

## A Two-Stage Approach to a Patient with Hydatid Cysts inside the Right Pulmonary Artery and Multiple Right Lung Involvement

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**We report on the surgical treatment of a patient in whom hydatid cysts inside the right pulmonary artery and multiple right lung involvement were detected. Since the right pneumonectomy carried a high risk of cyst rupture, and migration of the cysts to the opposite lung during ligation of the pulmonary artery, a two-stage surgical approach was scheduled. Hydatid cysts located at the proximal pulmonary artery were removed by performing a longitudinal arteriotomy along the pulmonary artery to the bifurcation. Subsequently, a right pneumonectomy was performed on a safe right pulmonary artery in a different session. (Ann Thorac Cardiovasc Surg 2006; 12: 349–51)**

**Key words:** Echinococcus granulosus, hydatid cyst, heart, pulmonary artery

### Introduction

Hydatidosis is a parasitic disease, mostly encountered in sheep and cattle-dealing areas of the world. It is an infestation caused by larvae of *Echinococcus granulosus* parasites. Since the parasite enters the human body via the gastrointestinal route, liver is the organ most commonly involved (75%). Lungs are the second most common (15–20%) sites, although the disease can be seen in any organ system.<sup>1)</sup>

Pulmonary artery involvement, which is very rare, can be seen either by direct implantation of the embryo to the arterial wall or by embolization of daughter vesicles from intracardiac cyst.<sup>2–4)</sup> Location of the cyst inside the pulmonary artery is associated with a high mortality rate due to secondary dissemination, embolization and anaphylactic shock after cyst rupture.<sup>5)</sup> Here, we present the management of a patient with multiple hydatid cysts in the right lung and right pulmonary artery.

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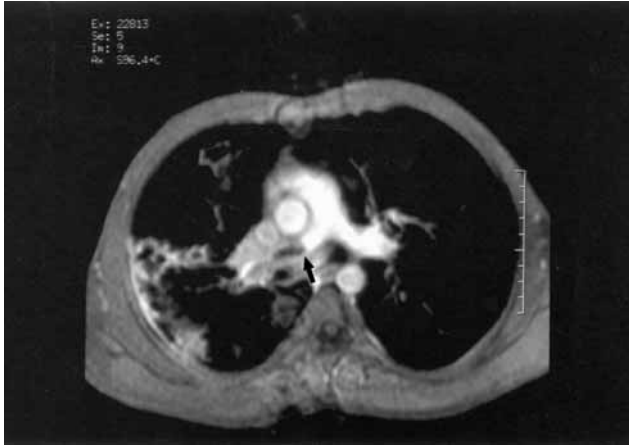
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### Case Report

A 24-year-old male patient was admitted to our hospital, complaining of cough and blood stained sputum for two weeks. He had undergone surgical resection of a hepatic hydatid cyst 12 years previously. A chest x-ray demonstrated a non-homogenous, irregular increased density in the hilum and lower zones, as well as an apical nodular lesion in the right lung.

Computerized tomography (CT) of the chest displayed a para-hilar cavitated mass-like lesion, a cavitary lesion in the posterior segment of the upper lobe and disseminated parenchymal infiltration in the right lung, together with multiple hilar and subcarinal lymph nodes. It was also noticed on CT that the right pulmonary artery was occluded with a hypo-dense lesion (Fig. 1). In fiberoptic bronchoscopy, the obstruction of right intermediary bronchus with white membranous structure was seen. Pathological examination of the biopsy revealed “the germinative membrane of a hydatid cyst”.

In order to demonstrate the lesion within the right pulmonary artery, magnetic resonance (MR) angiography was performed and, it showed complete occlusion of the right pulmonary artery (Fig. 2). In spite of complete obstruction of the right pulmonary artery, the patient had no complaints that could be attributed to pulmonary artery occlusion.



**Fig. 1.** CT scan showing right pulmonary artery occlusion with a para-hilar mass-like lesion and disseminated parenchymal infiltration (arrow, site of right pulmonary artery occlusion).



**Fig. 2.** MR angiography showing complete occlusion of the right pulmonary artery (arrow, site of right pulmonary artery occlusion).

The patient was scheduled for two-stage operation with the diagnosis of hydatid cysts inside the right pulmonary artery and the right lung. The patient was operated on via a median sternotomy approach to remove the cysts inside the right pulmonary artery. Standard cannulation for cardiopulmonary bypass (CPB) was established as the heart was beating. The right pulmonary artery was dissected from the main trunk to the truncus anterior branch of the right pulmonary artery without undue manipulation to prevent cyst rupture or migration. The right pulmonary artery was clamped at its proximal part intrapericardially. Then the artery was dissected outside the pericardium. Truncus anterior branch and descending interlobar pulmonary arteries were individually encircled and controlled with silastic loops. After that, a longitudinal arteriotomy was performed along the right pulmonary artery. We found that the right pulmonary artery was occluded with hydatid cysts and the arterial wall was thickened (Fig. 3). The endoarterial hydatid cysts were removed by forceps. The distal part of the pulmonary artery was then aspirated and cleared. The arteriotomy was extended until lobar arterial branching and the pulmonary artery was cleaned; and the back flow encountered was not satisfactory. After closure of pulmonary arteriotomy, the patient was de-cannulated. CPB time was 110 minutes.

