Case Report
Biliobronchial Fistula after Liver Surgery for Giant Hydatid Cyst

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Background. Biliobronchial fistula (BBF) is a rare complication in the natural history of liver hydatid disease by Echinococcus granulosus. We present a case of BBF after resection of a giant liver hydatid cyst in a 72-year-old woman. Case Report. A total cystpericystectomy was done, leaving the left lateral section of the liver that was fixed to the diaphragm. Postoperatively, the patient developed obstructive jaundice. An ERCP showed an obstruction at the junction of the left biliary duct and the main biliary duct and contrast leak. At reoperation, the main duct was ischemic, likely due to torsion along its longitudinal axis. A hepatotomy was done at the hilar plate, and the biliary duct was dissected and anastomosed to a Roux-en-Y jejunal loop. She was discharged without complications. Five months later, the patient developed cholangitis and was successfully treated with antibiotics. However, she suffered repeated respiratory infections, and four months later she was admitted to the hospital with fever, cough, bilioptysis, and right lower lobe pneumonia. The diagnosis of BBF was confirmed with 99mTc Mebrofenin scintigraphy. At transhepatic cholangiography, bile duct dilation was seen, with a biliothoracic leak. She underwent dilatation of cholangiojejunostomy stricture with placement of an external-internal catheter. The catheter was removed 3.5 months later, and two years later the patient remains in very good condition. Conclusion. An indirect treatment of the BBF by percutaneous transhepatic dilation of the biliary stenosis avoided a more invasive treatment, with satisfactory outcome.

1. Introduction
Bronchobiliary fistula (BBF) is defined as an abnormal communication between the biliary tree and the bronchial tract. It is a rare complication that may appear in the natural history of liver hydatid disease or after its surgical treatment [1, 2], trauma [3], congenital malformation [4–6], tumor [7], surgery [8–10], biliary lithiasis [11], hepatic abscesses [12], liver transplantation [13], or even radiofrequency treatment [14, 15].

The most prominent sign is bilioptysis or the presence of bile in the sputum. Other possible symptoms and signs are cholangitis, jaundice, and cutaneous fistula [12]. The first case of BBF was described by Peacock in 1850 [16] in a patient with a hydatid cyst.

We present a case of BBF that appeared after the surgical treatment of a giant liver hydatid cyst.

2. Case Report
A 72-year-old woman with abdominal complaints for several months was found to have a large right upper quadrant cystic mass (16 × 17 × 18 cm), consistent with a multivesicular liver hydatid cyst that replaced the right lobe of the liver, with compensatory hypertrophy of the left lateral segment (LLS) (Figure 1). After discussing the high risk of postoperative
Complications, the patient agreed to undergo surgery. We performed a total open cyst pericystectomy; after detaching the right lobe ligaments, we opened the cyst, extracted the fluid, and instilled hypertonic saline. The right lobe elements were ligated, and the entire cyst with a margin of pericystic inflammation was resected. The LLS was sutured to the diaphragm.

Postoperatively, the patient developed progressive jaundice. She was afebrile and had otherwise normal liver function. Ultrasound and CT were notable for intrahepatic biliary dilation. ERCP showed an obstruction at the level of the biliary confluence and a contrast leak (Figure 2). The patient was taken back to surgery for exploration where we found a fibrotic and ischemic main bile duct, likely due to torsion along its longitudinal axis. We dissected the bile duct until the liver parenchyma, but there was not viable tissue. A hepatotomy was performed at the hilar plate and a bile duct of about 1 cm in diameter was dissected and opened longitudinally. A Roux-en-Y jejunal loop was prepared. A side-to-side biliary jejunal anastomosis was performed with interrupted 6–0 PDS sutures. The patient recovered without further complications, and she was discharged one week later.

Five months after the initial surgery, the patient was admitted to the hospital with ascending cholangitis. She was successfully treated with antibiotics and discharged.

However, the patient suffered repeated respiratory infections in the outpatient setting. Nine months after the original operation, the patient presented to the hospital with fever and productive cough. She did not have jaundice. On physical examination, the patient seemed uncomfortable and expectorated bile-stained sputum several times. Chest plain film showed right lower lobe opacity consistent with right lower lobe pneumonia. Because of her persistent bilioptysis, she underwent 99mTc Mebrofenin scan (Bridatec, Amersham Health, Gipharma, Saluggia, Italy) which confirmed the diagnosis of biliobronchial fistula (BBF). A fistulous tract connected the liver and the right lower lobe; the patient’s sputum contained enough radioisotopes to be detected by the gamma camera (Figure 3).

Given the postsurgical anatomy, the patient underwent percutaneous transhepatic cholangiography which was notable for bile duct dilatation, stenosis of the biliojejunal anastomosis, and biliothoracic leak (BBF) (Figure 4). The anastomotic stricture was dilated, and an external-internal catheter was left in place. There was immediate cessation of bilioptysis and marked improvement of the patient’s condition.
isotope-containing sputum. The use of IDA scan was sug-

Follow-up studies showed good contrast passage, with-

complication is diagnosed [2].

The reason BBFs develop is not completely known. In some

cases, inflammation by bile from the intra-abdominal com-

partment could permeate the diaphragm [8]. In our case,

anastomotic stricture at the cholangiojejunostomy in-

creased pressure in the biliary system, resulting in a pro-

inflammatory collection of bile that extended through the

diaphragm to communicate with pulmonary tissue.

BBF is not easily diagnosed. Bile in the sputum is pathog-

nomic of BBF, but very often the patients have respiratory and/or biliary symptoms for weeks or months before this

c complication is diagnosed [2].

In our case, BBF was not suspected until the patient

started to have bilioptysis, and the suspicion was confirmed

with a Mebrofenin scan, showing the fistulous tract and an

fistula itself [8]. A biloma can be found, as a subphrenic col-

lection, sometimes associated to a pleural effusion. In other

instances, biliary dilatation can be seen, as in our case.

CT is widely used, but it rarely serves to visualize the

fistula itself [8]. A biloma can be found, as a subphrenic col-

lection, sometimes associated to a pleural effusion. In other

instances, biliary dilatation can be seen, as in our case.

MRCP has been useful in some reports [8, 10]. It has lim-

itations in the visualization of nondistended ducts and

when there is pneumobilia, as is expected after cholangio-

jejunostomy.

Treatment of BBF has traditionally been surgical repair

[12, 18]. In 1990, Brem et al. reported the first case of suc-

cessful endoscopic treatment of BBF, in a patient with biliary

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