### A Large Intracardiac Hydatid Cyst with Concomitant Cervical and Hepatic Involvement: A Case Report

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#### Abstract

Cardiac hydatidosis is a relatively rare complication of echinococcosis, with a potentially life-threatening condition. Here, we reported a large interventricular septal hydatid cyst with bulging in the left ventricle accompanied by a huge cervical lamp with recurrent hepatic cysts that underwent cardiac surgery to excise the cyst uneventfully.

#### Title page

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## A Large Intracardiac Hydatid with Concomitant Cervical and Hepatic Involvement: A Case Report

#### Key Clinical Message

Cardiac hydatidosis is a relatively rare complication of echinococcosis. Understanding the atypical manifestations, potential associated risk factors, and epidemiology leads to optimal and timely management.

#### Abstract

Cardiac hydatidosis is a relatively rare complication of echinococcosis, with a potentially life-threatening condition. Here, we reported a large interventricular septal hydatid cyst with bulging in the left ventricle accompanied by a huge cervical lamp with recurrent hepatic cysts that underwent cardiac surgery to excise the cyst uneventfully.

#### Introduction

A 68-year-old woman with a history of hepatic hydatidectomy two years ago complained of a large right-sided cervical mass. In ultrasound imaging, a huge  $(45 \text{mm} \times 75 \text{mm})$  mass was detected in the base of the right side of the neck. Moreover, multiple cystic lesions involving the two lobes of the liver were found. Despite the absence of cardiac symptoms, transthoracic echocardiographic examination revealed a large  $(33 \text{mm} \times 42 \text{ mm})$  echo lucent cystic mass plugged into the interventricular septum. A positive serology test confirmed the diagnosis of hydatidosis. Treatment with antiprotozoal medication was started, and after two weeks, the patient successfully underwent surgical hydatidectomy using the cardiopulmonary bypass. This case highlights the rare concomitant cardiac, cervical, and hepatic involvement with hydatid disease. Together, a good understanding of the atypical manifestations, potential associated risk factors, and epidemiology leads to the optimal and timely management of such patients to minimize worse outcomes.

#### Case Presentation

A 68-year-old woman presented to a health care center with a clinical manifestation of a slow-growing and painless lump on the right side of the cervical region over several weeks. She had no cardiac symptoms. History-taking revealed working in a sheep-farming area in her twenties. Past medical and surgical history included hypertension and hepatic hydatidectomy two years ago.

Clinical examination through ultrasound imaging revealed a  $47 \text{mm} \times 75$  mm cervical cyst expanded to superior mediastinum with neither inflammatory response nor spasm of the cervical muscles. The cervical cyst consisted of a bilayer membrane with several membrane-attached scolices, indicating an active hydatid cyst (cystic echinococcosis type 1, CE1). The lesion was lateral to the common carotid artery and posterior to the internal jugular vein with no cervical lymphadenopathy. Besides, the abdominal ultrasound examination showed multiple active, recurrent hepatic cysts in both the right and left lobes (stage 1), encompassing all liver segments. There was no evidence of biliary dilatation as well.

In the transthoracic echocardiographic (TTE), a bulging and well-defined echo-lucent cystic mass in the interventricular septum measuring  $33 \times 42$  mm was detected (figure 1). A slight compression effect was present on the right ventricle (RV) cavity. The LV size and LV outflow tract (LVOT) were normal, with a mild systolic dysfunction (eye-ball estimation of LV ejection fraction = 45-5%). The valvular functions were normal, with no pericardial effusion. Other echocardiographic findings were unremarkable. The hydatid serology was positive, in which the enzyme-linked immunosorbent assay (ELISA)-based qualitative assessment of *E. granulosus* IgG antibodies confirmed the echinococcosis. Finally, the patient underwent cardiac surgery using cardiopulmonary bypass (CPB) for cystectomy to minimize the risk of spillage of cyst contents. The CPB technique was established by cannula inserting into the ascending aorta, superior vena cava (SVC), and inferior vena cava (IVC) after the routine median sternotomy. Following the cold cardioplegia, the established hypothermia was recorded at 32 °C. The outlines of the isolated cardiac cyst seemed to be complete and clear. Conservative procedures were further performed to sterilize and evacuate the cyst contents. The RV cavity was entered, and the cyst was exposed carefully. Thereafter, ten milliliters of its contents were aspirated.

An equal amount of hypertonic saline (NS 20%) was injected into the cyst, and after several minutes, the exposed cyst was evacuated completely (figure 2). Following successful excision and secured hemostasis, the cyst specimen, containing 8 ml colorless turbid fluid, was sent to the histopathological examination, which further vouched for the diagnosis of a hydatid cyst (figure 3).

