

Case Report



Pulmonary Artery Embolism Due to a Ruptured Hepatic Hydatid Cyst Into the Inferior Vena Cava: Clinical and Radiologic Imaging Findings

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ABSTRACT

Background and Aim: Pulmonary embolism because of hydatid cysts is a very uncommon and lethal complication caused by a hydatid heart cyst rupture or a visceral hydatid cyst released into the venous circulation. By utilizing contrast-enhanced computed tomography (CT) and magnetic resonance imaging (MRI), hydatid pulmonary embolism can be differentiated from other types of pulmonary embolism. MRI mainly displays the cystic nature of lesions better than CT. Pulmonary embolism should be kept in mind in patients with hepatic hydatidosis if there is a sudden occurrence of chest pain and dyspnea, particularly in regions where hydatidosis is endemic. This report aims to present the clinical and radiographic features and discuss the diagnosis and treatment procedure of our patient.

Case Presentation: Here, we report a 45-year-old man with pulmonary embolism as a consequence of a ruptured hydatid liver cyst in the inferior vena cava. Multiple intra-arterial pulmonary hydatid cyst emboli originating from a hepatic hydatid cyst ruptured into the hepatic portion of the inferior vena cava were seen in our patient. The patient refused the surgical treatment. Therefore, the patient was treated using Andazol (Albendazole) and Cetirizine hydrochloride.

Conclusion: The present case is interesting because pulmonary embolism caused by hydatid cysts is a very uncommon clinical entity. There may be difficulties in diagnosing and treating hydatid cysts, and a definitive diagnosis was possible only by a histopathological examination.

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1. Introduction

Hydatic cyst, also known as Echinococcosis, is a zoonotic disease caused by tapeworm larvae of the genus *Echinococcus granulosus* [1]. Canidae, the final host for this parasite, pass the parasite eggs via defecation. Humans may be infected by the ingestion of fluids and foods contaminated by these eggs [2]. Echinococcosis is mainly endemic in sheep-raising areas such as South America, the Middle East, the Mediterranean coast, and Oceania [3].

The prevalence of Echinococcosis cyst is 6.8 cases per 1000000 in Iran. The Northeast (15.2%) and Southeast (0.7%) of Iran have the greatest and lowest prevalence rates, respectively. A significant difference is reported between males and females in terms of prevalence (5.8 versus 7.9, $P=0.001$) [4].

This parasite usually affects the liver and the lungs (55-75% and 15-25%, respectively), but it can affect any other organ (kidney, muscles, skin, etc.). The cardiovascular system may also be involved in less than 2% of cases. Hydatid pulmonary embolism is an unusual disease. It is usually seen in cardiac hydatidosis following the rupture of the cyst wall in the atrium or right ventricle. More rarely, it can be seen following the rupture of the hydatid cyst into the hepatic vein or the inferior vena cava (IVC) and spread to the lungs via the bloodstream [3, 5].

We report a case of hydatid cyst embolization to pulmonary arteries through the IVC vein. The diagnosis was made clinically and confirmed by biological and radiological findings. The aim here is to investigate the radiological and clinical findings of this rare complication.

2. Case Presentation

A 43-year-old man was referred to our radiology center to evaluate his intermittent hemoptysis. He had abdominal pain in the right upper quadrant (RUQ), chest pain, mild dyspnea, and intermittent cough for three weeks before admission.

He had liver hydatid cyst marsupialization 20 years ago. His medical history was otherwise unremarkable.

On physical examination, he had RUQ tenderness. All laboratory findings were within normal range, except for white blood cell count (17000 cells/cum), eosinophilia

(>5%), and the presence of anti-hydatid cyst antibody (measured by ELISA).

Abdominal ultrasonography (US) reported multiple cystic structures within hepatic parenchyma; some of which were calcified, and one had a compressing effect on the IVC lumen, with its germinal membranes having developed into the IVC. Suspecting a pulmonary embolism in the patient, a pulmonary CT-angiography was performed. Its abdominal cuts confirmed the presence of calcified (i.e. former) cysts and a filling defect in the intrahepatic segment of the IVC (Figure 1 A and B, respectively).

The pulmonary cuts of the CT-angiogram showed several hypodense filling defects (Figure 2, A) (i.e. an expansible thrombosis, Figure 2 B) in the pulmonary arteries, multiple cystic parenchymal masses in lower lobes of both lungs, and a cystic embolus in the right atrium.