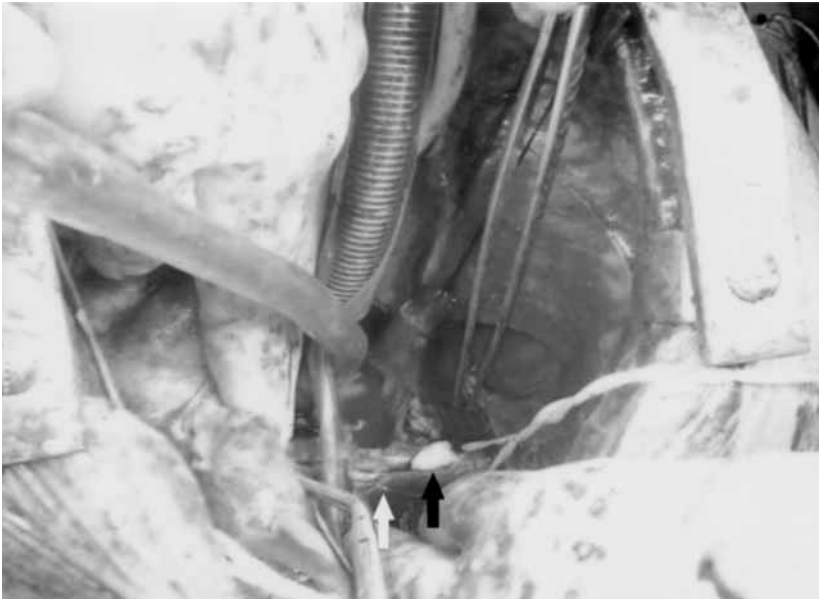
Two weeks after the first operation, the patient underwent a posterolateral thoracotomy and right pneumonectomy because multiple cysts in the lung had destroyed the lung parenchyma. Postoperative course was unevent-

ful and patient was discharged from the hospital on 8th postoperative day on albendazole therapy. He is in regular follow-up now after 14 months and no pathology has been detected.

## Discussion

Localization of hydatid cysts inside the pulmonary artery is extremely rare and there are few theories on this. Firstly; there is a hydatid cyst inside the heart chambers and due to rupture and embolism of these cysts, some embryos locate in the pulmonary artery wall resulting in hydatid cyst development.<sup>6)</sup> The second theory is that embryos settle in the artery wall coming directly from systemic circulation, probably from ruptured hepatic cysts.<sup>7)</sup> In our case, no cysts were observed at the cardiac level. However, the patient had a history of hydatid cyst in the liver, which was operated 12 years previously supporting the second theory.

As the patient had a pathologically proven hydatid cyst in the right lung and also absence of signs of acute thromboembolism despite total occlusion of the right pulmonary artery, it was reasonable to consider that the lesion inside the pulmonary artery was a slowly growing hydatid cyst. A hydatid cysts located in an organ is known to grow 2–3 cm per year. In the case of intra arterial location, they grow slowly into the lumen and finally occlude it. In the pulmonary arteries this process may last long enough allowing adequate pulmonary perfusion to be established through



**Fig. 3.** Germinative membrane of the hydatid cyst inside the pulmonary artery (black arrow, the germinative membrane; white arrow, the right main pulmonary artery).

the bronchial arteries. This may explain why our patient had no clinical signs of acute pulmonary artery occlusion in spite of total right pulmonary artery occlusion.

The preferred treatment for hydatidosis is surgery. This may or may not be followed by adjuvant medical therapy with albendazole. However, surgical treatment of hydatid cysts located inside the pulmonary artery is demanding and needs to be meticulous in its approach. Surgical intervention may be complicated by spillage of the cystic contents inflow direction, (which may lead to secondary dissemination), embolism, anaphylactic shock and pseudo aneurysm formation.<sup>2,4)</sup> Thus, surgical treatment has to focus on the prevention of the cyst migration and rupture, which may cause fatal complications. In previous reports, the surgical treatment of hydatid cysts located at the pulmonary artery is not detailed. Blind embolectomy, which is the mostly reported surgical technique, may cause rupture of the cysts and serious anaphylactic reactions. In the current case, the hydatid cysts located at the proximal pulmonary artery were removed via sternotomy by performing longitudinal arteriotomy under CPB on the beating heart. Since right pneumonectomy carried a high risk of cyst rupture or migration of the cysts to the opposite pulmonary artery during ligation of the pulmonary artery, a two-stage surgical approach was scheduled.

Due to the rarity of the event, no standard surgical technique has been recommended in the management of patients with hydatid cysts, located at the pulmonary artery. Two-stage operations with removal of the intra-arterial

cyst first and then the management of the lung cyst with parenchyma saving operation or lung resection is crucial in a patient with concomitant hydatid locations inside the pulmonary artery and the lung parenchyma. Surgery necessitates meticulous handling and appropriate technique to prevent dissemination, anaphylactic shock and embolism due to the cyst rupture and migration.

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