Case Report

Nepalese Heart Journal 2022; Vol 19(1), 75-78

A cardiac hydatid cyst; four years of postoperative follow up

Navin Chandra Gautam[1], Rheecha Joshi[1], Marisha Aryal[1], Rupak Pradhan[1], Rabindra Bhakta Timala[1]

[1] Department of Cardiovascular Surgery, Shahid Gangalal National Heart Center, Bansbari, Kathmandu.

Corresponding Author: Navin Chandra Gautam
Department of Cardiovascular Surgery, Shahid Gangalal National Heart Center, Kathmandu, Nepal.
Email: navincg@hotmail.com
ORCID ID NO: 0000-0003-2416-4096.

Cite this article as: Gautam N.C., Joshi R., Aryal M., et al. A cardiac hydatid cyst; four years of follow up. Nepal. Nepalese Heart Journal 2022; Vol 19(1) : 75-78

Submission date: 15th December 2021
Accepted date: 22nd February 2022

Abstract

Hydatid disease is a parasitic disease that mainly involves liver and lung tissues. Isolated cardiac involvement is very rare. We report a 40-year-old woman who presented in our emergency department for non-specific chest pain. She had no prior history of a hydatid disease Her transthoracic echocardiography illustrated a cystic mass in the right ventricular apex. Her contrast-enhanced computed tomographic scan of the chest showed a complex cystic lesion in the right ventricular wall at region of apex with enhancing thin wall and internal septations. Her immunological IgG test was positive for Echinococcosis. No other hydatid cysts were seen in the other organs such as liver and lungs by ultrasound scan of abdomen and computer tomography scan of chest respectively. After a week course of Albendazole, 400mg twice a day, she underwent cystectomy with cappitonage surgery under cardiopulmonary bypass. The patient was discharged from the hospital after an uneventful postoperative recovery. Echocardiographic and cardiac contrast enhanced computer tomography (CECT) evaluation in fourth year of follow-up revealed no evidence of recurrence of hydatids and ventricular function remained normal.

Keywords: Hydatid cyst, Cardiac involvement, Right ventricle

DOI: https://doi.org/10.3126/njh.v19i1.45311

Introduction

Hydatid disease is a zoonosis parasitic infection caused by Echinococcus granulosus, E. multilocularis, or E. vogeli. The liver, followed by the lung, are the most common site of involvement. Cardiac involvement of the hydatid cyst is uncommon (less than 2%), and cardiac involvement without hepatic involvement is rare. However, the most common sites for cardiac involvement are the ventricular septum and the left ventricular free wall. The embryo usually reaches the myocardium via the coronary circulation from the left side of the heart. The cyst is then formed within a period of one to five years. The myocardial reaction consists of a fibrous adventitial pericyst layer surrounding the laminated membrane.[1] We report a 40 years old woman with the hydatid cyst of the right ventricular free wall without hepatic involvement. She underwent cystectomy with cappitonage and uneventful postoperative coarse. Her fourth years follow up with echocardiography and cardiac CECT shows free of recurrence. She remains clinically fine till date

Case Report

A 40-year-old female presented in our emergency department with the complaint of non-radiating chest pain for 1 week with no other associated symptoms. She had no significant past medical history or comorbidities. Transthoracic echocardiographic examinations illustrated normal cardiac structures and function. However, there was large pericardial effusion with fibrin deposits with no features of cardiac tamponade and a cystic mass in right ventricular apex measuring 2.8 cm x 3.7 cm. Thoracic and abdominal cavity multislice CT scan with intravenous contrast showed a complex cystic lesion in right ventricular wall at the region of apex with enhancing thin wall and internal septa, gross pericardial effusion. There were no other such cystic lesions in the lung or liver.

