A, Müller-Stöver I, Dedelen H, et al. Anaphylactic shock ensuing therapeutic puncture of an echinococcal cyst. *Parasitol Res* 2015; 114(2): 763–766. <https://doi.org/10.1007/s00436-014-4275-0>.
14. Quail JF, Gramins DL, Dutton WD. Hydatidosis of the liver and posterior mediastinum. *Int J Surg Case Rep* 2015; 7C: 26–28. <https://doi.org/10.1016/j.ijscr.2014.12.031>.
15. Tamarozzi F, Vuitton L, Brunetti E, Vuitton DA, Koch S. Non-surgical and non-chemical attempts to treat echinococcosis: do they work? *Parasite* 2014; 21: 75. <https://doi.org/10.1051/parasite/2014071>.

How to cite this article: Aljaber NN, Alshoabi SA, Qurashi AA, Daqqaq TS. Cardiac hydatid cyst in the right ventricle: An unusual case at a rare site. *J Taibah Univ Med Sc* 2020;15(3):249–252.