

A Multistage Hydatid Cyst Case With Multicentric Localization and Transdiaphragmatic Invasion

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Abstract

A 29-year-old female patient presented with intermittent left shoulder pain that had been present for nearly two years. The patient had been operated for a hydatid cyst six years ago. PA lung X-rays yielded homogeneous radio opacity in the left middle and lower lobes. With the help of further radiological investigations, the patient was diagnosed as a case of multistage hydatid cyst with multiple localization and transdiaphragmatic invasion. (A surgical intervention was performed by the staff of the Department of Thorax and Cardiovascular Surgery). The cysts were excised with

cystotomy and capitonnage. The diaphragm was also found to be attacked by the cysts and was primarily repaired by capitonnating the cyst walls and the cysts above and under the diaphragm were excised. Albendazole was started in a dose of 10 mg/kg/day. The patient had an uneventful recovery.

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Introduction

Hydatid cyst is primarily a zoonosis caused by *Echinococcus granulosus*. However, transmission to humans is very common and this creates an important public health problem (1). These parasites frequently invade the liver, lungs being the second most common location. The invasion of the organs and the tissues by these parasites leads to a clinical picture known as unilocular hydatid cyst (2). The disease is endemic in Turkey with a reported prevalence of 0.87-20 per 100 000 (3).

Case Report

A 29-year-old female patient presented to our department in February 2000 with a complaint of intermittent left shoulder pain that had persisted for the past two years. In 1994, the patient had undergone an operation for hydatid cyst. She had not received any medical treatment before or after the surgery. On inspection, an incision scar was noted on the left hemithorax. On percussion, there was dullness on the lower parts of the left lung. Respiratory sounds were could not be heard in the left lower zones at auscultation. Examination of other systems revealed no noteworthy findings.

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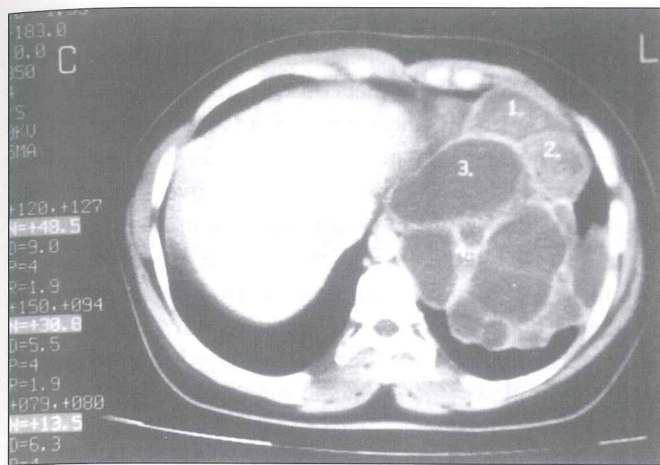


Figure 1. On thorax CT, multilobulated cystic lesions of different sizes in the posterior abdomen

PA chest radiography showed, in the left hemithorax, a homogeneous radioopacity with lobulated and well-defined borders that totally covered the middle and lower zones. The left costophrenic angle was totally obliterated; the limit between the borders of the heart and the mass could not be easily identified.

In computerized tomography (CT) of the thorax, multilobulated cystic lesions of different sizes with multiple septa that started from the left lung lower segment were observed. These lesions were mostly intraparenchymal (Figure 1). As the lesions progressed through the basal part, they touched the pleura and the interphase between the diaphragm and the lesion could not be easily traced. In the posterior parts of the abdomen the lesions extended extra peritoneally as far down as the level of the L-2 vertebrae.

A tentative diagnosis of hydatid cyst with transdiaphragmatic invasion was made and thoracoabdominal multiplanar magnetic resonance imaging (MRI) was planned in order to demonstrate this invasion.

In the multiplanar MRI sections, the lesions were observed from 1 cm below the tracheal bifurcation, without leading to a significant compression atelectasia at the level of their intraparenchymal localizations. They were adherent to the pericardium at the cardiac apex. At the basal part of the lung they stuck to the pleura with a large base; their largest diameter was 30mm. Multiple septa were also observed within the lesions. In T1 and T2 weighted series, the mass lesions had a fluid intensity with accompanying septal formations. These lesions were evaluated as stage I-II hydatid cysts. The coronal sections demonstrated that the cysts, by transdiaphragmatic invasion, descended as far down as the lower pole of the left kidney in the extraperitoneal cavity (Figure 2). The spleen was displaced medially due to significant compression by the cysts. The left kidney was mildly compressed.

Abdominal ultrasonography (US) revealed a normal liver, spleen and gall bladder; the left kidney was enlarged (to a diameter of 130 mm) and displaced anteriorly and caudally.

