1. Introduction

Pulmonary hydatid cyst embolization is a rare complication of cardiac or hepatic echinococcosis. A review of the world medical reports has revealed only a few cases, most of them diagnosed at autopsy. We report our clinical and surgical therapeutic approach in a case of a patient who developed acute pulmonary embolism due to multiple hydatid cysts.

2. Case report

A 57-year-old male was admitted to our department with a 2-day history of left pleuritic pain and increasing shortness of breath. His past medical history revealed surgical excision of a hepatic hydatid cyst at age of 35, left liver lobectomy at age of 48 and cholecystectomy-choledochoduodenostomy due to recurrent abdominal echinococcosis at age of 49.

On admission, the patient was dyspneic and mildly cyanotic. On auscultation there was a faint systolic murmur 2/6 at the pulmonic area. Left lower lung was poorly ventilated. Liver and spleen were palpable but no render. There was no ascites or edema. Laboratory evaluation was within normal limits except a mild eosinophilia. The titer of anti-echinococcal antibodies was positive 1:3200 (ELISA, bioMérieux®, titer of negative control <1:100). ECG was normal. Two-dimensional echocardiogram showed no cysts in the cardiac chambers or pericardial cavity, but the pulmonary artery was distended. Chest roentgenogram proved a rounded mass at the left hilum and multiple smaller masses at the right lower pulmonary field. Spiral CT scan of the chest revealed complete occlusion of the left pulmonary artery and partial occlusion of distal branches of the right pulmonary artery by cystic lesions. Spiral CT scan of the abdomen showed multiple hydatid cysts in the peritoneal cavity. MR-angiography confirmed the presence of multiple cysts in the left pulmonary artery and some smaller cysts in the distal branches of right pulmonary artery (Fig. 1).

With the diagnosis of acute pulmonary hydatid embolization the patient was operated on an emergency basis. The operation was performed without using extracorporeal circulation through a left anterolateral thoracotomy. The pleural cavity was entered through the fifth intercostal space and the pericardium was opened anterior to the phrenic nerve. The main pulmonary artery was carefully dissected from the neighboring aorta along the fifth intercostal space and the pericardium was opened anterior to the phrenic nerve. The main pulmonary artery was carefully dissected from the neighboring aorta along with the proximal portion of the left pulmonary artery and a tape was passed beneath for proximal control. The surrounding tissues were protected with wet sponges soaked in 15% hypertonic saline solution. Left pulmonary artery was occluded proximally and a longitudinal arteriotomy was made in the extrapericardial portion. A significant number of hydatid daughter cysts were removed using a long sponge forceps (Fig. 2). A Fogarty catheter was then passed distally into each of the smaller branches and some smaller cysts were withdrawn. Bright red backbleeding was encountered indicating that the distal circulation was open and that the embolectomy was expected to be successful. The pulmonary artery tree was copiously irrigated with saline solution and the arteriotomy was closed with a running 5.0 polypropylene suture.
The postoperative course was uneventful. (Fig. 3) The patient was discharged on a regimen of albendazole, 800 mg daily for four cycles as treatment against abdominal hydatidosis. We noticed a spectacular regression of abdominal hydatid cysts and a moderate regression of distal pulmonary artery cysts. The patient is in close follow-up every 6 months in our department and after 42 months of follow-up, he is asymptomatic.

3. Discussion

Cystic hydatid disease or Echinococcosis is a parasitic infection caused by the small tapeworm Echinococcus granulosus. It has a wide geographic distribution and a human may be infected incidentally as intermediate hosts in the parasite’s life cycle. The parasite can reach any part of the body although the organs most commonly affected are liver (75%) and the lungs (15%). Heart is rarely involved with less than 2% of all cases [1].

Hydatid pulmonary embolism can occur either as a result of hepatic or abdominal cyst rupturing into the hepatic veins or the inferior vena cava or directly from the ruptured cyst in the right cardiac chambers. Surgical and autopsy findings indicate that the embolism is caused by vesicles or daughter cysts that act purely mechanically by obstructing the blood flow and there are no blood clots or added thrombosis. The cause of pulmonary embolism in our patient was probably the rupture of an abdominal cyst into the inferior vena cava, as there were disseminated hydatid cysts within his abdominal cavity.

Hydatid pulmonary embolism is classified in three groups according to the clinical presentation: (a) acute fatal cases; (b) subacute pulmonary hypertension with death in less than year and (c) chronic pulmonary hypertensive cases. The majority of cases appear to follow a course of prolonged pulmonary hypertension punctuated by acute embolic episodes [2].

The diagnostic investigation of patients with suspected hydatid pulmonary embolism should involve two-dimensional echocardiography, spiral CT scan, MRI, and possibly conventional pulmonary angiography. If there is no previous history of hydatid disease its existence can be suspected by the presence of anti-echinococcal antibodies and eosinophilia in blood tests. Eosinophilia is uncommon except from cyst rupture [2].

Two-dimensional echocardiography is the imaging modality of choice for locating hydatid cysts in the heart and pericardium. Spiral CT scan and MRI have been used successfully in the diagnosing hydatid cysts of the lungs and the heart. However, MRI is more advantageous than spiral CT-scan for examination of the heart and the great vessels, because with images in multiple phases it gives a more complete anatomic picture [1,3]. Although there has not been previous experience with MR-angiography in hydatid pulmonary embolism, we consider it pathognomonic and the additional performance of conventional pulmonary angiography unnecessary.

Median sternotomy and the use of extracorporeal circulation must be the surgical approach of choice, when cysts are present in the cardiac chambers [4]. We preferred the
approach of left thoracotomy, because there were no cysts within the cardiac chambers or the main pulmonary trunk and the main site of embolism was in the left pulmonary artery [5].

Medical therapy is advocated for patients with recurrent hydatidosis or in whom surgical intervention involves a high rate of morbidity or mortality [2]. Our patient had a significant regression of the abdominal cysts after four cycles of albendazole.

In conclusion, a high index of suspicion is required to diagnose hydatid pulmonary embolization. An extensive work up was carried out to this patient to rule out the presence of hydatid disease elsewhere in the circulatory system. Early diagnosis, particularly in the acute form of pulmonary embolization, is of utmost importance because surgical intervention is the only treatment that could be life saving.

References