

Bilateral Pulmonary Artery, Inferior Vena Cava, and Cardiac Echinococcosis: A Rare Presentation of Zoonotic Diseases

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ABSTRACT

Cystic hydatidosis is a zoonotic disease that is mostly observed in the Mediterranean region. This infectious disease had different manifestations, which may lead to a delay in diagnosis process and various complications for the patients. Most of the cases are usually diagnosed through imaging techniques and its related management could be medical or surgical depending on the patient's clinical condition and disease severity.

This case study is a report of a 50-year-old female patient with a positive history of liver, splenic hydatid cysts, hemoptysis, and dyspnea. The diagnosis of pulmonary and cardiac involvement was made using computed tomography angiography. Despite the immediate surgery, the patient died from hemorrhage after 3 days.

The management of pulmonary embolism induced by *Echinococcus* highly depends on the clinical diagnosis. Moreover, the success of the surgery outcomes is related to the history and level of disease severity.

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Introduction

Cystic hydatidosis is a chronic parasitic disease that is caused by a tapeworm. This zoonotic disease can occasionally infect humans through contaminated foods. This parasitic disease is endemic in Iran; however, the incidence of human hydatidosis has been recently decreasing due to the enhancement of public health education and knowledge (1). Cystic hydatidosis can involve many body organs, especially the liver and lung. Although this issue is of significant importance, there is a dearth of research on the rare presentations of the disease in the literature (1).

Pulmonary or systemic embolisms caused by *Echinococcus* spp. are the rare complications of

hydatidosis. Cystopen to inferior vena cava and daughter cysts or cardiac cysts may rupture into pulmonary arteries and cause embolisms (2). The various manifestations of this disease may range from cough, dyspnea, chest pain to severe hemoptysis. The patients affected with this disease require prompt management, especially those with massive hemoptysis and chest pain. The management of these cases usually ends up with surgical procedures, and the survival depends on the severity of the disease and comorbid conditions (3). This study reported a case of pulmonary and the embolism of the right heart chambers due to the rupture of hydatid cysts.

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Case Presentation

The case in this study was a 50-year-old female patient with a previous history of liver hydatid cyst and spleen surgery for about 20 years ago. She was admitted to the emergency department due to recent exertional dyspnea (Functional class III), fever, and hemoptysis. The medical history of the patient was unremarkable, except for a previous surgery of liver hydatid cyst and a history of treated pulmonary tuberculosis.

At the time of admission, the vital signs included an axillary temperature of 37.8 °C, a respiratory rate of 20 breath/min, a pulse rate of 100 beats/minute, a systolic blood pressure of 140 mmHg, a diastolic blood pressure of 90 mmHg, and an oxygen saturation of 88%. In addition, pulmonary auscultation was unremarkable. Supportive therapy for massive hemoptysis was initiated following oxygen therapy and taking an intravenous line.

The patient's electrocardiography was unremarkable and no sign of leukocytosis or anemia was observed in the complete blood count. A high-resolution computed tomography (HRCT) was performed in order to determine the possible etiology and bleeding source of hemoptysis. The results of HRCT revealed scattered thick-walled cystic centers in the lung fields. The maximum diameter of these cysts with air-fluid level was 43*33 mm resembling hydatid cysts. Moreover, the increased diameter and tortuosity of pulmonary artery branches were noted.

Multiple multicystic zones were observed in the upper segment of the left liver lobe. Based on the results of HRCT and the cystic masses, an optic fiber bronchoscopy was performed; however, it failed to visualize any end bronchial lesions. According to the possible signs of pulmonary hypertension on HRCT, the patient was required to have transthoracic echocardiography. The results of transthoracic echocardiography revealed 50% of ejection fraction. The D-shaped configuration of left ventricular and abnormal septal motion was compatible with the right ventricular pressure overload.

The right atrium (RA) was severely enlarged (RA area: 33 cm²). In addition, there were a severe right ventricular enlargement and systolic dysfunction in the patient. Moreover, she was diagnosed with severe tricuspid regurgitation and pulmonary artery hypertension (110 mmHg). A hypermobile oscillating mass was attached to the entrance orifice of inferior vena cava protruding to RA. The inferior vena cava was in top normal size with a respiratory collapse higher than 50%.

According to the abovementioned results, the case was subjected to a color Doppler ultrasound

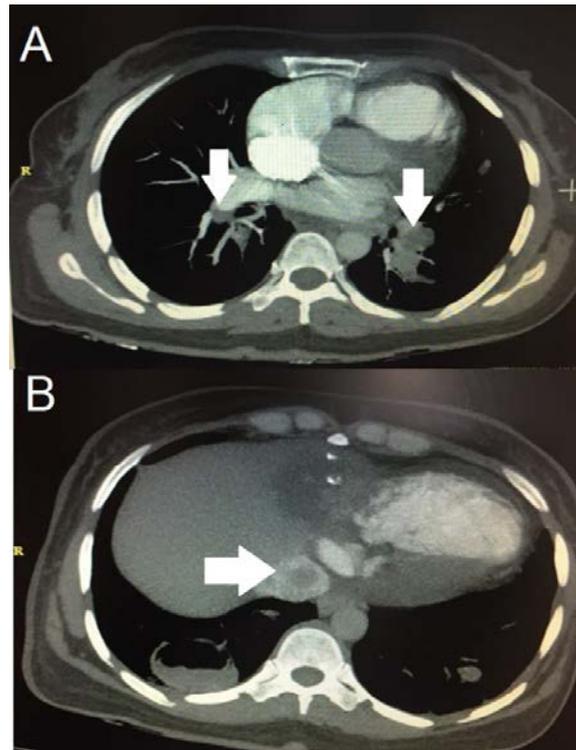


Figure 1. Computed tomography image; A) filling defects in both pulmonary arteries; B) an inferior vena cava filling defect extending toward the atrium

of the lower limbs and computed tomography angiography (CTA). The results of the lower limb Doppler ultrasound were normal. The results of thoracic CTA revealed filling a defect in RA extending toward the distal of inferior vena cava (figures 1 and 2). Multiple filling defects suggested the emboli lobar and segmental branches in the lungs.

