CASE REPORT

Pulmonary artery embolism due to a ruptured hepatic hydatid cyst: clinical and radiologic imaging findings

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Abstract Hydatid pulmonary embolism is an uncommon condition resulting from the rupture of a hydatid heart cyst or the opening of a visceral hydatid cyst into the venous circulation. We report a rare case with multiple intra-arterial pulmonary hydatid cyst emboli originating from a hepatic hydatid cyst ruptured into the hepatic segment of the inferior vena cava. We present the ultrasonography findings of hepatic hydatid cyst and multidetector computed tomography pulmonary angiography images demonstrating both multiple hydatid cyst emboli and their hepatic origin.

Keywords Hydatid disease · Pulmonary embolism · Ruptured hydatid cyst · MDCT · IVC

Introduction

Echinococcosis is a parasitic disease affecting most commonly the liver and the lung [1, 2]. Pulmonary artery embolism due to hydatid cyst is an extremely rare entity that can be seen secondary to cardiovascular system invasion. It is usually seen in cardiac involvement but it can be also seen due to inferior vena cava (IVC) or hepatic vein invasion. It may have an acute fatal or subacute or chronic clinical presentation. But, in any of these clinical

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forms, the prognosis of pulmonary embolism due to hydatid disease is poor. We present ultrasonography (US) and multidetector computed tomography (MDCT) pulmonary angiography findings of a case of echinococcal embolization to the pulmonary artery.

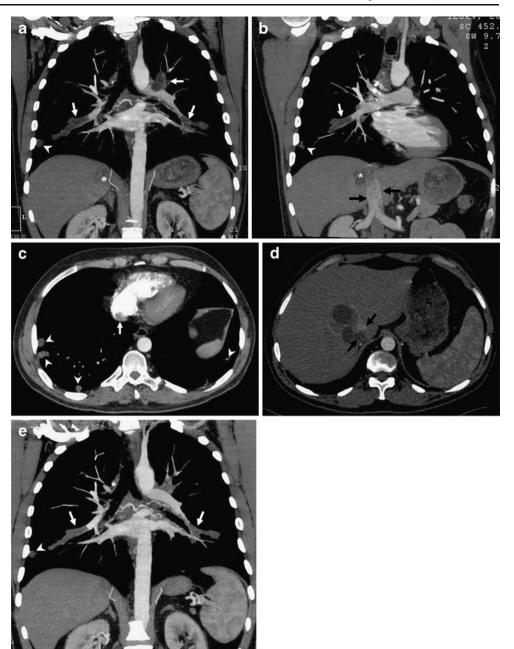
Case report

A 43-year-old male presented to our radiology unit for an evaluation of intermittent hemoptysis. Three weeks prior to admission, he experienced a pain in the right upper quadrant of the abdomen and episodes of shortness of breath and chest pain. A MDCT pulmonary angiography was performed for a suspicion of a pulmonary embolism. The MDCT examination revealed multiple cystic emboli in the pulmonary arteries causing vessel enlargement, multiple cystic parenchymal nodules predominantly located in the lower lung lobes, and cystic embolus in the right atrium. In addition, we determined a multilocular mass with solid and cystic components adjacent to the IVC in the liver parenchyma. On CT images, the border between IVC and cystic component of the mass was undetermined and the intrahepatic segment of the IVC was not totally filled with contrast agent (Fig. 1). Furthermore, an abdominal US examination was performed and a 4 cm type 3 cyst hydatid (daughter cysts filling the lesion) located in the liver adjacent to the IVC was demonstrated. The lumen of the IVC was partially thrombosed or compressed due to vascular wall destruction and hydatid emboli. On sonographic images, we revealed that the border between the wall of the hydatid cyst and IVC was missing (Fig. 2).

The patient refused surgical intervention for the semisolid cystic mass adjacent to hepatic segment of IVC.



