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CASE REPORT

Intrahepatic Biliary Fistulisation of Primary Hydatid Disease of the Spleen Causing Cholestasis

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ABSTRACT

Hydatid disease is a serious health hazard, especially in endemic regions. Primary hydatid disease of the spleen is rare, although the spleen is a relatively common site for secondary involvement. A 67-year-old male with primary disease of the spleen causing obstructive jaundice is reported. He presented with upper abdominal pain of 10 days' duration. Magnetic resonance imaging examination and magnetic resonance cholangiopancreatography revealed splenic hydatid cyst with suggested intrahepatic biliary fistulisation causing dilatation of the segment III bile ducts. Surgical exploration revealed primary hydatid cyst of the spleen fistulising to the third segment of the liver with daughter cysts and germinative membranes causing obstructive dilatation of the segmental bile ducts. To the best of our knowledge, this is the first reported case of primary hydatid disease of the spleen fistulising to the intrahepatic bile ducts causing jaundice.

Key words: *Hydatid disease, Spleen, Biliary dilatation*

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ÖZET

İntrahepatik Safra Yollarına Fistülize Olarak Kolestaza Yol Açan Primer Dalak Kist Hidatiği

Hidatik hastalık, özellikle endemik bölgelerde önemli bir sağlık sorunudur. Dalağın sekonder tutulumu sık olarak görülmesine rağmen, primer dalak hidatiği nadirdir. Bu olguda primer dalak hidatiğine bağlı tıkanma sarılığı görülen 67 yaşındaki bir erkek olgu bildirilmektedir. Hasta 10 gündür devam eden karın ağrısı şikayetiyle başvurdu. Yapılan manyetik rezonanslı kolanjiyopankreatografik inceleme, karaciğere fistülize olarak üçüncü segment safra yollarında genişlemeye yol açan dalak hidatiği ile uyumluydu. Yapılan cerrahi eksplorasyonda primer dalak hidatik kistinin üçüncü segment safra yollarına fistülize olduğu, kız kistler ve germinatif membranlar yoluyla segmental safra yollarında tıkanmaya bağlı dilatasyona yol açtığı saptandı. Bu olgu, intrahepatik safra yollarına fistülize olarak sarılığa yol açtığı bildirilmiş olan ilk primer dalak kist hidatik olgusudur.

Anahtar kelimeler: *Kist hidatik, Dalak, Safra yolu dilatasyonu*

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INTRODUCTION

Hydatid disease is a common cause of hepatobiliary health problems, especially in endemic regions of the world like the Middle East, the Mediterranean, South America, Australia, New Zealand, and East Africa[1]. The disease most commonly manifests itself as a cystic mass lesion localised to the liver, although many different intraabdominal and extraabdominal organs have been reported to be affected by this parasitic infection[2-4]. Even though secondary involvement of the spleen is a relatively common finding, primary hydatid disease of the spleen is rare[5]. Ozdogan et al. reported seven patients with isolated splenic disease operated on over a period of 20 years, with an overall incidence of 1.2% among all hydatidosis cases[6]. Splenic hydatid cysts do not present with jaundice unless the liver is the primary organ of involvement. Primary hydatid cyst of the spleen fistulising into the liver and causing obstructive jaundice is, to the best of our knowledge, reported for the first time with this case.

CASE REPORT

A 67-year-old male was referred with upper abdominal pain and hyperbilirubinemia of five months, duration. His surgical history revealed bilateral inguinal hernia repair eight years before. Ten days before his referral, he presented to another institution with severe abdominal pain. Abdominal ultrasonographic examination revealed a simple splenic cyst measuring 5.5 cm in diameter. Intrahepatic bile ducts were dilated on the left side and the common bile duct measured 9 mm at its distal end. No lesions of the liver parenchyma or pancreas were identified. On admission, he was mildly icteric with total bilirubin level of 9.55 mg/dL (range: 0.3-1.2 mg/dL), direct bilirubin level of 5.63 mg/dL (range: 0-0.2 mg/dL), alkaline phosphatase (ALP) level of 197 U/L (range: 28-112 U/L), gamma glutamyltranspeptidase (GGT) level of 507 U/L (range: 5-46 U/L), aspartate aminotransferase (AST) level of 175 U/L (range: 0-35 U/L), and alanine aminotransferase (ALT) level of 269 U/L (range: 0-45 U/L). His prothrombin time was slightly longer than normal at 15.5s (upper normal limit: 13s). A magnetic resonance cholangiopancreatography (MRCP) was obtained to evaluate the liver and biliary tract and showed a subdiaphragmatic lesion on the left side measuring 6 x 7 cm in diameter consistent with a hydatid cyst (Figure 1). MRCP also suggested intrabiliary fistulisation in the left liver lobe with left-sided dilatation of the intrahepatic biliary tree (Figure 2). A simple cyst of the spleen was also noted.