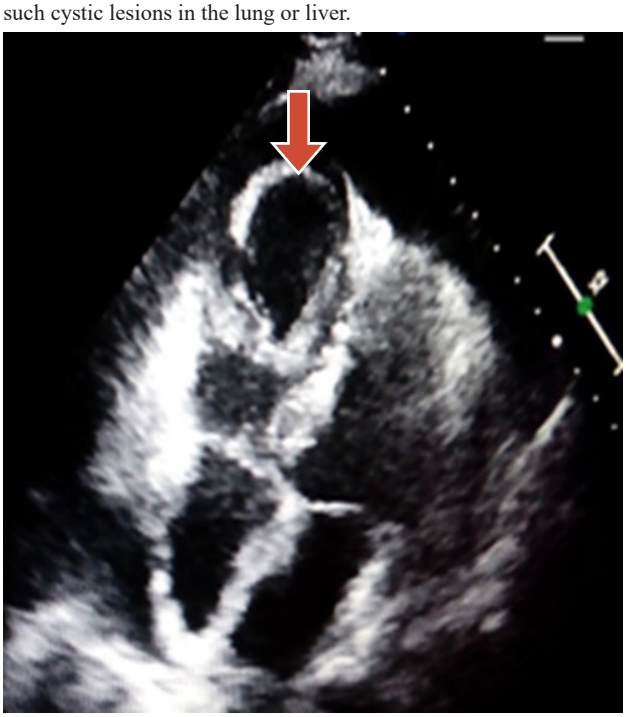


Figure 1: *Transthoracic Echocardiography showing a mass at right ventricular apex*

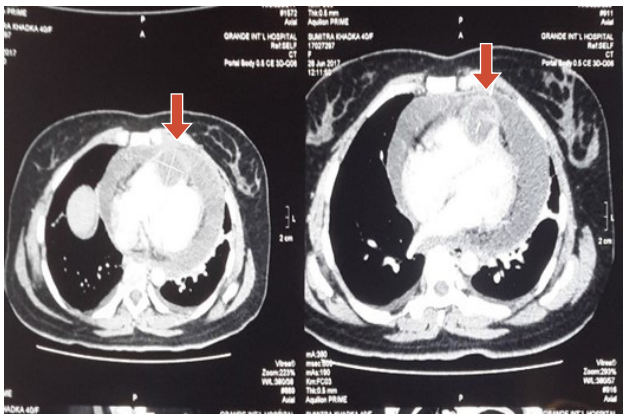
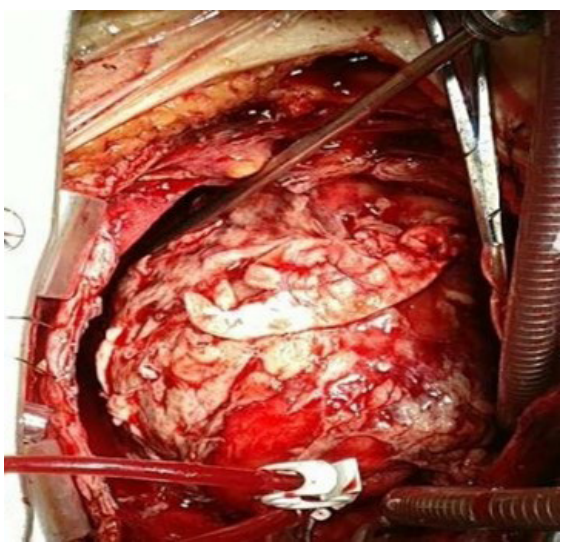
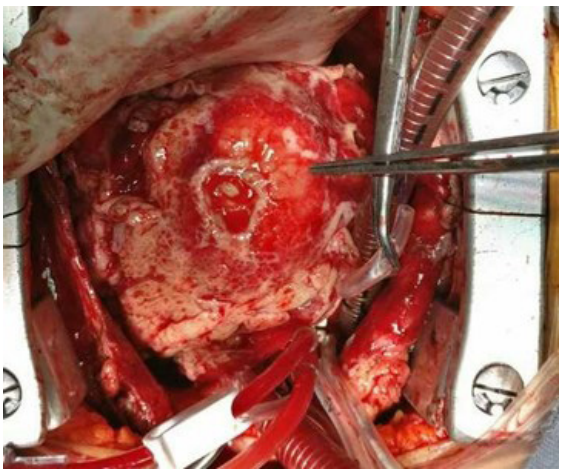


Figure 2: *Contrast enhanced computer tomography (CECT) scan showing the mass with multiple septa and pericardial fluid collection*

