Hydatid Cyst in the Distal Pulmonary Artery: A Case Report

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Abstract

We report a 21-year-old female with two hydatid cysts localized in the right paracardiac zone and in the left lower lobe. She underwent sequential thoracotomies. Thoracotomy for the right side revealed a hydatid cyst in the pericardial cavity with tight adhesion to the right atrial wall. The second operation confirmed the presence of a distal intrapulmonary arterial hydatid cyst, which was treated with common basal segmentectomy. We report this case because of the rarity of the natural disease and also for choice of method of surgical intervention in such cases.

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Introduction

Hydatidosis is a parasitic infection caused by the larvae of *Echinococcus granulosus* and encountered mainly in Mediterranean countries. Cardiac involvement of hydatid disease is rare and occurs in 0.02-2% of all cases of human hydatidosis (1,2). *Echinococcus* infection of the pulmonary artery has also been reported in the absence of intracardiac and hepatic hydatid cysts (3) as a rare complication occurring after removal of a hepatic hydatid cyst (1). Here we describe a patient with an intrapulmonary arterial localization of hydatid disease without hepatic or cardiac chamber involvement, who was treated by segmental pulmonary resection. We believe there are no previous reports of similar patients who underwent a similar treatment.

Case Report

A-21-year old female patient was referred to our clinic for assessment. The patient had no complaints. She had undergone a left thoracotomy for hydatid cyst eleven years ago. During her medical follow-up, no evidence of hydatid disease was found until her last chest follow-up visit, at which time her chest x-ray revealed opacity in the left lower zone and computerized tomography revealed cystic lesions located in the right paracardiac zone and in the left lower lobe. Physical examination revealed no abnormal

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findings. The titer of anti-ecchinococcal antibodies was positive. MRI images of both lesions can be seen in Figures 1 and 2. Echocardiography revealed that the cystic lesion was located outside the pericardium. Sequential operations were planned. First a right thoracotomy was performed. The cyst was found to be intrapericardial in location, with tight adhesion to the right atrial wall. It was removed with partial resection of the right atrial wall and the pericardial defect was reconstructed with otologous pleura. After an uneventful postoperative period of 20 days, left thoracotomy was performed. After exploration and dissection of tight pleural adhesions, the cyst was noted to be located in the common basal segment of the lower lobe and was found to show continuity with the interlobar pulmonary artery. Needle aspiration from the enlarged and bulged left common basal segmentary artery revealed that the hydatid cyst was located in the pulmonary artery. Longitudinal arteriotomy was performed after dissection and clamping of the interlobar pulmonary artery. The left common basal segmentery artery was found to be completely obstructed at its origin by hydatic membrane. Hydatid daughter cysts were removed. Since no back flow was maintained and no regular intima was noted in the pulmonary artery, a formal left lower lobe common basal segmentectomy was performed.

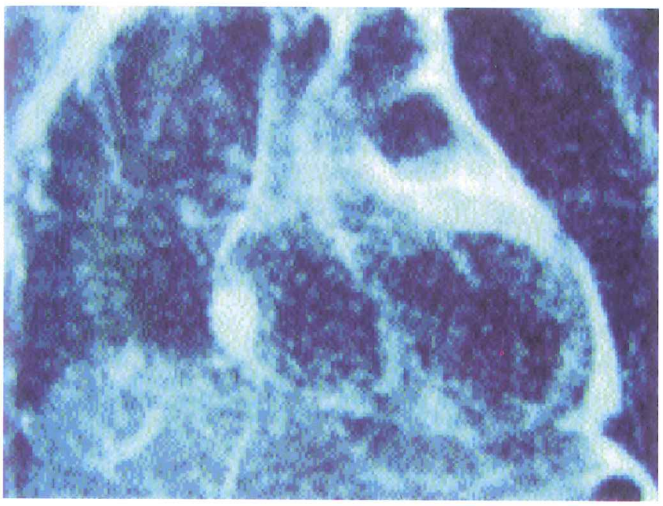

Figure 1. *Paracardiac localization of the hydatid cyst.*

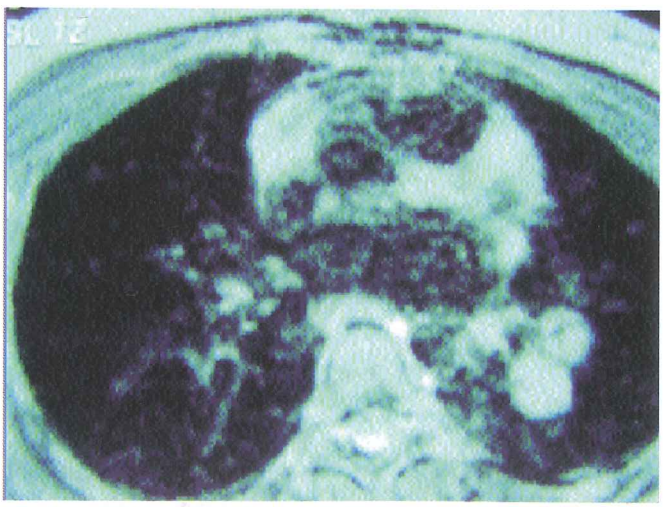

Figure 2. *MRI image of the hydatid cyst in distal pulmonary artery.*

The patient was discharged on the 5th day of the operation under albendazole (Andazol, Biofarma, Istanbul) treatment, to be given 3 times a day and continued for 3-4 weeks.