#### Discussion

Due to well-developed transportation, echinococcosis, as a zoonotic disease, has become a serious global health problem, affecting more than one million people by hydatid disease worldwide [4]. Compared with visceral hydatidosis commonly occurring in the liver, cardiac HC is presented by wide clinical manifestations, leading to an early diagnosis challenge [13]. According to the WHO-Informal Working Group on Echinococcosis (WHO-IWGE) ultrasound classification, hydatid cyst consists of three stages, including active (CE1, CE2, with a high risk of rupture), transitional (CE3), and inactive or calcified cysts (CE4, CE5, with a low risk of rupture) [1]. Although most cardiac hydatic cysts in the literature are reported in young patients, here. we reported an old lady (68 y/o) with a recurrent hepatic hydatidosis accompanied by a huge homogenous cystic mass in the cervical and intracardiac regions. In our case, the cystic lesions in the vicinity of portal venous confluence and the left portal vein may be considered the leading cause of extra-hepatic hydatidosis. It is worth noting that the multiplicity and dispersion of the lesions, typical imaging findings, a history of husbandry procedures, a history of hepatic cysts, geographic location, and positive result of serologic test strongly established the hydatid cyst diagnosis. A previous study represented a 70 y/o female with no history of being in a sheep-rising area with signs in favor of right heart failure and cardiac hydatidosis complicated hydatid cyst and pre-tamponade [14]. Shojaei et al, in Iran, also indicated a cardiac HC in a 70-year-old farmer with dyspnea. The isolated lesion was diagnosed by echocardiography and further confirmed with cardiac MRI. Despite successful surgical excision, he died due to a progressive arrhythmia [15]. Another report in Iran has also documented an echinococcal infection involving an intramyocardial multicystic lump in the posterolateral and basal inferoseptal segments of LV in a 57-year-old farmer man referred with chest pain, and diagnosed by echocardiography, CMR, and positive ELISA-based serologic test. In contrast to our finding, EKG examination showed pathological Q and negative T waves. Similarly, surgery was the treatment of choice, followed by albendazole as a complementary therapy [3].

In conclusion, a good understanding of the atypical manifestations, potential associated risk factors, and epidemiology lead to the optimal and timely management of patients with rare echinococcosis to minimize worse outcomes.



Figure1: Transthoracic echocardiography shows (A, D) in a four-chamber view a large, well-defined intramyocardial cystic lesion (with no obvious septation) in the mid part of the septum with mild compression effect on RVOT without gradient. (B, C) The parasternal long axis and short axis view revealed a cystic lesion.



Figure 2: (A) Surgery revealed a complete and clear cardiac cyst (B, C, D). After sterilizing and evacuating the cyst contents, the cardiac cyst was completely resected upon aspiration.



Figure 3: (A, B) Protoscolices of Echinococcus granulosus in cytology,  $\times 400$  magnification, Papanicolaou Stain.

#### References

1. Ameen A, Hilal K, Shaikh A, Khan F, Fatimi S. Cardiac hydatid cyst presenting as ventricular arrhythmia: a case report. The Egyptian Heart Journal. 2021;73(1):105.

2. Separovic Hanzevacki J, Gasparovic H, Reskovic Luksic V, Ostojic Z, Biocina B. Staged management of a giant cardiac hydatid cyst: a case report. BMC Infectious Diseases. 2018;18(1):694.

3. 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):75-7.

4. Dong Z, Yusup M, Lu Y, Tang B. Hydatid cyst of the heart as a rare cause of arrhythmia: A case report and review of published reports. HeartRhythm Case Reports. 2022.

5. Mesrati MA, Mahjoub Y, Abdejlil NB, Boussaid M, Belhaj M, Limem H, et al. Case Report: Sudden death related to unrecognized cardiac hydatid cyst. F1000Research. 2020;9.

6. Bajdechi M, Manolache D, Tudor A, Orghidan M, Gurghean A. Cardiac hydatid cysts in a young man: A case report and a literature review. Experimental and Therapeutic Medicine. 2022;24(3):1-10.

7. Manuel V, Neto MP, Garcia ZS, Delgado C. Swiss Cheese Heart: Cardiac Hydatid Cysts. Canadian Journal of Cardiology. 2022.

8. Oner T, Korun O, Celebi A. A cardiac hydatid cyst mimicking a pericardial tumour in a paediatric case. Cardiology in the Young. 2019;29(2):244-6.

9. Gupta A, Mishra SC, Jaiswal S, Pande S. Intracardiac hydatid cyst located in right ventricular outflow tract: a rare site. Indian Journal of Thoracic and Cardiovascular Surgery. 2021;37(5):588-90.

10. Al-Dairy A, Abo Kasem R. Surgical excision of a cardiac hydatid cyst from the right ventricle in a child. Clinical Case Reports. 2021;9(8):e04714.

11. Kankilic N, Aydin MS, Gunendi T, Goz M. Unusual hydatid cysts: cardiac and pelvic-ilio femoral hydatid cyst case reports and literature review. Brazilian Journal of Cardiovascular Surgery. 2020;35:465-72.

12. AlShamlan RA, Almousa AM, Al Saeed MJ, Al-Dera FH, Alobaydun MA. Cardiac hydatid cyst successfully managed with Albendazole: A case report. Cureus. 2019;11(12).

13. Bzikha R, Bouhmou A, Messouak M. Cardiac hydatid cyst disease in a young patient. Cirugia Cardiovascular. 2021;28(5):290-2.

14. El Boussaadani B, Regragui H, Bouhdadi H, Wazaren H, Ajhoun I, Laaroussi M, et al. Primary cardiac hydatid cyst presenting with massive pericardial effusion: a case report. The Egyptian Heart Journal. 2020;72(1):1-4.

15. Shojaei E, Yassin Z, Rezahosseini O. Cardiac hydatid cyst: a case report. Iranian journal of public health. 2016;45(11):1507.

