The Heart Surgery Forum #2003-91404
7 (1), 2004 [Epub November 2003]

Online address: www.hsforum.com/vol7/issue1/2003-91404.html

Hydatid Cyst in the Pulmonary Artery: An Uncommon Localization

(#2003-91404 . . . September 14, 2003)

Ihsan Bakir, Yavuz Enc, Sertac Cicek

Dr. Siyami Ersek Thoracic and Cardiovascular Surgery Center, Department of Cardiovascular Surgery, Istanbul, Turkey

ABSTRACT

Pulmonary artery involvement of hydatid disease caused by the *Echinococcus granulosus* parasite is an uncommon condition resulting from the opening of a visceral hydatid cyst into the venous circulation or the rupture of a cardiac hydatid cyst. We report a case of a 31-year-old woman with a hydatid cyst located in the right pulmonary artery. Clinical presentation was fatigue, cough, and dyspnea. Diagnosis was made by chest x-ray, computed tomography, and magnetic resonance imaging. The cyst was extracted under total circulatory arrest. Diagnosis and surgical therapy of the intraluminal pulmonary arterial hydatid cyst prevented possible occurrence of severe complications, such as cyst rupture, anaphylactic shock, and sudden death.

INTRODUCTION

Hydatidosis is an endemic zoonosis in Middle Eastern countries, the Mediterranean coast, and South America. Cardiac involvement with echinococcosis is an infrequent disease and is seen in 0.5% to 2% of patients with echinococcal disease [Dighiero 1958, Bayezid 1991]. Hydatid pulmonary embolism generally results from the rupture of a hydatid cyst in the right chambers of the heart or a venous migration of daughter vesicles to the right heart and then pulmonary arteries [Lahdhili 2002].

We report a case of hydatid cyst in the right pulmonary artery following opening of a visceral hydatid cyst into the venous circulation and discuss in detail the clinical and surgical approach to treatment.

CASE

A 32-year-old woman with a history of hydatid disease was hospitalized for fatigue, dry cough, and exercise-induced dyspnea. She had undergone hepatic lobectomy 5 years previously because of hydatid disease. During that procedure anaphylactic shock occurred due to cyst rupture.

Physical examination revealed crepitant and bronchial rales in the upper lung fields and a pulse rate of 60/min. Complete blood count and liver and cardiac enzymes were within normal limits.

The chest x-ray showed hilar opacities of the right lung and no evident anomalies of cardiac outlines. The serial electrocardiograms and the transthoracic echocardiography did not show any abnormalities. Computed tomography of the thorax showed a cystic lesion of the right pulmonary artery in the bifurcation region with a diameter of $3 \times 2 \times 2.5$ cm (Figure 1A). Magnetic resonance angiography of the thorax showed a perfusion defect in the middle and lower branches of the right pulmonary artery (Figure 1B). Abdominal ultrasonography was normal. Previous hepatic involvement of hydatid disease, imaging findings, and the fact that hydatid cysts are still endemic in Turkey led the authors to the diagnosis of hydatid disease in the pulmonary artery.

One week after the diagnosis, the patient underwent surgery through a median sternotomy incision and under guidance of transesophageal echocardiography (TEE). TEE showed no lesion in the cardiac chambers but a cystic lesion in the bifurcation region of the right pulmonary artery. Cardiopulmonary bypass (CPB) was initiated and the patient's body temperature was cooled to 16°C to commence total circulatory arrest (TCA). Surgical procedure included longitudinal incision of the distal portion of the right pulmonary artery under the superior vena cava and then puncture and aspiration of the cyst content, irrigation with hypertonic solution, and extraction of the cyst as far distally as possible. After irrigation with saline solution, the arteriotomy of the pulmonary artery was closed. TCA and CPB were terminated without any complication. Surgical findings were a 3×4–cm diameter cyst full of daughter vesicles that was located in the distal portion of the right pulmonary artery and extended into the middle and lower branches of the right pulmonary artery (Figure 2A). The cyst was composed of a laminated outer layer and an inner germinal layer filled with a transparent fluid (Figure 2B). Pathological analysis of the surgical specimen confirmed a hydatid cyst. The patient was extubated in the intensive care unit after 7 hours and discharged after 9 days with albendazole (10 mg/kg daily) treatment.

Received August 28, 2003; accepted September 14, 2003.

Address correspondence and reprint requests to: Ihsan Bakir, MD, Yazmaci Tahir Sok Derya Apt No. 11/4 Çatalçesme—Suadiye/Istanbul 81110 Turkey; 90-216-3727474; fax: 90-216-3734427 (e-mail: ihsanbak@yahoo.com).

