

Pediatric Hydatid Cyst with Ventricular Aneurysm and Surgical Treatment With Dor Procedure, Case Repot

Ayşegül Durmaz¹, Oguz Omay¹, Cagri Duzyol¹, Sibel Gür¹, and Ekin İLKELİ²

¹Kocaeli Universitesi Tıp Fakultesi

²Düzce Atatürk Devlet Hastanesi Department of Cardiovascular SurgeryDüzce Tukey

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Abstract

Cardiac involvement of hydatid cyst disease is a rare presentation but may lead to life-threatening complications such as cyst rupture and should be treated surgically. A 10-year-old male patient with cranial and complicated cardiac hydatid cyst disease lesions that caused lower extremity peripheral arterial occlusion and aneurysmatic dilatation in the left ventricular apex is presented. Although the patient was in the pediatric age group, the Dor procedure was successfully applied to preserve the ventricular geometry. The Dor procedure for a ventricular aneurysm caused by a cardiac hydatid cyst was rarely applied in the pediatric age group. In conclusion, this case differs from other cardiac hydatid cysts previously reported in the literature due to the advanced stage of the disease, atypical clinical presentation, and rare complications despite the young age of the case. The surgical method used in treating the patient makes the subject more interesting.

Pediatric Hydatid Cyst with Ventricular Aneurysm and Surgical Treatment With Dor Procedure, Case Repot

Ayşegül DURMAZ, MD¹

Oğuz OMAY, MD¹

Çağrı DÜZYOL, MD¹

Sibel GÜR, MD¹

Ekin İLKELİ²

Kocaeli University Medical Faculty, Department of Cardiovascular Surgery, Kocaeli, Turkey¹

Düzce Atatürk Devlet Hastanesi, Department of Cardiovascular Surgery, Düzce, Turkey²

Corresponding Author: Ayşegül Durmaz, Kocaeli University Medical Faculty, Department of Cardiovascular Surgery, drayseguldurmaz@gmail.com, ORCID: 0000-0001-5116-4361, Phone: 0090 531 792 3282

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Abstract:

Cardiac involvement of hydatid cyst disease is a rare presentation but may lead to life-threatening complications such as cyst rupture and should be treated surgically. A 10-year-old male patient with cranial and complicated cardiac hydatid cyst disease lesions that caused lower extremity peripheral arterial occlusion and aneurysmatic dilatation in the left ventricular apex is presented. Although the patient was in the pediatric age group, the Dor procedure was successfully applied to preserve the ventricular geometry. The Dor procedure for a ventricular aneurysm caused by a cardiac hydatid cyst was rarely applied in the pediatric age group. In conclusion, this case differs from other cardiac hydatid cysts previously reported in the literature due to the advanced stage of the disease, atypical clinical presentation, and rare complications despite the young age of the case. The surgical method used in treating the patient makes the subject more interesting.

Keywords: Cardiac, Hydatidcyst, Leftventricle, Rupture, Aneurysm, Systemicembolism

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Introduction:

Hydatid cyst disease (HCD), a zoonotic parasitic infection caused by *Echinococcus granulosus*, *E. multilocularis*, or *E. vogeli*, is a significant health problem and has a worldwide distribution with a higher incidence in underdeveloped and developing countries. Humans become intermediary carriers from consuming food contaminated with parasite eggs. The parasite embryo can reach the systemic circulation through the intestine and then reach any organ with a different prevalence. Multiple organ involvement can be seen in cases.[1] Cardiac hydatid cyst (CHC) is extremely rare and represents 0.5-2% of all human cases.[2] However, it can lead to severe and life-threatening complications. Occurrence in children is infrequent since the cyst grows slowly by about 1 cm per year.[3] The primary recommended treatment is surgery.

Here, we present a rare case of a hydatid cyst (HC) in childhood with cardiac involvement and intracardiac rupture that caused a true ventricular aneurysm treated with the Dor procedure for minimizing the akinetic tissues and restoring natural ventricular geometry.

