Endoscopic Treatment of Bronchobiliary Fistula

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Summary: *We tried to treat 9 patients with bronchobiliary fistulas (BBFs) endoscopically for the last 6 years. In most of the patients the reason was Hydatid cyst operations. Six patients had biliocutaneous fistula (BCF) at the same time. All BBFs and BCFs were closed after the NBD placement without any complication. Three patients who developed fistula recurrence healed with endoscopical treatment. From those observations we can conclude that endoscopical treatment is a good alternation to surgical treatment.*

Key Words: Bronchobiliary fistula, Nasobiliary drain.

Bronchobiliary fistula (BBF) is a rare condition. The most common causes are echinococal and amoebic disease of the liver. BBFs can occur spontaneously or following diaphargm injury which make take place during abdominal or thoracoabdominal surgery. Bile-stained sputum is pathognomonic in diagnosis. Surgical procedures in the treatment carry high risk and re-operation may be needed in some cases (10,11). There are some reports about the treatment of Biliary-cutaneous fistulae (BCF) by an endoscopically placed Nasobiliary drain (NBD). We treated successfully 9 patients with BBFs, 6 of which also had BCFs, in this way. All fistulas healed within 6 weeks. No complication occurred. The results of this simple therapy seem more satisfactory compared to highed-risked surgery.

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PATIENTS

In the last 6 years we have attempted to treat endoscopically 9 patients with BBF which developed post operatively (8 pts) and spontaneously (1 pt). The characteristics of the patients are shown in the table I. The causes of the operations were only Hydatid Disease of Liver (HDL) in 3 patients (case 5,6,9), HDL with perforation into the right pleural space in 2 patients (case 1,2), HDL and pulmonary in one patient (case 4). One patient (case 7) had undergone thoracotomy 20 years ago because of Hydatid cyst of liver and lung and, 4 years and 2 years ago she had undergone laparotomy because of the recurrence of HDL and cholelithiasis respectively. Following cholecystectomy bilio-cutaneous fistula developed and persisted for 1.5 years. Six months ago before her referral to our hospital laparotomy was performed for the treatment of BCF during which the fistula was ligated. Following this operation BBF developed and persisted for 6 months until her admission. In case 3 BBF developed spontaneously because of the HDL with perforation into the right pleural space. Following the operation (thoracotomy+cyst evacuation+lower and middle right pulmonary lobectomy) the degree of bilioptysis increased.

In summary either Hydatid disease or the operation because of Hydatid disease was the cause of BBFs in 7 patients. In one patient (case 4) BBF was secondary to biliary operation. The 7th case had both Hydatid cyst and cholelithiasis and so she had undergone cyst

Table I: Characteristics of the patients with BBF

Case	Sex	Age	Cause of operation	Kind of operation	Duration of bilioptysis	Associated lesions
1	F	19	HDL with perforation into the right pleural space	Laparotomy + cyst evacuation +chest drainage + repair of diaphragm injury	4 week	BCF
2	M	30	HDL with perforation into the right pleural space	Laparotomy + cyst evacuation + chest drainage	2 years	BCF+SSC
3	M	12	Spontaneous BBF (Cause: HDL with perforation into the right pleural space)	Thoracotomy + cyst evacuation + lower and middle pulmonary lobectomy + repair of diaphragm injury	13.5 months	BCF
4	M	35	Cholelithiasis, CBD stones	Cholecystectomy + CBD exploration	6 months	Stones in the CBD and IHBD
5	F	31	HDL	Laparotomy, cyst evacuation + primary suture	2 months	Debris in CBD
6	M	32	HDL	Cyst evacuation	3 years	BCF + SSC
7	F	55	For the treatment of BCF	Ligation of Fistula	6 months	BCF + stones in the CBD
8	M	34	HDL+pulmonary hydatid cyst	Thoracotomy + laparotomy + cyst evacuation + repair of diaphragm injury	10 days	No lesion
9	M	26	HDL	Cyst evacuation	7 months	BCF + stones in the CBD

BBF= Bronchobiliary fistula, BCF= Bilio-cutaneous fistula, HDL= Hydatid disease of liver, CBD= Common bile duct, IHBD= Intrahepatic bile duct, SSC= Secondary sclerosing cholangitis

evacuation (20 and 4 years ago) and cholecystectomy (2 years ago). In this case it was difficult to identify the real causes, but probably both might have an affect in the development of BBF.

The duration of the BBF ranged 10 days to 3 years. Six of the 9 patients had BCF simultaneously. Two patients (case 2,6) had secondary sclerosing cholangitis due to injurious affect of the HDL at the extrahepatic and intrahepatic bile ducts.