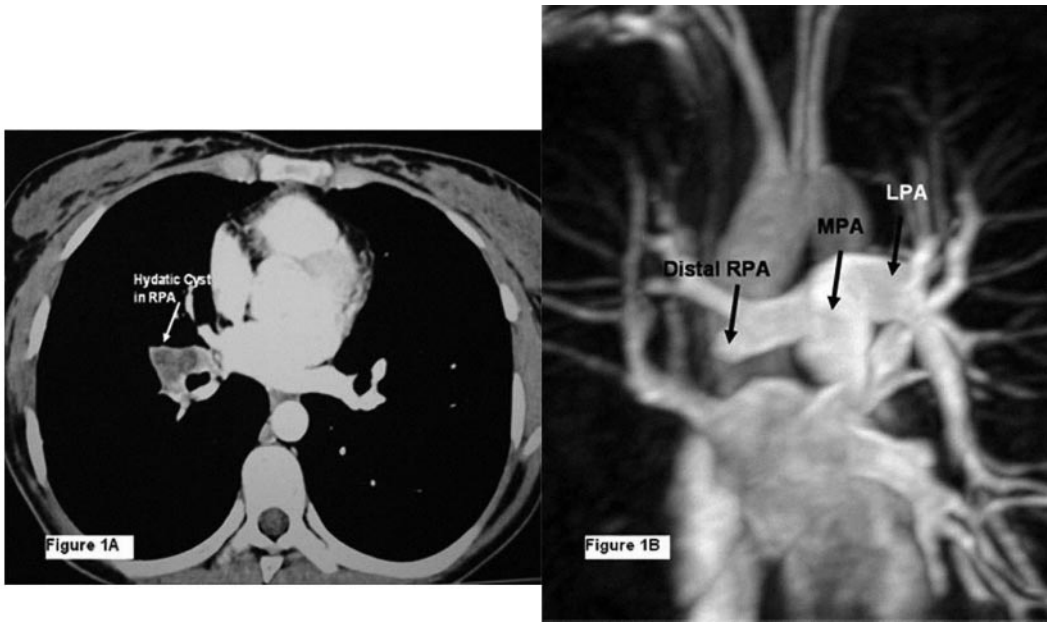

Figure 1. A, Computed tomographic image of hydatid cyst (arrow) occupying the right pulmonary artery (RPA) lumen. B, Magnetic resonance angiographic image showing obstruction of the distal part of the right pulmonary artery with a perfusion defect in the middle and lower branches of the RPA.

COMMENT

In human beings, the most frequent locations of hydatid cysts are the liver (in more than 65% of cases) and the lungs (25% of cases). A mean of only approximately 0.5% to 2% of cases of hydatid cysts are located in the heart [Miralles 1994]. Pulmonary embolism of hydatid disease generally occurs after a cardiac rupture of the right ventricle hydatid cyst or a venous migration of daughter vesicles to the right heart and then pulmonary arteries. The literature contains reports of approximately 50 cases of hydatid pulmonary embolism due to rupture of intracardiac hydatid cysts [Lahdhili 2002]. Although secondary dissemination by venous route is a well-known complication of surgery for hydatid cyst removal, reports of growth of hydatid cyst in the lumen of the pulmonary artery are extremely rare in the literature. In the

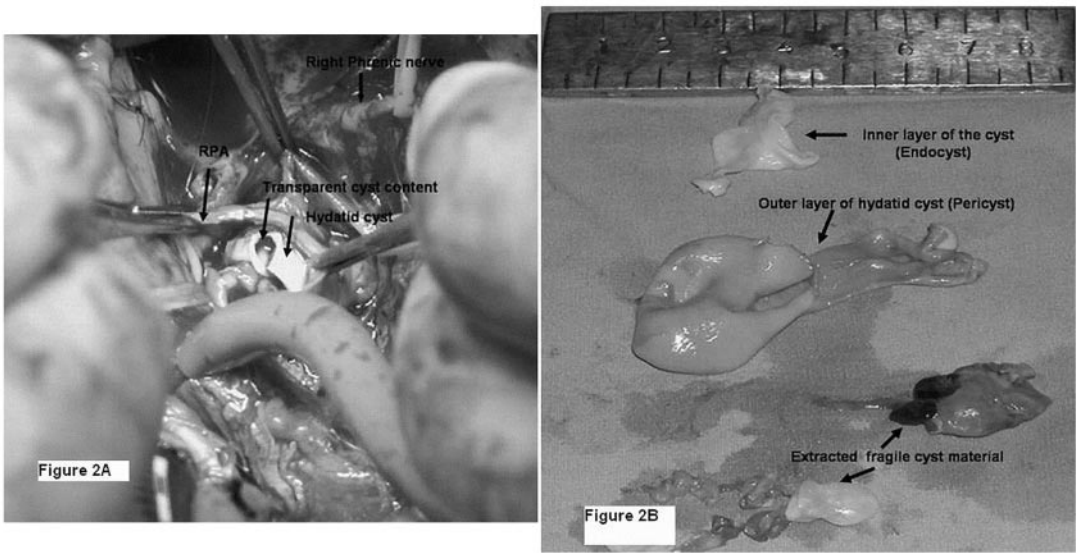

Figure 2. A, Surgical view of hydatic cyst in the right pulmonary artery. B, Extracted hydatid cyst and its content. Pathological analysis of the surgical specimen confirmed a hydatid cyst.

present case, secondary dissemination of hydatid disease by venous migration after left lobectomy of the liver performed 5 years previously led to the development of a cyst in the pulmonary artery.