Echinococcus IgG test was positive. She was planned for urgent surgery. Preoperatively, tablet Albendazole 400 mg twice daily was given for two weeks. Surgical cystotomy of cardiac hydatid cyst via a median sternotomy on cardiopulmonary bypass (CPB) was planned. Standard cardiopulmonary bypass through cannulation of the right atrium and the ascending aorta was established. Myocardial protection was achieved by administration of antegrade cardioplegia and mild hypothermia. Thickened and inflamed pericardium with fibrinous deposits all over the pericardial cavity was found. There was an intra myocardial hydatid cyst (4 cm x3cm) with septation and daughter scolices at right ventricular apex. The cyst was surrounded by povidone-soaked gauze packs to prevent dissemination of cyst. An oblique incision on cystic mass was given and the cyst was excised from myocardium together with its capsule. Myocardial cavity was washed thoroughly with povidone iodine solution and normal saline. The cavity was filled with hemostatic powder. The shape and geometry of right ventricle was protected. The Teflon patch (felt) was used to suture myocardial cavity using pledged-U-sutures.



[A]



[B]

Figure 3: *A: Intraoperative appearance of right ventricular cyst, B: Reconstruction of right ventricular wall after cystotomy.*

Successful weaning from extracorporeal circulation was completed with minimal catecholamine support. The postoperative period was uneventful. We prescribed her albendazole 400mg twice daily for 12 weeks. Yearly follow up of the patient was done. On the fourth year follow up, she had no postoperative complications and no evidence of relapse of the disease as evaluated by transthoracic echocardiography (figure 4) and CECT chest (figure 5).

The patient's consent has been obtained, and reporting of the case for publication was approved by the institutional review committee of our institution.

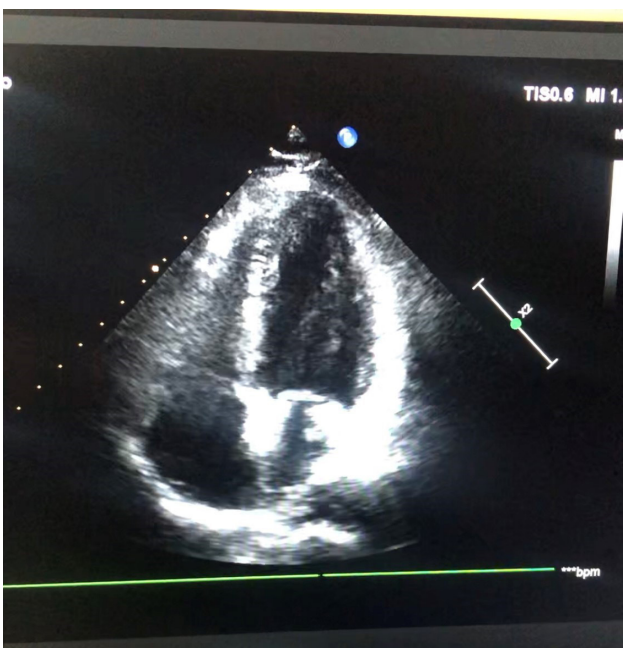


Figure 4 *Echocardiography on fourth year of follow up*

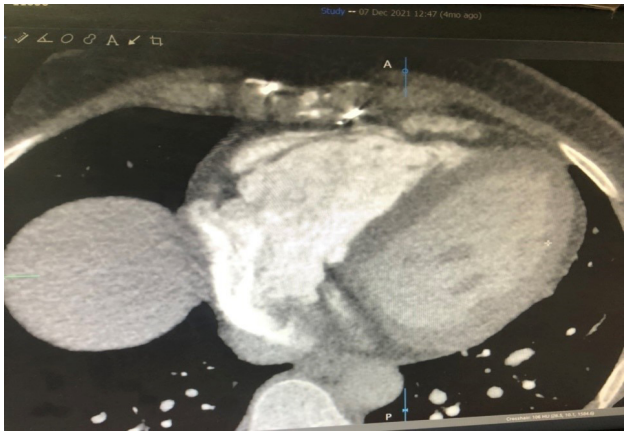


Figure 5: *Cardiac contrast enhanced computer tomography (CECT) scan of the patient on the fourth year of follow up*