METHODS

We performed ERCP with Olympus GIF TJF IT10 model flexible duodenoscope and tried to show fistula tract and the other possible lesions. EST was made with standard sphincterotome (Olympus, Wilson Cook, Microvasive, Hoobs Medical) when needed. Any common bile duct (CBD) stone or debris was cleared from the CBD, following EST. Then we placed 7F naso-biliary drain (NBD) (Hoobs, Wilson Cook) into the CBD. A repeat cholangiogram was performed 4 weeks after the NBD placement. If the fistula tract could not be visualized in an asymptomatic patient (without bilioptysis and bile flow trough BCF tracts) NBD was removed.

We present here two (case 1 and 3) of 9 patients.

Case 1

A 19-yr-old female had been operated for hydatid disease of the liver in which hydatid cysts had perforated into the right pleural space. During the operation cyst evacuation

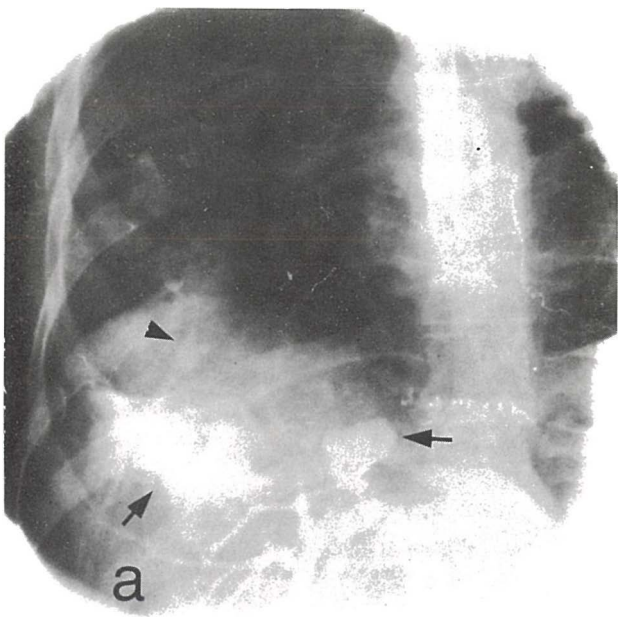

Figure 1: (case 1)

a: ERCP showing dilatation of intrahepatic bile duct and many pouches communicating ith the bile ducts (arrows). BBF tract is seen arising from the big pouch (arrowhead). b: Control cholangiogram four weeks later showing disappearence of the pouches and the fistula tract.

had been performed and a thorax drain had been placed. After the operation bile in sputum had developed and bile flow through thorax drain had not ceased. Four weeks later, he was referred to our unit. On admission, he had bile drainage of 500cc/day from the site of the thorax drain and additionally had bile in sputum. ERCP revealed dilatation of bile ducts and many intrahepatic pouches that communicated with these (Fig 1a); also a BBf tract was seen. A NBD was inserted into the common bile duct. Bile drainage through the NBD was 800 cc/day. Bile flow from the site of thorax drain and bile in sputum decreased gradually and disappeared completely within 3 days and 3 weeks respectively, after NBD placement. On catheter cholangiogram performed in the fourth week the fistulas and pouches were no longer seen (Fig 1b) and the NBD was removed. The patient was asymptomatic during the follow-up period of six years.

Case 3

In a 12-yr-old male patient who had been suffering from bile in sputum for a year, hydatid disease of the liver had been diagnosed. The hydatid cysts had perforated into the right pleural space. He had undergone cyst evacuation together with lower and middle pulmonary lobectomy during which a thorax drain was placed. After the operation bile in sputum increased and bile flow through the thorax drain did not cease. Therefore he was referred to our unit 1.5 months after the operation. On admission he was seen to have bile in sputum, and additionally a bile flow of 500 cc/day through the thorax drain. ERCP revealed contrast material extravasation into the right pleural space (Fig 2a). A NBD was inserted into the CBD and daily bile flow was observed to be 700 cc through this drain. Bile flow through thorax drain decreased gradually and ceased completely within 2 weeks following the NBD placement. In the third week the thorax drain was removed. Within the fourth week bile in sputum disappeared. On catheter cholangiogram performed in the fifth week, contrast material extravasation could no longer be seen (Fig 2b). After this procedure the NBD was removed. The patient was asymptomatic during the follow-up period of 5 years.