The clinical symptoms induced by hydatid cysts are due to related complications, such as obstruction, rupture, or compression [Aydogdu 2001]. Symptoms may include anaphylactic reactions such as fever, eruptions, or even circulatory collapse when the cyst ruptures [Tejada 2001]. Although hemoptysis is the most frequent sign of hydatid pulmonary embolism, the presenting symptoms of our case included dry cough, fatigue, and exercise-induced dyspnea. The possible occurrence of complications, such as cyst rupture and sudden death, makes cyst extraction the most appropriate treatment [Miralles 1994, Tejada 2001].

Radiodiagnostic techniques and serologic tests can be used for diagnostic purposes [Oguzkaya 1997]. Chest x-ray, computed tomography (CT), and magnetic resonance imaging (MRI) are useful in making diagnosis and planning the surgical procedure. In our case, pulmonary hydatid cyst was confirmed by CT scan and magnetic resonance angiography. Hydatid cyst of the heart should be investigated if there is pulmonary hydatidosis. In the majority of cases hydatid cyst of the pulmonary arteries is caused by embolization due to rupture of hydatid cysts located in the heart [Lahdhili 2002]. Echocardiography is the imaging method of choice for diagnosis of cardiac cysts, even better than CT and MRI, which are more useful in the study of extracardiac echinococcosis [Men 1999, Tejada 2001]. The detection of eosinophlia in some patients provides very useful complementary data. Serologic tests have a higher sensitivity and specificity, but they are positive only in approximately 60% of pulmonary and 90% of hepatic lesions. The most useful tests are latex agglutination and immunoelectrophoresis, but excision and pathological examination of the lesion are required to confirm the diagnosis [Miralles 1994].

In the present case, median sternotomy with CPB and TCA was used to obtain a bloodless field and to decrease the risk of dissemination of the cyst content in case of any accidental cyst rupture. The sternotomy can also provide access to treat lung hydatid cysts during the same stage. Hydatidosis of the heart can be investigated with transesophageal echocardiography during the procedure, and any misdiagnosed hydatid cysts can be removed concurrently. In our case the postoperative period was uneventful and albendazole (10 mg/kg daily) was used during follow-up treatment.

Serologic tests and echocardiographic, CT, and MRI controls are recommended for 5 years after extraction to detect recurrences after surgical manipulation or cysts that were not detected at the time of surgery [Tejada 2001].

In conclusion, diagnosis of the disease by radiodiagnostic techniques and surgical therapy of the hydatid cyst in the pulmonary artery prevented possible occurrence of severe complications, such as cyst rupture, anaphylactic shock, and circulatory collapse. The most frightful complication is rupture during surgical manipulation, with subsequent dissemination and liberation of the cyst content, producing anaphylactic shock [Miralles 1994, Men 1999]. Given the significant risk of cyst rupture and anaphylactic shock, the operation should be performed in a short time period after diagnosis, and extraction of the lesion is recommended, even in asymptomatic patients.

REFERENCES

Aydogdu T, Sahin N, Ulusan V, Gurpinar F, Turkay C, Bayezid O. 2001. Right atrial hydatid cyst associated with multiple organ involvement: case report. J Thorac Cardiovasc Surg 121:1009-11.

Bayezid O, Ocal A, Isik O, Okay T, Yakut C. 1991. A case of cardiac hydatic cyst localised on the interventricular septum and causing pulmonary emboli. J Cardiovasc Surg 32:324-6.

Dighiero J, Canabal EJ, Hazan J, Horlales JO. 1958. Echinococcus: disease of the heart. Circulation 17:128-31.

Lahdhili H, Hachicha S, Ziadi M, Thameur H. 2002. Acute pulmonary embolism due to the rupture of a right ventricle hydatic cyst. Eur J Cardiothorac Surg 22:462-4.

Men S, Yucesoy C, Edguer TR, Hekimoglu B. 1999. Intraaortic growth of hydatid cysts causing occlusion of the aorta and of both iliac arteries: case report. Radiology 213:192-4.

Miralles A, Bracamonte L, Pavie A, et al. 1994. Cardiac echinococcosis. Surgical treatment and results. J Thorac Cardiovasc Surg 107:184-90.

Oguzkaya F, Akcali Y, Kahraman C, Emirogullari N, Bilgin M, Sahin A. 1997. Unusually located hydatid cysts: intrathoracic but extrapulmonary. Ann Thorac Surg 64:334-7.

Tejada JG, Saavedra J, Molina L, Forteza A, Gomez C. 2001. Hydatid disease of the interventricular septum causing pericardial effusion. Ann Thorac Surg 71:2034-6.