Intraarterial pulmonary hydatid cysts revealing ventilation-perfusion mismatch on lung scan

A. Fuat Yapar¹, Mehmet Aydın¹, Dalokay Kılıç², Aysun Sükan¹, Mehmet Reyhan¹

¹Başkent Üniversitesi, Nükleer Tıp Anabilim Dalı, Adana Uygulama ve Araştırma Merkezi, Adana
²Başkent Üniversitesi, Göğüs Cerrahisi Anabilim Dalı, Adana Uygulama ve Araştırma Merkezi, Adana

ABSTRACT
We report a rare case with multiple intra-arterial pulmonary hydatid cysts revealing dramatically large ventilation-perfusion mismatch defects on lung scintigraphy. The causes of ventilation-perfusion mismatches are also reviewed.

Key Words: Ventilation-perfusion mismatch, lung scintigraphy, and hydatid cyst.

Introduction
Hydatid disease (also known as echinococcosis or hydatidosis) is a parasitic disease affecting most commonly liver and lungs. The location of echinococcal cysts inside pulmonary artery is extremely rare (1-6).

Although ventilation-perfusion mismatches are typically attributed to pulmonary thromboembolism (PE), numerous diseases can cause this finding on lung scintigraphy. Herein, a case with multiple intra-arterial pulmonary hydatid cysts is reported in which dramatically large ventilation-perfusion mismatch defects were seen on lung scintigraphy. The causes of ventilation-perfusion mismatches are reviewed.

Case report
A 44 year-old man with multiple hydatid cysts in bilateral pulmonary artery was referred for lung scintigraphy for the quantitative analysis. His past medical history revealed surgical excision of a hepatic hydatid cyst 10 years ago, and he was asymptomatic for a long time. Through the last two months, he has been suffering from mild dyspnea, and cough. Radiographs of the chest showed multiple nodules in the left and right lung. Ultrasound revealed multiple hepatic cysts, and echocardiography depicted a cyst 3.5 × 5 cm in size in the posterior aspect of the left ventricle.

Lung scan was used for evaluation of lung ventilation-perfusion visually and quantitatively. Perfusion scintigraphy was carried out by a slow intravenous injection of 111 MBq 99mTc macroaggregated albumin (MAA) in the supine position. Ventilation scintigraphy was performed by the inhalation of aerosolized 1 GBq 99mTc DTPA on the following day. The aerosol was inhaled in the sitting position and the breathing pattern of the patient was tidal breathing during inhalation of the aerosol. Anterior, posterior, left and right anterior and posterior oblique, left and right lateral views were obtained with a dual head gamma camera (E-cam, Siemens, Illinois, USA) equipped with a low-energy high-resolution parallel hole collimators. Perfusion scintigraphy revealed absence of left pulmonary perfusion, and large perfusion defects on the mid and lower lobes of the right lung (Fig. 1). Only the upper lobe of the right lung had normal perfusion. On the ventilation study, both lungs were visualized. Nearly all of these perfusion defects were normally ventilated except a relative hypoventilation on the apical region of the left lung. So, dramatically large ventilation-perfusion mismatch defects on the entire left lung and two lobes (mid and lower) of the right lung were detected. In quantitative analysis, the distribution of ventilation was 40% to the left lung and 60% to the right lung. Measurement of forced expiratory volume in 1 s (FEV1) was 2.81 L/s in June 2005
After possible middle and lower lobectomy of the right lung, the postoperative predicted lung ratio and the FEV1 value were 70% and 1.97 L/s, respectively (critical FEV1 value > 0.8 - 1 L/s).

In this case, chest X-ray and computed tomography (CT) showed multiple cysts in the lungs. In order to assess further arterial embolism of cysts, MR-angiography was performed with contrast enhanced three-dimensional technique. A dynamic pulmonary magnetic resonance angiography (MRA) image showed absence of pulmonary arterial branches on both lungs except for the right upper lobe artery (Fig. 2A). Multiple cystic structures with septations within the left main pulmonary artery and descending branch of the right pulmonary artery were seen (Fig. 2B).

As a surgical approach; hydatid cyst and membranes were extracted from the right and left pulmonary arteries, and right middle and lower lobectomy was performed via the median sternotomy using cardiopulmonary bypass technique. Unfortunately, the patient died as a result of pulmonary hypertension and right ventricle insufficiency 10 hours later.