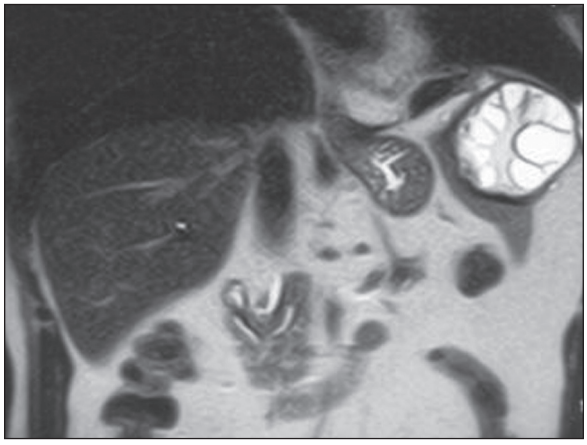

Figure 1. Left-sided infradiaphragmatic cystic lesion on the magnetic resonance imaging consistent with hydatid disease.

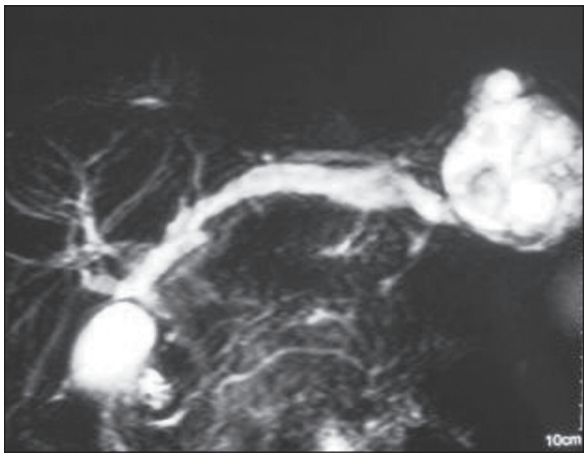

Figure 2. Magnetic resonance cholangiopancreatography image demonstrating intrabiliary fistulisation of the hydatid cyst at the left liver lobe with dilatation of the left intrahepatic biliary tree.

Extrahepatic hydatid cyst with intrabiliary fistulisation was diagnosed preoperatively and surgery was performed. Intraoperatively, a hydatid cyst originating from the upper pole of the spleen with fibrous adhesions to the diaphragm and the third segment of the liver was identified. A tubular fibrous structure of approximately 1 cm in diameter was connected with the liver parenchyma. This structure was divided to reveal that it was in direct communication with the intrahepatic biliary tree. Cholecystectomy was performed and the intrahepatic biliary tree was probed and irrigated via the cystic duct. Multiple daughter cysts and germinative membranes were extracted from the fistula opening in the third segment. Saline irrigation was repeated until clear bile was draining from the

opening in the liver surface. The spleen was removed, the cystic duct was tied, and the fistula opening in the third segment of the liver was closed with figure of eight sutures. The patient had an uneventful postoperative course and was discharged on the third postoperative day on oral albendazole therapy. Histopathological examination confirmed hydatid disease of the spleen.

Three months postoperatively, his follow-up examination was unremarkable and his albendazole treatment was stopped.

DISCUSSION

Hydatid disease can present with various clinical signs and symptoms. Lesions of the liver can present with abdominal pain or intrabiliary rupture resulting in obstructive jaundice or even free intraabdominal rupture causing abdominal hydatidosis. Almost every organ of the human body has been reported to be affected by this disease. Up to 70% of hydatid cysts are located in the liver, with the right lobe being affected in 85% of patients[7]. Primary hydatid disease of the spleen is rare and is an indication for splenectomy.

Our patient presented with jaundice, which is not expected with isolated extrahepatic involvement. Preoperative imaging revealed the cystic lesion to be in communication with the intrahepatic biliary tree, which was unexpected. In fact, to the best of our knowledge, there has been no other reported case of a splenic hydatid cyst with intrahepatic biliary fistulisation.

It may be assumed that the chronic irritation caused by the growing cyst eroded the liver capsule and eventually resulted in fistulisation of the cyst contents into intrahepatic bile ducts.

Preoperatively, we discussed the option of obtaining an endoscopic retrograde cholangiopancreatography (ERCP), which would have provided the opportunity to explore the biliary tract non-operatively. However, this was not performed since the spleen was the source of the disease, and a surgical exploration of the abdomen with splenectomy was planned. A surgical exploration of the common bile duct was also planned but later deemedun unnecessary, since after adequate irrigation through the cystic duct, we were convinced that the intrahepatic biliary system was clear of any residual disease.

ERCP can provide valuable information preoperatively in cases where biliary fistulisation is suspected. Endoscopic sphincterotomy can facilitate drainage of the biliary system. Our patient underwent MRCP, which clearly indicated biliary involvement; therefore, we did not feel like the added risk of ERCP was justified.The value of preoperative diagnostic studies such as MRCP can not be overemphasized in this case. In our case, intrahepatic biliary dilatation was evident, clearly suggesting communication of the splenic cyst with the biliary tree.

Hydatid disease can manifest itself in many different forms, especially in endemic regions. Although we believe that intrahepatic fistulisation of a splenic hydatid cyst is reported for the first time with this case, various forms of intra and extraabdominal fistulisations such as spleno-colic, hepato-colic, adrenojejunal, gastric and cutaneous have been reported[8-12]. This entity has to be kept in mind when encountering a splenic cyst. Every hepatobiliary surgeon practising in endemic regions should be familiar with its manifestations and management.

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