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Both biliopleural fistula and portal hypertension with giant hydatid cyst of the liver

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To the editor,

Hydatid disease still remains an important health problem in endemic regions such as the Mediterranean. The most common complication of liver hydatid cyst is rupture in the biliary tree. Biliopleural fistula and portal hypertension are rare complications due to hydatid cyst. However, these two complications have not previously been reported in the same case. We present a case report in which a giant liver hydatid cyst was complicated with both biliopleural fistula and portal hypertension.

A 34 year-old woman was admitted with gradually increasing dyspnea and abdominal distension over a period of one year. Abdominal examination revealed remarkable abdominal distension and venous collaterals (Fig. 1), hepatomegaly and splenomegaly. On pulmonary auscultation there was no breath sound in the basal and middle zones of the right lung. Chest radiography showed an opacity in the right lobe. Abdominal ultrasonography revealed a 21 × 17 cm mass on the right hepatic lobe, which had cystic and hyperechogenic components involving milimetric calcifications (Gharbi stage 3). Laboratory tests were as follows: white blood cell, 13400/mm3, direct bilirubin 0.51 g/dL. Indirect hemagglutination test for cyst hydatid was 1/1280 titer positive. Upper abdominal magnetic resonance imaging and a cholangiography revealed a mass of $187 \times 156 \times 205$ mm dimensions that filled almost all of the right hepatic lobe, with the cyst compressing the pancreas, spleen, stomach, vena cava inferior and vena portae. In light of these findings, surgery was decided upon. Abdominal exploration showed that the mass was adherent to the right diaphragm and compressing on the vena portae and vena cava inferior. Cystotomy was performed and 1500 cc of liquid and 150-200 daughter vesicles were extracted. Fistula was not present. After surgery, abdominal venous collaterals disappeared. Because of persistent pleural effusion, endoscopic retrograde cholangiopancreatography was performed showing contrast leakage from the left intrahepatic biliary tree to the thorax (Fig. 2). A nasobiliary drainage catheter was placed. Thereafter, pleural effusion gradually decreased. Bile drainage was stopped at the twelfth postoperative day. Control cholangiography revealed no leakage to the thorax. The patient was discharged from the hospital and was well after a three month follow-up period.



Fig. 1. — Abdominal examination showing venous collaterals due to portal hypertension.

Hydatid cyst can rarely fistulize to the pleural cavity and lead to portal hypertension. The placement and size of the cyst are important factors related to those complications. There are several factors that lead to pleural fistula. A cyst in the superior and posterior portions of the liver can grow upwards in the chest, eroding through the diaphragm (1). A giant cyst leads to increased pressure on the diaphragm, causing thinner diafragmatic fibres and rupture in the pleural cavity (2). In our case, most probably, the fistula occured because of a giant cyst and due to its placement in the liver.

On the other hand, the present case also had portal hypertension. In the literature, a few cases of hydatid cyst of extrahepatic organs with portal hypertension have been reported (3,4). Hydatid cysts had originated from

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Fig. 2. — Endoscopic retrograde cholangiopancreatograph showing contrast leakage from the left intrahepatic bile tree into the thorax.

the spleen or pancreas in those cases. Portal hypertension developed due to the cyst pressing on the portal and splenic veins. In our case, a giant liver hydatid cyst led to portal hypertension by compressing the vena portae and vena cava inferior.

In conclusion, although hydatidosis is a benign disease, it may lead to serious complications. To our knowledge, this is the first report in which a giant liver hydatid cyst was complicated with both biliopleural fistula and portal hypertension. Surgery may not be sufficient when a liver hydatid cyst is large and has more than one complication. Hydatid cyst should be remembered among the causes of portal hypertension and pleural effusion.

References

- MALIK A.A., BARI S.U.L., AMIN R., JAN M. Surgical management of complicated hydatid cysts of the liver. World J. Gastrointest Surg., 2010, 2: 78.84
- CRAUSAZ P.H. Hydatid cyst of the lung and hydatid disease of the liver with intrathoracic evolution. J. Thorac. Cardiovasc. Surg., 1967, 53: 116-129.
- KANTARÇEKEN B., ÇETINKAYA A., BÜLBÜLOĞLU E., DEMIRPOLAT G. Splenic hydatic cyst as a cause of sinistral portal hypertension and isolated gastric variceal bleeding. *Turk. J. Gastroenterol.*, 2010, 21: 317-320.
- SZANTO P., GOIAN I., AL HAJJAR N., BADEA R., SEICEAN A., MANCIULA D., SERBAN A. Hydatid cyst of the pancreas causing portal hypertension. *Maedica (Buchar)*, 2010, 5: 139-41.