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Case Report

Endoscopic Treatment of a Bronchobiliary Fistula Due to Complicated Hydatid Cyst After Surgical Intervention: A Case Report

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ABSTRACT

Hydatid cyst is an infectious parasitic disease often caused by Echinococcus granulosus, or to a lesser extent by Echinococcus alveolaris. The liver is the organ most frequently involved.

We report a case with hydatid cyst of the liver and lung that many years after surgery developed bronchobiliary fistula and biloptysis. The patient had been operated 2 times previously but yet the problem was present. Endoscopic retrograde cholangiopancreatography (ERCP) was used successfully for treatment. We present the result of ERCP and endoscopic sphincterotomy in the management of biliary hydatid disease.

Bronchopleural fistula after hydatid cyst surgery could be cured by ERCP and sphincterotomy with or without stent insertion.

Keywords: Hydatid Cyst, ERCP, Fistula

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INTRODUCTION

We report a case with hydatid cyst of the liver and lung that many years after surgery presented with bronchobiliary fistula and bilioptysis. Although the episodes of bilioptysis were severe and frequent, the patient underwent ERCP with sphincterotomy with successful result.

Hydatid cyst is an infectious parasitic disease often caused by Echinococcus granulosus or to a lesser extent by Echinococcus alveolaris. The liver is the most frequently involved organ (70 %). An enlarged cyst leads to distension of neighboring bile duct secondary to compression of the duct; and erosions occur in the extremely distended bile duct, resulting in a fistula. The incidence of postoperative biliary fistula ranges from 4% to 28% .(1) Bronchobiliary fistula which often presents with bilioptysis is a rare but serious complication after hepatic resection.

Biliary fistula develops in 4%-28% of patients after hepatic hydatid disease (HHD) surgery.

Although endoscopic retrograde cholangiopancreatography (ERCP) and endoscopic sphincterotomy (ES) are helpful in the treatment of this complication, persistent fistulas may occur. (2,3), In managing bronchobiliary fistula, it has been reported that surgical approaches should be considered only after failure of aggressive attempts through nonsurgical interventional techniques such as endoscopic retrograde cholangiography

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or percutaneous transhepatic cholangiography. This is because reoperative procedures tend to be complicated with significant morbidity and mortality. (4), Before the advent of endoscopy and imaging techniques, the only modality for diagnosis and treatment was surgery. In more recent years, ERCP has been extensively and successfully used in both diagnosis and treatment of biliary complications of hepatic hydatid cyst. We present the results of ERCP and endoscopic sphincterotomy in a patient with biliary hydatid disease.

CASE REPORT

A 20-year-old man was admitted in our center with anorexia, abdominal pain, dyspnea and bilioptysis. The patient had a history of surgery 14 years ago for hydatid cysts of the liver, left and right lungs. He had another recent operation, 2 months before, due to bilioptysis and recurrence of hydatid cysts in the liver and lower lobe of the right lung. After surgery, bilioptysis was not improved and there was a persistent biliary leak through the draining catheters of the liver and lung. The patient chest X-ray is shown in Figure 1.

Figure 1: Postero-anterior chest X-ray view reveals a few relatively thick-walled cavitary lesions with fluid levels, which denote concurrent presence of fluid and air, dispersed throughout the right pulmonary field.

There was continued bilioptysis despite surgery and after chest tube insertion at right lung as is shown in Figure 2.

Figure 2: Postero-anterior chest X-ray view demonstrates a few remarkably thick-walled cavitary lesions occupying the right pulmonary field lower moiety containing both fluid and air, resulting in fluid levels. Note the right-sided chest tube placement and resultant right chest wall subcutaneous emphysema.

First, to determine any abnormal communication or fistula between biliary system and pleura, MRCP was requested which confirmed broncho-biliary fistula. In our center, ERCP for the patient was performed as shown in Figure 3.

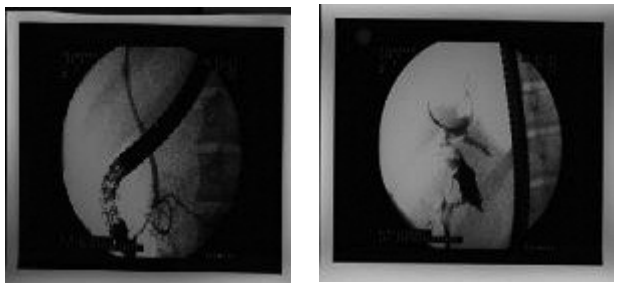

Figure 3: ERCP shows extravasation of contrast to outside of biliary system.

During ERCP, endoscopic sphincterotomy was performed as endoscopic treatment of broncho-biliary fistula. There was no need for stent insertion.Gradually, during two months after endoscopic sphincterotomy, the hepatic and pleural biliary leaks through the draining catheters were ceased and the repeated ERCP showed improvement of broncho-biliary fistula (Figure 4).