The branches of pulmonary artery diameter were increased in contrast to other common embolism patterns. Multiple cystic masses were also present in the left lobe of the liver and near inferior vena cava resembling hydatid cysts. Overall, the results were indicative of hydatid emboli with pulmonary hydatidosis that could be secondary to the fistulization of the liver hydatid cyst into the inferior vena cava.

The diagnosis of the pulmonary artery and heart hydatid disease was established according to the results of CTA and echocardiography. Pulmonary hypertension was considered to be secondary to hydatid disease. The patient received 400 mg of albendazole twice a day for approximately 20 days. According to the mentioned lesions and the extension to heart and inferior vena cava, the patient was a candidate for the surgical resection of the cysts from the inferior vena cava, heart, and pulmonary artery.

The patient underwent a successful right-side pneumonectomy and the masses were removed. However, the patient passes away due to massive



Figure 2. Intra-atrial, intra-inferior vena cava, and pulmonary artery hydatid cysts

hemoptysis 3 days after the surgery. The pulmonary cysts, intra-cardiac cysts, and inferior vena cava cysts were confirmed by pathology examination as hydatid cysts (Figure2).

Discussion

Embolism caused by *Echinococcus* spp. is a rare complication and a life-threatening medical condition that requires prompt management. This zoonotic disease is common in the Mediterranean countries. However, the diagnosis of this well-known infectious disease is difficult in some cases and requires clinical attention. Fast progressive symptoms and mimic cardiologic or pulmonary disorders are regarded as rare presentations of this disease.

In this regard, it is of utmost importance to consider the patient's clinical medical history and clinical presentation to prevent unnecessary therapeutic procedures. Similar to the present report, these patients are prone to have a history of contact with farm animals, such as sheep or goat. Furthermore, a medical history of treated hydatid disease or suspected features of hydatid cysts using imaging techniques is another strong evidence for the presence of this infectious disease.

Similar to other clinical diseases, there are some important differential diagnoses for these patients. The primary atrial tumors and pulmonary thromboembolism are the two main differential diagnoses. Such diagnosis can be ruled out using imaging techniques with or without contrast. In the present case, CTA could clearly demonstrate the mass extension. The detection of hypodense masses within the pulmonary artery in patients with a longstanding history of hepatic hydatid cysts near inferior vena cava is a key for a timely diagnosis of pulmonary embolism due to hydatid disease (4).

In addition, hemoptysis in patients who are

prone to have hydatid cysts could be a warning sign of vascular involvement, which requires close vital sign monitoring and reanimation. The management of complicated cases of pulmonary embolism due to hydatidosis is controversial and strongly depends on the patients' clinical conditions. Those cases who refuse the surgery can be discharged by oral albendazole (5).

Pre-operative and post-operative medications for hydatid disease is effective for killing spilled scolices in ruptured cysts (6). Anaphylactic shock and vascular rupture are the main fatal complications of treating hydatid cysts. Moreover, these patients may chronically progress to respiratory failure and cor pulmonale (7). Pulmonary embolism caused by *Echinococcus* spp. is the rare complication of hydatid disease that could be a life-threatening condition. Although the managements of these cases are controversial and strongly related to clinical diagnosis, the surgery is the last option for some patients. The surgery outcomes depend on the patient's previous diseases and the level of disease severity.

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Conflict of Interest

The authors declare no conflict of interest.

References

1. Rokni M. Echinococcosis/hydatidosis in Iran. Iran J Parasitol. 2009;4:1-16.
2. Jaafari A, Nedia F, Boukhriss B, Ehlem B, Moez T, Habib BM. Fatal hydatid pulmonary embolism.

- Report of two cases. *Ann Cardiol Angeiol.* 2009; 58:125-8.
3. Yuan Z, Maihesuti M, Wenya L, Cheng Z, Biyemu N, Haitao W. Rare reason for pulmonary embolism: one case of pulmonary hydatid cyst and review of the literature. *J Thromb Thrombolysis.* 2015; 40:126-9.
 4. Akgun V, Battal B, Karaman B, Ors F, Deniz O, Daku A. Pulmonary artery embolism due to a ruptured hepatic hydatid cyst: clinical and radiologic imaging findings. *Emerg Radiol.* 2011;18:437-9.
 5. Poyraz N, Demirbaş S, Korkmaz C, Uzun K. Pulmonary embolism originating from a hepatic hydatid cyst ruptured into the inferior vena cava: CT and MRI findings. *Case Rep Radiol.* 2016; 2016:3589812.
 6. Ghassemof H, Esfehni RJ. Hydatid disease presented as acute abdomen, an interesting incidental finding: a case report. *Arch Clin Infect Dis.* 2015;10:e29632.
 7. Serraj M, Smahi M, Kamaoui I, El Houari A, Sahnoune F, Ouadnoui Y, et al. Hydatid pulmonary embolism: a rare complication of hepatic hydatid cyst. *Rev Mal Respir.* 2013;30:215-21.