Discussion

Hydatid cyst is a parasitic disease caused by a tapeworm of E. granulosus, E. multilocularis, or E. vogeli.[2] The most common site of its involvement is the liver followed by lungs. The incidence of concomitant liver and lung hydatidosis ranges from 5.8 to 13.3%.[1] The primary echinococcosis of the heart is rare and it can occur at around 0.5% and 2% of all hydatid cyst involvement in comparison with the liver (70%) or lung (20%). In primary cardiac hydatidosis, the tapeworm larvae reach the myocardium passing through the coronary circulation, via the pulmonary circulation or a patent foramen ovale. As the left ventricle has maximum myocardial mass and abundant blood supply, its invasion by echinococcus is 55-60%. Likewise, incidence of interventricular septum is 5–9%, right ventricle is 15%, right atrium is 3–4% and pulmonary artery, left atrium, and pericardium is up to 7–8%.[3] The primary pericardial cysts are among the rare phenomenon and are seen to develop after a rupture of myocardial cyst or contamination during surgical intervention.[4] The differential diagnosis of the cardiac hydatid cyst includes intracardiac tumors, congenital cysts and aneurysms.[5]

The clinical presentation of cardiac echinococcosis depends on the size and location of the cyst.[1] The growth of hydatid cyst is usually slow and asymptomatic and just about 10% of patients with cardiac hydatid cyst are symptomatic.[6] The symptoms and signs of cardiac echinococcosis (if present) are extremely variable. Non-specific features such as weight loss, cough, fever, and dyspnea are likely to be the presenting symptoms especially in children.

Hydatid disease is endemic in cattle-raising areas of the world, notably in the Mediterranean countries, the Middle East, South America, Australia, and New Zealand.[7,8] The incidence of hydatidosis in the Turkish population has been reported as 1:20,000.8

To our knowledge, this is the first case report of isolated cardiac hydatid cyst in the country. Out of total 20000 cardiac operation performed at this center; this is the first operation for cardiac hydatid cyst. The incidence of hydatidosis in Nepal is not known.

Mild, recurrent non-specific chest pain is the most common complaint, which may be due to an episode of partial rupture into the pericardium, with resultant pericarditis. The patient may present with typical angina because of the compression of the hydatid cyst on the adjacent myocardium and may result in wrong diagnosis, especially in elderly patients. If a cardiac cyst is located in the right atrium, it could even be mistaken with the myxoma of the atrium. Moreover, it can present as a conduction disturbance when it is located near the conduction system, particularly the interventricular septum. A case was reported on 57-year-old man who presented with syncope due to ventricular tachycardia and was managed with antiarrhythmic drugs for an acute episode.[9]

The most severe complication of this disease is the rupture. Intrapericardial rupture of hydatid cyst develops in about 10% of the cases resulting in acute pericarditis and eventually constrictive pericarditis. During intracardiac rupture embolization in the pulmonary or systemic circulation may occur, leading to dissemination of the disease in different organs. The patient may develop anaphylactic reaction with severe circulatory collapse and even death may ensue. The frequency of intramyocardial perforation is high i.e., 25-40%. The right atrium hydatid cyst can give rise to pulmonary hypertension secondary to cystic embolization.

There are different methods in order to investigate cardiopericardial hydatidosis and echocardiography is the procedure of choice. Echocardiography is highly sensitive and specific in the diagnosis of hydatid cysts [6] and positive serological tests can help the diagnosis of this disease. Thameur et al. have suggested that CT scan is better than echocardiography considering its ability to distinguish solid from liquid tumors and also CT scan is an effective and reliable tool for the surgeons which can provide the exact site of the abnormality.[10] Two-dimensional echocardiography and magnetic resonance imaging are currently the best diagnostic modalities to demonstrate a cardiac hydatid cyst.[11] Pericardiocentesis and invasive diagnostic procedures should be avoided due to the risk of rupture of cyst.[12]

Serological tests can be false-negative in 10–20% of patients with hepatic hydatid cysts, in 40% with pulmonary cysts and in 50% with cardiac cysts; this is most likely linked with an insufficient immune response.[13] The enzyme-linked immunosorbent assay (ELISA) is one of the most specific serological tests that can be used, and a positive result for echinococcus antibodies confirms the diagnosis[14]