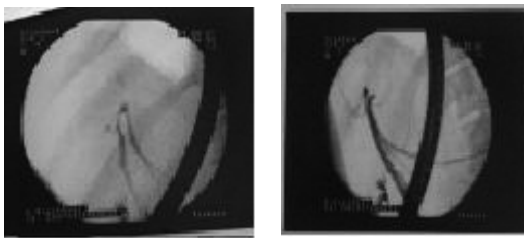

Figure 4: ERCP two months after sphincterotomy shows no biliary leakage as well as normal biliary drainage.

DISCUSSION

An irritating cough and expectoration of yellowish sputum, termed bilioptysis, are the typical presentations of bronchobiliary fistula (BBF). Sputum analysis verifies the levels of direct and indirect bilirubin, which doubtlessly indicates a direct communication between the biliary system and bronchial tree. Mild to moderate jaundice generally occurs in some cases. Generally, the clinical conditions of the patients are not encouraging due to the underlying chronic illness or previous surgical intervention. Patients are persuaded that the fistula should be treated without delay following intense supportive therapy, which includes proper antibiotics and high calorie intake. Radiology reveals patch densities dominantly in the right lower lobe; however, these patches may also be present throughout the right lung and even in the left side. (5,6), Thoracic and upper abdominal CT scans appear to be the best tools for the initial evaluation of lung and liver pathologies to plan further investigations. Biliary fistula is the most frequently seen complication after surgical treatment of hydatid cyst. Increased pressure in the biliary system leads to enlargement of small fissure-like connections, with resultant bile flow into the cavity and drainage of bile from the surgical drains placed in the cyst. The reason for the bile flow into the cavity may be Oddi's stenosis or obstruction due to cystic material. On the basis of the above pathology, the mainstay of fistula treatment is to reduce the pressure in the bile ducts. (7), This can be done through surgery; i.e., biliodigestive anastomosis, or by means of an endoscopic approach in which ES and/or nasobiliary drainage are carried out or a stent is applied. Endoscopic techniques have been used with success in postoperative biliary drainage, including that associated with treatment of hydatid cyst. The most commonly performed technique is ES. There are reports in the literature about the use of ES and its success in treating biliary fistula as a postoperative complication of hydatid cyst. The time from ES to closure of fistula is reported to be 3-21 days. (8), Saritas et al. (9), reported 87 patients over a period of 8 years, by means of ERCP, with postoperative biliary symptoms who had previously undergone surgery for HHD. The overall success rate of endoscopic treatment was 86 %, and a second surgical intervention was required only in six patients. Findings from ERCP included biliary fistula in 55 patients (63.2 %), biliary stricture in 16 (18.4 %), and residual hydatid material within the bile duct in 14 (16.1 %) cases. No serious complication was encountered after endoscopic procedures and concluded that endoscopic treatment modalities are safe and helpful methods for the treatment of biliary complications of hepatic hydatid cyst in the postoperative period. Simsek et al. (10), evaluated the use of diagnostic and therapeutic ERCP in pre- and postoperative patients with HHD. For 8 years, ERCP was performed in 39 patients with hepatic echinococcal disease. Indications in the preoperative group of patients (n = 19) included a cholestatic enzyme profile in all cases; jaundice and acute cholangitis also were present in 14 and 7 cases, respectively. In the postoperative group (n = 20), indications for ERCP included persistent external biliary fistula after surgery in 10 patients, jaundice in 8, acute cholangitis in 7, and right upper quadrant pain in 2 patients. The most common ERCP finding in the postoperative group was external biliary

fistula (10 patients). Other findings consisted of hydatid cyst material within the bile duct, bile duct stenosis, cystobiliary fistula and hydatid cyst material in the bile duct, cystobiliary fistula, hydatid membranes in the gallbladder, extrinsic compression to bile ducts, and a normal cholangiogram. In the postoperative group, sphincterotomy (with balloon or basket extraction as needed) was performed in 19 patients, stents were placed in 2 patients, 1 patient underwent balloon dilatation, and 1 had nasobiliary drainage; there was complete resolution of the findings in 14 of the 20 patients. They concluded that ERCP and related therapeutic maneuvers are safe and valuable in the pre- and postoperative management of patients with HHD. (11), Khandelwal et al. (12), reported a case with bilioptysis; a 73 year-old woman with a hepato-cellular carcinoma who developed a bronchobiliary fistula. Endoscopic biliary sphincterotomy and insertion of a prosthesis led to successful resolution of symptoms and restoration of normal bile flow.

CONCLUSION

Bile behaves as a strong irritant when present outside the biliary channel and gastrointestinal system. Bronchopleural fistula after hydatid cyst surgery could be cured by ERCP and sphincterotomy with or without stent insertion.

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