Discussion

In this case, the lung scintigraphy showed ventilation-perfusion mismatch defects due to intra-arterial cysts without bronchial obstruction. Hydatid disease affects most commonly the liver and the lung parenchyma. The location of echinococcal cysts inside pulmonary artery is extremely rare (1-6). 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. In this case, 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.

Echocardiography, spiral CT scan, MRI, CT-angiography, MR-angiography, and conventional pulmonary angiography have been used in diagnostic investigation of patients with suspected hydatid pulmonary embolism. 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. 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. MR-angiography, CT-angiography and conventional angiography depict hydatid pulmonary embolism. Contrast-enhanced pulmonary MR angiography has been proposed as a noninvasive and nonionizing alternative to contrast enhanced CT for the evaluation of various pulmonary vascular dis-
Intraarterial pulmonary hydatid cysts revealing ventilation-perfusion mismatch on lung scan

Figure 2. (A) Contrast-enhanced 3D breath-hold pulmonary magnetic resonance angiography (MRA) images showed absence of pulmonary arterial branches on both lungs except the right upper lobe artery. (B) T2-W MR images at coronal plane demonstrated multiple cystic structures with septations within the left main pulmonary artery and descending branch of the right pulmonary artery.

eases (7, 8). Some investigators have found that in animal experimental studies, the diagnostic capability of contrast enhanced MR angiography for small pulmonary embolisms was equal to or better than contrast-enhanced CT (9).

The treatment of choice is surgery. However, surgical intervention can be complicated by rupture of the artery and/or the cyst. Dissemination of the disease, anaphylactic shock, embolism and pseudoaneurysm formation can then happen.

The preoperative prediction of postsurgical loss in lung function can be made accurately by using the quantitative ventilation-perfusion lung scan technique. Either a regional ventilation study or perfusion study may be used for the prediction, but analysis of the regional ventilation distribution appears to be a better parameter than that of perfusion distribution for the prediction of postoperative loss of FEV1 (10). In this case, ventilation scan was used for quantitation of function, instead of perfusion scan because of large perfusion defects.

Ventilation-perfusion scintigraphy has been accepted as a very useful screening modality for pulmonary thromboembolism. Two or more large mismatched segmental defects or the equivalent in moderate sized defects indicate a high probability of PE (≥80%) according to revised PIOPED criteria (11). While most of them quite uncommon, ventilation-perfusion mismatches may occur secondary to vast number of diseases. The pathological process of these lesions involves the pulmonary vasculature occurring either in the lumen, the vessel wall or the perivascular tissue; and ultimately causing vascular occlusive findings. Causes of ventilation-perfusion mismatches on lung scan include various forms of arterial occlusion (thrombus due to acute or previous pulmonary thromboembolism, tumor, fat, parasite-dirofilaria immitis, air, talc, cotton fiber due to intravenous drug abuse, sickle cell disease, septic emboli, pulmonary leukostasis), primary vascular diseases (vasculitis, connective tissue disorder, stenosis, tuberculosis, previous radiation therapy), compression of pulmonary arteries without involvement of bronchi (lung cancer, nodal enlargement-sarcoidosis/tuberculosis/metastases, emphysema, fibrosing mediastinitis-histoplasmosis, lymphangitic carcinomatosis, aortic aneurysm, traumatic pulmonary artery pseudoaneurysm, interruption of inferior vena cava, bronchopleural fistula, idiopathic pulmonary fibrosis, bronchogenic cyst, hemangioendotheliomatosis, pulmonary artery sarcoma), vascular anomalies (pulmonary arterial agenesis or stenosis, peripheral coarctation, pulmonary arteriovenous malformation, systemic arterial supply), immunologic pulmonary edema, pneumonia, pulmonary hypertension, chronic intermittent atelectasis, pulmonary contusion, and artifacts (hiatal hernia/intrathoracic stomach, swallowed aerosol and paralyzed diaphragm, wedging of Swan-Ganz catheter) (12-20).

The importance of the present case lies in the demonstration of the scintigraphic findings revealing multiple ventilation-perfusion mismatches due to the intra-pulmonary arterial hydatid cysts.
References

Corresponding Author
A. Fuat Yapar
Başkent Üniversitesi, Adana Uygulama ve Araştırma Merkezi, Nükleer Tıp Anabilim Dalı, 01250, Yüreğir, Adana
Tel: 3 322 3272727-1020
Fax: 3 322 3271270
E-mail: fyapar@dr.com