The diagnosis was partial pulmonary embolism due to a hydatid cyst. The patient refused the surgical treatment. Besides, pulmonary embolectomy was not an option for treatment due to the risk of an anaphylactic reaction. Therefore, the patient was treated using Andazol (Albendazole) 10 mg/kg/day for a 30-day course, in 2 divided oral doses, and Zyrtec (cetirizine hydrochloride) daily oral 10 mg tablet. After ten days, the patient's dyspnea and chest pain disappeared, and no progression was seen in his hepatic lesions. After two months, some arterial lesions had diminished, and the patient had no more symptoms.

Unfortunately, no clinical data is available on the patient's follow-up.

3. Discussion

Hydatid cysts are caused by the cystic or larval stage of *Echinococcus granulosus*. Organisms in hepatic cysts are carried to the right atrium and lungs via IVC and pulmonary arteries. They may be brought to a different organ via systemic circulation [6]. Anaphylactic shock, tamponade, and rupture of the cyst wall in the pericardium are the main complications. Hemoptysis is the most common symptom, but the clinical findings are not specific to pulmonary arterial hydatid cyst embolization [3].

It should be noted that the pulmonary arterial hydatid cyst embolization might cause sudden death (29% of cases) [7]. There have been a few case reports of embolization following the rupture of hepatic cyst into IVC [2]. In our case, hepatic cysts were developing in the IVC.

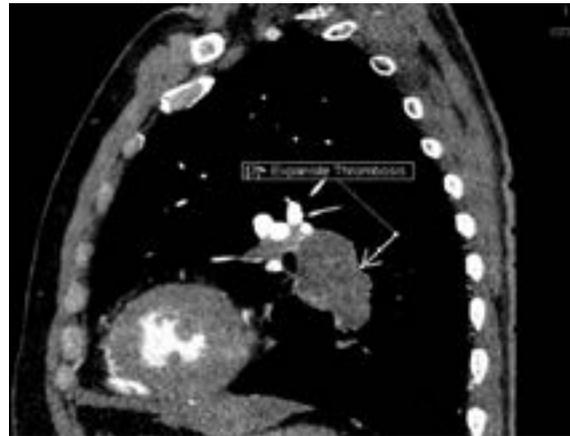


Figure 1. A) Multifocal calcified cystic structures within hepatic parenchyma. Calcifications indicate previous marsupialization. B) Filling defects in the hepatic section of the IVC.

We postulated that the larvae entered the right atrium via IVC, then moved to pulmonary arteries, and caused the embolism.

Laboratory studies, skin tests, and serology help diagnose the diseases due to hydatid cysts, but they lack prognostic value to determine the intravenous rupture of hydatid cyst. Clinical and radiological findings of hydatid pulmonary embolism make the diagnosis more effective. Intra-arterial cyst reveals the typical hypodense appearance on CT-angiography; therefore, no contrast enhancement in such a case is against a diagnosis of malignant structures [2]. Chest imaging may have an

essential role in guiding the diagnosis, especially CT-angiography and MRI. They can study the morphology of hydatid cysts, locate them, and reveal their extension to the vasculature [8].

Surgical treatment is the primary approach for pulmonary artery hydatid embolism. Often, embolectomy is the preferred option. The main complication of surgical intervention is a cyst and or arterial rupture during the surgery. Patients who refuse surgical intervention should be treated with Albendazole [2]. In our case, we treated the patient with Albendazole and Cetirizine hydrochloride because he refused surgical intervention.



Figure 2. A) Pulmonary lobar filling defects. B) Indicating an expansile thrombosis with a multicystic appearance.

4. Conclusion

A pulmonary embolus due to a ruptured hepatic cyst is extremely rare and can present similar to primary arterial neoplasms and pulmonary thromboembolism; therefore, in a patient with hydatid cyst near the IVC, it should be included within the differential diagnoses of an intra-arterial pulmonary mass.

Ethical Considerations

Compliance with ethical guidelines

The patient granted informed consent for the publication of this case report. Also, the authors have removed any identifying information from accompanying images to maintain patient confidentiality.

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Authors' contributions

Diagnosis the case and treatment: Parham Rabiee and Sajjad Rezvan; Searching the literature and writing the manuscript: Mohamad-Amin Khajeh-Azad, Mohammad-Hossein Mokhtarian, and Alireza Sharifi Mohamad-Amin Khajeh-Azad, Final editing: Mohammad-Hossein Mokhtarian.

Conflict of interest

The authors did not have any conflict of interest to report.

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