For the cardiac hydatid cyst, the recommended treatment is enucleation of the cyst under cardiopulmonary bypass with topical scolicidal agents in the surrounding operative field.[15, 16]

In addition, adjuvant treatment with oral Albendazole may reduce the size of the cyst and prevent recurrence. The recommended treatment is enucleation, pericystectomy or cystectomy with cappitonage of the cyst under cardiopulmonary bypass via median sternotomy with topical scolicidal agents in the surrounding operative field by instillation of scolicidal agents such as 2% formalin, 0.5% silver nitrate solution, 20% hypertonic saline solution, 1% iodine solution or 5% cetrimide solution.[17]

During surgery, it is important to minimize spillage of the cystic contents in order to prevent intraoperative dissemination and eventual recurrence. This may be accomplished by delivery of intact cyst or by cystic fluid aspiration with use of scolicidal agent and preoperative therapy with Albendazole. As a rule, the heart should not be manipulated before application of cross clamp and cardiopulmonary bypass.[18] Surgical excision is also indicated in asymptomatic patients [17] because if left untreated it may transform to large cavity with a high risk of rupture.[19]

In our patient, she is in fourth years of follow up and there are no cardiac symptoms. Her echocardiography and CECT is free from recurrence. In a study from China, 15 patients with cardiac hydatid cyst were followed up from ten months to 22 years, there were four recurrences of myocardial cysts after 12 months.[20] About 10% of all cardiac hydatid cysts tend to recur after surgery, but this rate may decrease with proper medical treatment and proper prevention of spillage of protoscoleces during surgery.[21]

Conclusion

Cardiac hydatid cysts are rare and may present with a variety of signs and symptoms, especially in the endemic zones. Due to the high risk of associated complications, cardiac hydatid cysts should be removed surgically as early as possible, even in asymptomatic patients. During surgery, measures should be taken to prevent perioperative embolization of a germinative membrane. Surgical

excision under cardiopulmonary bypass is the treatment of choice. Regular follow up with echocardiograhy and CECT is recommended to detect recurrence.