Fig. 1 Pulmonary artery embolism due to a ruptured hepatic hydatid cyst in a 43-year-old man is well-demonstrated on coronal maximum intensity projection (MIP) image (a), oblique coronal MIP image (b) and axial CT images (c, d). MDCT images show complete occlusion and vessel enlargement of the right and left inferior segmental, and the left superior segmental pulmonary arteries by cystic lesions (mean density value 18 HU) (white arrows in a, b). In addition, MDCT images also show multiple cystic parenchymal nodules (white arrowheads), cystic embolus in the right atrium (white arrow in c), and hepatic hypodense cystic mass (asterisk) adjacent to the IVC (black arrows). In followup MDCT examination, coronal MIP image (e) shows narrowing of some pulmonary arterial lesions (white arrows) causing arterial expansion



Pulmonary embolectomy was not decided as a therapeutic option due high risk for an anaphylactic reaction. For these reasons, the patient was treated with albendazole (Andazol®) 10 mg/kg/day in two divided oral doses and cetirizine hydrochloride (Zyrtec®), oral 10 mg tablet, once a day of a 30-day duration. After a 15-day interval, treatment was restarted and is still going on. In the following MDCT and US controls, no progression was determined in the liver lesion. On the other hand, some of the pulmonary arterial lesions causing arterial expansion were narrowed, and a few of the parenchymal lesions became smaller (Fig. 1e). The patient became asymptomatic during the second month of the medical therapy.

Discussion

Echinococcus granulosus is the causative parasite for cystic hydatid disease that is endemic in sheep-raising areas. Carnivores are the definitive hosts while humans are "accidental" intermediate hosts. This parasitic infestation is seen in humans by ingestion of eggs passed into the feces of infected carnivores. After ingestion, the embryos in the eggs liberate and migrate to the liver, lung, and many other organs. In these organs, they develop into metacestode forms and form different types of hydatid cysts. Liver and lungs are the two most common organs involved [3].





Fig. 2 Axial US image shows a 4 cm type 3 hydatid cyst located in liver adjacent to the IVC (*white arrows*). The missing border between the wall of the hydatid cyst and IVC, and incomplete thrombosis of the hepatic segment of IVC are well-demonstrated

Pulmonary embolism due to hydatid disease is rarely seen and usually has been reported as a serious complication of cardiac hydatid disease [4, 5]. But this complication may also develop after invasion of the cardiovascular system components other than the heart. There are a few reports of embolization following cyst rupture into the IVC or hepatic veins based mainly on postmortem examinations [6, 7]. In our patient, cyst rupture into the IVC was the cause of chronic embolism of the pulmonary arteries.

Clinical presentation of the hydatid pulmonary embolism mimics that of thromboembolic disease. It may cause hemoptysis and hemorrhage due to invasion of vascular structures such as bronchial and pulmonary arteries. It can also cause chest pain, dyspnea, cough, and/or an anaphylactoid reaction [7]. Hydatid pulmonary embolism has been classified clinically based on presentation as acute fatal, subacute with pulmonary hypertension and death in less than 1 year, or resulting in chronic pulmonary hypertension [4]. Our case is a chronic hydatid pulmonary embolism stabilized with a medical therapy lasting for a year following an acute onset.

The diagnosis of the hydatid pulmonary embolism is made by the clinical and radiological findings. On enhanced CT, the intra-arterial cyst shows the typical hypodense appearance. The differential diagnosis in such cases should include other conditions associated with intraluminal defects such as pulmonary thromboembolism and primary arterial tumors [8]. The acute thromboembolic diseases are usually excluded by clinical means. In our case, no predisposing factor responsible for thromboembolic disease was noted and there was no deep vein thrombosis in the lower extremities. Intra-arterial hypodense masses did not show contrast enhance-

ment. That finding excluded primary arterial tumors that behave more aggressively and show contrast enhancement. Abdominal US examination is a helpful diagnostic method in determining the origin of the pulmonary hydatid emboli. The missing border between the wall of the hydatid cyst and IVC or hepatic veins can be well-demonstrated by US.

Surgical interventions such as embolectomy and/or enucleation are preferred treatment choices. However, rupture of the artery and/or the cyst during surgical intervention may cause dissemination of the disease, anaphylactic shock, embolism, and pseudoaneurysm formation [9]. The degree of the degenerative changes in the arterial wall, the localization of the lesions, and parenchymal changes should be evaluated well before a surgical intervention [10]. In the cases with recurrent hydatidosis, disseminated disease (including secondary lung or pleural hydatidosis), high risk of morbidity or mortality, and risk of spillage of hydatid fluid during surgical intervention and medical therapy, even when not curative, can improve the length of survival [1].

In conclusion, the location of echinococcal cysts inside pulmonary artery is extremely rare. Pulmonary hydatid cyst emboli should always be one of the differential diagnoses of the hypodense intra-arterial pulmonary mass in a patient with hepatic hydatid cyst adjacent to inferior vena cava or hepatic veins and radiological features of such cases should be well-known by the radiologist.

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