Discussion

It has been reported that hydatid cysts may rarely present within the pulmonary arteries as a result of the rupture of right atrial or ventricular cysts into the pulmonary arteries, leading to initially acute and then subacute and chronic recurrent embolization of the cysts (1-3). More rarely, hematogenous dissemination from a hepatic focus may cause intraarterial invasion (1). Other ways of dissemination, such as the parasite's crossing the arterial wall via previous small breaks of the intima which may occur in aneurysms, or by entering the vas nutritia, have also been suggested (1,5,6).

We believe that in our patient, the intrapulmonary arterial hydatid cyst was associated with the primary operation which had been performed 11 years ago and was due to dissemination of the parasites by crossing the arterial wall, as described above.

The surgical procedures used in the hydatid disease of the lung are well defined and include enucleation of the cyst, pericystectomy, cystotomy with or without capitonnage according to the localization (7). Birincioglu et al reported an off-pump technique for the treatment of 17 patients with cardiac echinococcosis and in long term follow-up of these patients, only one patient requires reoperation due to reoccurence of the lesion after 68 months (4).

To minimize the risk of rupture of the atrial cyst, we preferred to install surgical treatment before albendazole therapy. We preferred sequential thoracotomies instead of median sternotomy because it is difficult to perform left lower lung resections through median sternotomy. Presence of a large cyst involving more than 50% of the lobe, a cyst with severe pulmonary suppuration not responding to perioperative treatment, multiple unilobar cysts, and sequelae of hydatid disease, such as pulmonary fibrosis, bronchiectasis, or severe hemorrhage constitute the main indications for parenchymal resection (8). On the other hand, method of intervention in hydatid cysts located in the intrapulmonary artery is controversial. In the literature, different types of treatments have been reported in cardiac and intrapulmonary arterial hydatic cysts. Some have performed cardiopulmonary by-pass for the resection of cardiac and intrapulmonary arterial cysts. Embolectomy and enucleation tecniques have also been used for cysts located in the major pulmonary arteries (2).

Segmental resection for intrapulmonary arterial hydatic cyst has not been reported to date. Since the echocardiography,

CT and MR scans of our patient revealed that the intrapulmonary arterial hydatid cyst had a distal localization, we decided we did not need to prepare extracorporeal circulation for our patient. To protect our patient from complications due to the relapsing nature of hydatidosis, increased risks of complications by manipulation of the impacted cyst, we preferred performing a common basal segmentectomy. In our opinion, the two important points to be considered in systematic approach to intrapulmonary arterial localization of hydatid disease are 1) the localization of the pulmonary artery involved, and 2) the localization of the intrathoracic hydatid cyst.

If the pulmonary arterial localization is peripheral, it can be treated with resection which is more simple than a lobectomy, such as was performed in our patient. On the other hand, if the lesion is located in the right or left main pulmonary artery with or without involvement of contralateral pulmonary paranchyma, the surgeon should be prepared to perform a pulmonary embolectomy-cyst enucleation operation under cardiopulmonary by-pass with total circulatory arrest episodes. Decision on method of intervention definitely necessitates pulmonary hemodynamic and angiographic evaluation in the preoperative workup. The question is to be able to assess the right or left main pulmonary arterial localization and whether the contralateral parenchyma is completely clear or not. Resection of the lung could still be debatable with such a scenario, due to the relapsing nature of the disease. However, it should be kept in mind that embolectomy of the hydatid cyst from the main pulmonary truncus would cause higher morbidity and mortality. The surgeon performing this type of operation via thoracotomy should remember that in pulmonary artery involvement, feeding vessels are from the systemic circulation, since the pulmonary arterial flow is occluded. The dense adhesions and the vascular supply from the chest wall could cause trouble such as injuries to the pulmonary parenchyma and bleeding from the chest wall, as we experienced in our case.

In conclusion, in this paper we tried to comment on surgical approaches in intrapulmonary arterial hydatid disease and the role of lesser parenchymal resection. We believe that lesser parenchymal resections in patients with distal intrapulmonary artery localization could be an acceptable approach.

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