Case Presentation:

A 10-year-old male patient was evaluated in another center with a complaint of headache that started nine months ago. The cranial magnetic resonance imaging (MRI) examination showed multiple cystic lesions. The results of the serological tests were negative. Surgical treatment was performed, and intraoperative evaluation was compatible with HCD. The patient was referred to our clinic for treatment planning upon detecting transthoracic echocardiography (TTE) findings consistent with a CHC lesion in the left ventricular apex.

In the first evaluation, there were no pathological signs in the patient's neurological, abdominal, pulmonary, and cardiac physical examinations. Since bilateral lower extremity pulses could not be palpated, aorta-lower extremity computed tomographic angiography imaging (CTA) was performed in addition to cranial, abdominal, and cardiac(MRI). In the cranial MRI, a contrasting multiple hydatid cyst lesion was observed in the bilateral cerebral hemispheres, which was smaller than in the previous MRI. CTA of the aorta and lower extremities showed occluded abdominal aorta and iliac arteries in a 5 cm segment. Femoral arteries were filling with collaterals. On cardiac MRI, the hydatid cyst lesion at the apex of the left ventricle (LV) was seen as opening into the ventricle and causing aneurysmatic dilatation in the myocardium.[Figure 1]

The patient's occlusion of the abdominal aorta was compatible with systemic embolism due to a ruptured CHC. No other organ involvement was observed in the radiological evaluation. The only data we have regarding the etiology is that the patient had a brief history of feeding a stray cat at home two years ago. The patient was surgically repaired with the Dor procedure (aneurysmectomy and circular endoventricular patch plasty). [Figure 2]The postoperative period was uneventful. The patient was followed up regularly for five years by the local cardiovascular surgeon in coordination with our clinic.

Discussion:

HCD is an endemic parasitic infection that can develop in almost any body part, especially the liver (75%) and lungs (15%).[4] Echinococci reach the right heart cavities by the portal or lymphatic routes, reach the left heart through the pulmonary circulation, patent foramen ovale, or an intracardiac defect that causes a right-left shunt and settles into the myocardium via the coronary circulation.[5] LV, the part of the heart with the most abundant blood supply, is most frequently involved (60%), followed by the right ventricle (10%), pericardium (7%), pulmonary artery (6%), left atrial appendage (6%) the interventricular septum (4%) have been reported.[1] In this case, no focus of HC was detected, except for multiple organ involvement in the cranial and cardiac areas; accordingly, echinococci probably reached the left heart without attaching to the pulmonary capillaries.

HCD is generally asymptomatic, and signs and symptoms typically appear decades later because the cyst grows slowly.[6] The clinical presentation of CHC varies depending on the location, age, size, infection, number, and calcification.[5] Chest pain, palpitations, and dyspnea are the main symptoms of HCD. However, it can cause fatal complications such as sudden rupture, suppuration, anaphylactic shock, arrhythmia, and embolization.[5] HC in the LV are usually located subepicardially and rarely rupture into the pericardial cavity.[7] Intracavitary rupture in LV may cause systemic embolism, such as occlusion of the mesenteric or lower extremity arteries.[8] However, HC in the right ventricle is usually located subendocardially and causes pulmonary embolization, mainly due to intracavitary rupture.[9] Operative and surviving perforated CHCs have been rarely reported in the literature. In our case, there was occlusion in the distal abdominal aorta and bilateral iliac arteries due to the rupture of the cardiac cysts into the LV. The patient had no symptoms, both the CHC and its complications.

In addition, CHC can mimic left ventricular aneurysms (LVA) or cardiac malignancies. It can also be thought to cause cardiac aneurysms acquired in childhood since it is a progressive parasitic infection that forms cystic cavities. However, no information was found in the literature regarding this.[10] In our rare case, the evaluations show that the cyst was not mimicking an aneurysm but caused a true cardiac aneurysm by the destruction of the left ventricle apical wall that was noticed after it ruptured.