References

1. Sai Chandran BV, Durga Prasad R, Sudarsan Reddy P, Subbarao VK. Intramyocardial hydatid cyst: a mistaken identity and its successful removal on a beating heart. Interact Cardiovasc Thorac Surg. 2011;13(2):208-10. https://doi.org/10.1510/icvts.2011.267518
2. Shojaei E, Yassin Z, Rezahosseini O. Cardiac Hydatid Cyst: A Case Report. Iran J Public Health. 2016 Nov;45(11):1507-1510. PMID: 28028503; PMCID: PMC5182260.
3. Yaliniz H, Tokcan A, Salih OK, Ulus T. Surgical treatment of cardiac hydatid disease: A report of 7 cases. Tex Heart Inst J. 2006;33(3):333-9. PMID: 17041691; PMCID: PMC1592285
4. Vazhev ZG, Stoev HA. Cardiac Echinococcosis Involving Left Ventricular Myocardium in an 18-Year-Old Patient. Folia Med (Plovdiv). 2018 Jun 1;60(2):308-313. https://doi.org/10.1515/folmed-2017-0081 PMID: 30355821.
5. Khosravi A, Taghipour H, Fanaei SA, Assar O, Ghyasy MS, Mirlohi SM. Heart hydatid cyst close to the left descending artery in a thirteen-year-old boy. Iran Red Crescent Med J. 2014 Sep 5;16(9): e15164. https://doi.org/10.5812/ircmj.15164 PMID: 25593721; PMCID: PMC4270658.
6. Canpolat U, Yorgun H, Sunman H, Aytemir K. Cardiac hydatid cyst mimicking left ventricular aneurysm and diagnosed by magnetic resonance imaging. Turk Kardiyol Dern Ars. 2011 Jan;39(1):47-51. PMID: 21358231.
7. Ameli M, Mobarhan HA, Nouraii SS. Surgical treatment of hydatid cysts of the heart: report of six cases. J Thorac Cardiovasc Surg. 1989 Nov;98(5 Pt 2):892-901. PMID: 2682022
8. Salih OK, Celik SK, Topcuoğlu MS, Kisacikoğlu B, Tokcan A. Surgical treatment of hydatid cysts of the heart: a report of 3 cases and a review of the literature. Can J Surg. 1998 Aug;41(4):321-7. PMID: 9711167; PMCID: PMC395009
9. Singh SK, Singh V, Kumar S, Devenraj V, Bhandari M, Pandey AK. Right ventricular hydatid cyst presented as tachyarrhythmia. Asian Cardiovasc Thorac Ann. 2019 Jul;27(6):489-491. https://doi.org/10.1177/0218492318817665
10. Alizadeh-Ghavidel A, Kyavar M, Sadeghpour A, Totonchi Z, Mirmesdagh Y, Almassi N, Madadi S. Unusual clinical presentation of a giant left ventricle hydatid cyst. J Cardiovasc Thorac Res. 2013;5(4):175-8. https://doi.org/10.5681/jcvtr.2013.038 PMID: 24404351; PMCID: PMC3883543
11. De Castro S, Santilli M, Brandimarte C, Gualdi G, Migliau G.Role of noninvasive diagnosis in cardiac echinococcosis: pulmonary embolization from right atrial localization. Cardiologia. 1991;36(10):817-21.https://doi.org/10.1016/S0022-5223(94)70468-6
12. De Martini M, Nador F, Binda A, Arpesani A, Odero A, Lotto A. Myocardial hydatid cyst ruptured into the pericardium: cross-sectional echocardiographic study and surgical treatment. Eur Heart J. 1988;9(7):819-24. https://doi.org/10.1093/eurheartj/9.7.819
13. Soleimani A, Sahebjam M, Marzban M, Shirani S, Abbasi A. Hydatid cyst of the right ventricle in early pregnancy. Echocardiography. 2008 Aug;25(7):778-80. https://doi.org/10.1111/j.1540-8175.2008.00668.x. PMID: 18754937.
14. Aleksic-Shihabi A, Vidolin EP. Cystic echinococcosis of the heart and brain: a case report. Acta Med Okayama. 2008 Oct;62(5):341-4. https://doi.org/10.18926/AMO/30968. PMID: 18985095.
15. Tuncer E, Tas SG, Mataraci I, Tuncer A, Donmez AA, Aksut M, Yakut C. Surgical treatment of cardiac hydatid disease in 13 patients, Tex Heart Inst J, 2010; 37(2):189-193. PMCID: PMC2851433PMID: 20401292
16. Kabbani SS, Ramadan A, Kabbani L, Sandouk A, Nabhani F, Jamil H. Surgical experience with cardiac echinococcosis, Asian Cardiovasc Thorac Ann, 2007 Oct, 15:422-426 https://doi.org/10.1177/021849230701500513
17. 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 Mar-Apr;32(2):138-140. https://doi.org/10.21470/1678-9741-2016-0046 . PMID: 28492796; PMCID: PMC5409250.
18. Kaplan M, Demirtas M, Cimen S, Ozler A. Cardiac hydatid cysts with intracavitary expansion. Ann Thorac Surg. 2001 May;71(5):1587-90. https://doi.org/10.1016/s0003-4975(01)02443-2. PMID: 11383804.
19. Soydinc S, Davutoglu V, Oylumlu M. Progress of untreated massive cardiac echinococcosis--echocardiographic follow-up. Echocardiography. 2006;23(9):780-3.
20. Murat V, Qian Z, Guo S, Qiao J. Cardiac and pericardial echinococcosis: report of 15 cases. Asian Cardiovasc Thorac Ann. 2007 Aug;15(4):278-9. https://doi.org/10.1177/0218492307015004 02. PMID: 17664197.
21. Rr Fei Yan, Qiang Huo, Murat Abudureheman, Jun Qiao, SongFeng Ma, Hao Wen, Surgical treatment and outcome of cardiac cystic echinococcosis, European Journal of Cardio-Thoracic Surgery, Volume 47, Issue 6, June 2015, Pages 1053–1058, https://doi.org/10.1093/ejcts/ezu323