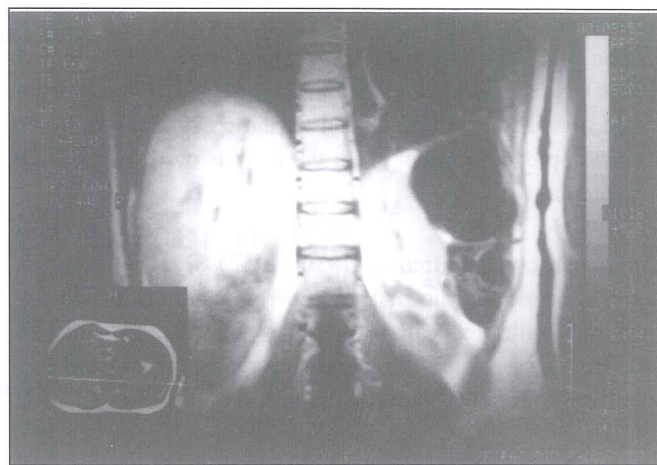


Figure 2. On coronal MR sections, cysts which have descended as far as the lower pole of the left kidney by transdiaphragmatic invasion.

In the posterior part of the spleen, the costodiaphragmatic sinus was obliterated and the retroperitoneally located lesions could easily be differentiated from the spleen parenchyma; the dimensions of the largest lesion were 26 x19 mm. The lesions contained multiple daughter vesicles. These lesions were evaluated as stage I-II hydatid cysts according to Garby classification.

Echocardiography was performed and showed that the heart was deviated to the right due to compression by the cysts. However, cardiac chambers were not collapsed; the diameters of the chambers, wall movements and thickness of the walls were evaluated as normal.

The Department of Thorax and Cardiovascular Surgery was consulted and a surgical intervention was planned for this patient with unilocular hydatid cyst. The intervention revealed the presence of approximately 40 cysts with diameters of more than 1.5 cm in the left hemithorax. The cysts showed adherence to the apex, to the pericardium and pleura and also to each other. There were also cysts of diameters of less than 1.5 cm in multiple numbers. The cysts localized in the parenchyma were excised by cystotomy and capitonnage and other localized cysts were excised by cystotomy and curettage. The diaphragm was primarily repaired by capitonnage of the cyst walls; the cysts above and under the diaphragm were then excised.

The patient did not have any postoperative complications. Albendazole was started in a daily dose of 10 mg/kg and the patient was asked to return for follow-up.

Discussion

Unilocular hydatid cyst can develop either primarily or secondarily after an invasion. The cysts that are formed by the inoculation of the oncosphere are generally single. With the spontaneous or iatrogenic rupture of these cysts, the daugh-

ter vesicles that are disseminated to the exterior are localized in the neighboring organs and tissues with their scolexes. These cysts are generally multiple in numbers. In our patient, based on a history of a previous hydatid cyst operation and the widespread nature of the invasion, the cystic disease was accepted as secondary.

Unilocular cysts are frequently single (75%). However, they can also have a multifocal or multiorgan localization. The frequency of multiple localization ranges between 11% and 30% (4). In our case, multiple cysts were present. Lung involvement is solitary in 70% of the cases, and multiple in 30% (5).

Unilocular cysts can remain asymptomatic for 5-20 years. The large masses in the lungs can displace the diaphragm and cause a mediastinal shift (6). Our case had a complaint of left shoulder pain due to diaphragmatic involvement, which had started 4 years after her operation.

The cysts are located in the pericardium and the heart in 0.5-2% of the patients with hydatid cyst. This involvement is generally seen in patients with multiple hydatid cyst. The best diagnostic tool is MRI for such cases. Echocardiography also aids in the diagnosis. In our case, cardiac involvement could not be demonstrated with MR and echocardiography. The presence of a cyst that was adherent to the pericardium was shown with MR with coronal plane. This finding was verified by surgery.

The best diagnostic tools are the radiological procedures. The structure of the hydatid cyst and that of the cyst wall, wall calcifications, daughter vesicles, septations and the presence of fluid within the cyst can be evaluated with imaging techniques. It is recommended that abdominal US be used routinely in patients with lung hydatid cysts because liver cysts can frequently accompany the lung lesions in these patients (1). The presence of diaphragmatic involvement is reported to be very rare in patients with hydatid cyst (7). In our case, the cysts attacked the diaphragm and this was verified during the operation. We evaluated this finding as a transdiaphragmatic invasion either by venous or lymphatic route.

Until the 1970's, the treatment of hydatid cysts could only be achieved with surgery (8). Studies on the medical treatment

of this parasitic disease started in more recent years. Today, it is believed that if the operation is performed after a brief course of albendazole or mebendazole, the success rates will be higher (9). The most important complication of the surgery is the development of a widespread secondary hydatid cyst disease due to the dissemination of live scolexes from the surgery site. Reported recurrence rates in the lungs range from 2 % to 14% (10). Recurrence rates are decreased by administering medical treatment in the preoperative and postoperative periods (6). We evaluated our case also as a recurrence. The patient had not received any treatment prior to or following her previous surgery.

In summary, a patient with hydatid cysts which were multiple, of different sizes and numbers, and which showed widespread localization, diaphragmatic invasion and involvement, is reported. The case was accepted to be secondary and also to represent a recurrence. We believe that cyst screening especially with CT and US, MRI when necessary should be performed in patients with hydatid cyst, as the chance of developing a secondary hydatidosis is very high, especially following surgery. For the early diagnosis of a recurrence in the postoperative period, close follow-up of the patients is essential.

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