The diagnosis of cardiac HCD is mainly based on clinical suspicion, cardiac imaging, and serological tests. Antibody assays are helpful primarily to confirm the possible radiological diagnosis. However, negative serological test results do not exclude the diagnosis.[11] TTE is a very sensitive and specific diagnostic tool that shows the effect of the lesion on ventricular or valve functions. CT is necessary in the diagnosis, but it can be misleading, especially in the case of myocardial cyst rupture. Cardiac MRI can provide valuable information about the lesion and its relationship to other cardiac and extracardiac structures.[12] In our patient, the negative serological tests and the absence of pathological findings in the abdomen and lung imaging caused a shift away from the diagnosis of HC in the unit that first evaluated the patient. However, cardiac involvement was detected by TTE, and MRI revealed that it caused intracavitary rupture and aneurysm in the apex of the LV.

CHC should be surgically removed even in asymptomatic patients because of the high fatal risk of complications.[13] Ventricular aneurysms are also rare in children, and theories of etiology differ.[14] Approaches to surgical treatment also differ among surgeons.[15] Typically, aneurysmectomy or primary closure with plication of the aneurysm sac is preferred.[14] Endoventricular circular patch plasty, also known as the Dor procedure, was described by Dor in adults to repair akinetic wall segments due to transmural ischemia. The procedure reshapes the left ventricle with a suture surrounding the transition zone between the contractile myocardium and aneurysmal tissue. It restores ventricular wall continuity with a patch, but the akinetic or dyskinetic portions of the anterior wall and septum are excluded from the procedure.[16-17] This surgical approach was rarely described in children, but it offers the same benefits as in adults, such as excision of noncontractile (fibrotic) ventricular wall to minimize akinetic or dyskinetic tissues and restore natural ventricular chamber geometry.[14-15] In this case, surgical treatment was deemed appropriate because of our patient's complicated CHC, the unknown presence of an intracardiac cystic structure, and the risks of additional complications related to the apical LVA.

Although some superficially located small cysts can be directly intervened, resection under cardiopulmonary bypass, since 1962, has been considered the safest method for its advantages, such as preventing systemic embolization by placing aortic cross-clamps and enabling the recognition of additional lesions missed before surgery.[18] Gentle and limited manipulation of the heart under cardiopulmonary bypass reduces the risk of operative complications.[19] The choice for surgical treatment is total excision of the cyst. However, if this is not possible, complete closure of the cyst by plication and obliteration of the cavity should be performed since complications are observed in cases of simple drainage or marsupialization of the cavity near the cardiac structures.[5] In our case, the LV was explored under cardiopulmonary bypass, and no pathological tissue sample of the HC was observed. Surgical repair was performed with aneurysmectomy and Dor procedure since complete excision would not be possible in the current state.

Serological and echocardiographic check-ups are recommended five years after surgical treatment to detect recurrences after manipulation or cysts not discovered during the operation.[20] No late cardiac problems or cyst recurrence was observed during the 5 years follow-up period.

Conclusion:

In conclusion, this case differs from other CHCs previously reported in the literature due to the advanced stage of the disease, atypical clinical presentation, and rare complications despite the young age of the case. Such cases should alert clinicians, especially if the patient comes from an area where the disease is endemic. Keep in mind the possibility of a CHC, even if the symptoms are not typical, and conducting rigorous investigations using detailed clinical evaluation and available diagnostic methods is highly important. If HCD is detected, systemic findings should also be investigated carefully. Regardless of the localization of KCC, its treatment is a surgical intervention without delay.

References:

1. Uysalel A., Aral A., Atalay S., Akalin H. Cardiac echinococcosis with multivisceral involvement. *Pediatr. Cardiol.* 1996;17:268–270.
2. Atilgan D, Kudat H, Tükek T, et al. Role of transesophageal echocardiography in diagnosis and management of cardiac hydatid cyst: report of three cases and review of the literature. *J Am Soc Echocardiogr.* 2002;15(3):271–4.
3. Maroto LC, Carrascal Y, López MJ, Forteza A, Pérez A, Zavanella C. Hydatid cyst of the interventricular septum in a 3.5-year-old child. *Ann Thorac Surg.* 1998;66(6):2110–1.
4. Beggs I. The radiology of hydatid disease. *AJR Am J Roentgenol.* 1985;145:639–48.
5. V. Kutay, H. Ekim and C. Yakut Infected myocardial hydatid cyst imitating left ventricular aneurysm *CARDIOVASCULAR SURGERY JUNE 2003 VOL 11 NO 3*
6. Akar R, Eryilmaz S, Yazicioglu L, et al.: Surgery for cardiac hydatid disease: an Anatolian experience. *Anadolu Kardiyol Derg.* 2003, 3:238-244.
7. Al-Naaman YD , Al-Omeri MM . Cardiopericardial echinococcosis causing myocardial insufficiency, right-sided heart failure and constrictive pericarditis. *J Cardiovasc Surg (Torino)* 1977; 11:303.
8. Ayteç A., Türkoğlu H., Paker T.Bilal S., Akçevin A., Bakay C., Sarioğlu T., Olga R., Öztürk M., Demiroğlu C. Kardiyak Hidatik Kist Hastalığı ve Cerrahi Tedavisi: 6 Şahsi Vak'aya Dayanan Derleme. *Türk Kardiyol Dern. Arş.* 19:340-344, 1991
9. Pasaoglu I, Dogan R, Hizan E, Oram A, Bozer AY. Right ventricular hydatid cyst causing recurrent pulmonary emboli. *Eur J Cardiothorac Surg* 1992;6:161–3.
10. Birincioglu CL, Bardakçi H, Küçüker SA, Ulus AT, Arda K, Yamak B, et al. A clinical dilemma: cardiac and pericardiac echinococcosis. *Ann Thorac Surg* 1999;68:1290-4.
11. Zhang W, McManus DP. Recent advances in the immunology and diagnosis of echinococcosis. *FEMS Immunol Med Microbiol* 2006;47:24-41
12. Uğur Canpolat, M.D., Hikmet Yorgun, M.D., Hamza Sunman, M.D., Kudret Aytemir, M.D. Cardiac hydatid cyst mimicking left ventricular aneurysm and diagnosed by magnetic resonance imaging *Türk Kardiyol Dern Arş - Arch Turk Soc Cardiol* 2011;39(1):47-51
13. Hafize Yaliniz, MD Acar Tokcan, MD Orhan K. Salih, MD Tumer Ulus, MD Surgical Treatment of

- Cardiac Hydatid Disease Texas Heart Institute Journal Volume 33, Number 3, 2006
14. Ramin Jamshidi, Lisa K. Hornberger, Tom R. Karl. Left Ventricular Aneurysm in a Child With Severe Combined Immunodeficiency Syndrome *Ann Thorac Surg* 88:1678 – 80
 15. Cikirikcioglu M, Antal AD, Tatar T, Didier D, Kalangos A. Surgical repair of a congenital left ventricular aneurysm. *J Card Surg* 2007;22:62–5
 16. or V. Left ventricular aneurysms: the endoventricular circular patch plasty. *Semin Thorac Cardiovasc Surg* 1997;9:123–30.
 17. Menicanti L, Di Donato M. The Dor procedure: what has changed after fifteen years of clinical practice? *J Thorac Cardiovasc Surg* 2002;124:886 –90.
 18. Tellez G, Nojek J, Juffe A et al. Cardiac echinococcosis: Report of 3 cases and review of the literature. *Ann Thorac Surg* 1971; 21:425.
 19. Hosseinian A., Mohammadzadeh A., Shahmohammadi G., Hasanpour M., Maleki N., Doustkami H., Haghdooost Z. Rupture of a giant cardiac hydatid cyst in the left ventricular free wall: successful surgical management of a rare entity. *Am J Cardiovasc Dis* 2013;3(2):103-106
 20. Kardaras, F., Kardara, D., Tselikos, D. et al. Fifteen year surveillance of echinococcal heart disease from a referral hospital in Greece. *Eur Heart J*, 1996, 17, 1265–1270

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