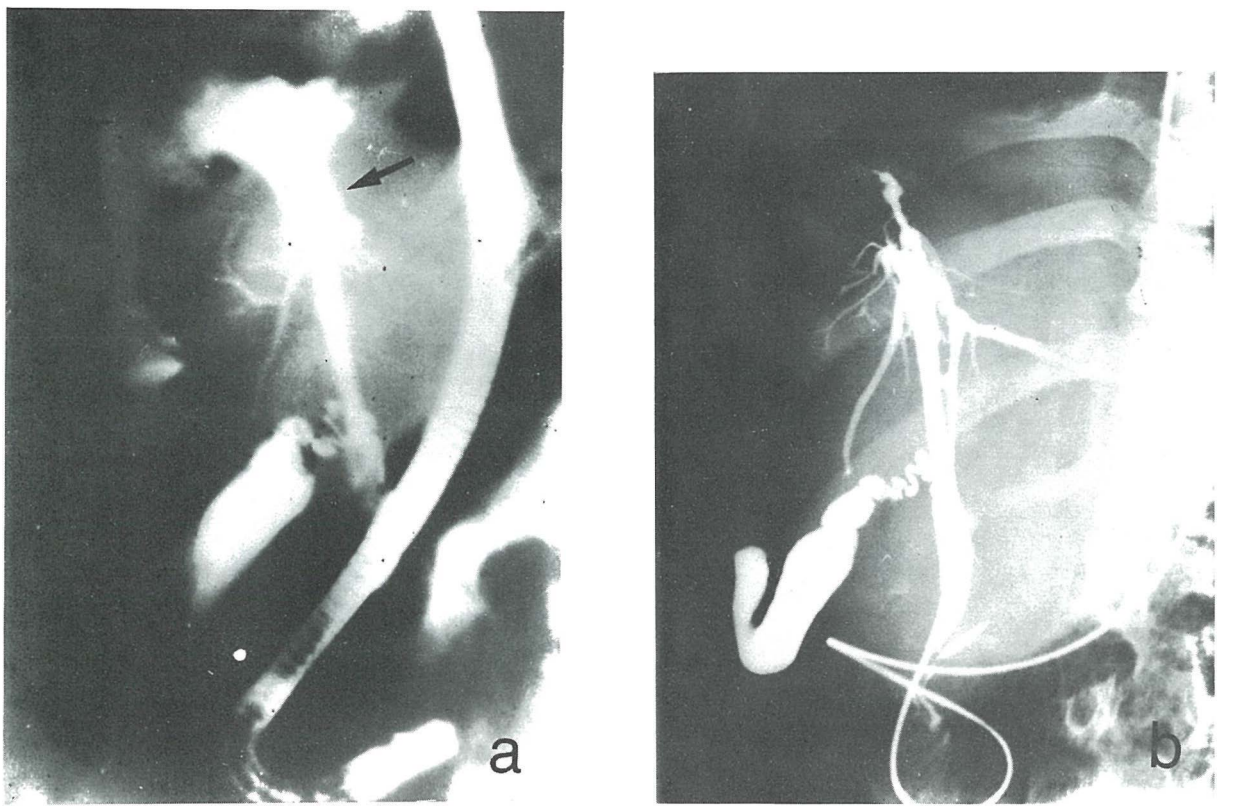

Figure 2: (case 3)

a: ERCP showing biliopleural fistula tract (arrow) and contrast material extravasation into the right pleural space (arrowhead). Contrast material is flowing out through the thorax drain. **b.** Four weeks after NBD placement; neither the fistula tract nor the extravasation are seen.

RESULTS

The BBFs healed in all patients within 6 weeks following endoscopical placement of NBD into the CBD. Bile drainage from the cutaneous fistula tracts disappeared completely. The results of the treatment are shown in the Table II. In 4 patients (1,2,3,8) only NBDs were used, because these patients had no lesion to cause extrahepatic biliary obstruction. Though there was sclerosing cholangitis in case 2, it was clinically not relevant. In the patients with stones (case 4,7,9) and with debris (case 5) in common bile duct, we firstly performed EST to clear the CBD and then placed a NBD. In one patient (case 6) with sclerosing cholangitis we performed EST to facilitate the NBD placement.

The main symptom "bilioptysis" decreased gradually and disappeared completely within 3 weeks in most patients. We usually removed NBD one week after bilioptysis disappeared and following control cholangiogram through NBD. In three patients (case 2,4,9) recurrence had occurred. In case 2 bilioptysis recurred 6 months after the first treatment. The patient also had secondary sclerosing cholangitis which was the cause of the recurrence. In this patient we placed endoscopic endoprothesis into the CBD after 4 week-treatment with NBD. The patient is now symptom free for a follow-up period of 3 years. In case 4 BBF recurred 1 year after the first treatment; on ERCP he had multiple stones in the CBD which were removed endoscopically and then NBD was placed. After 4 weeks of the treat-

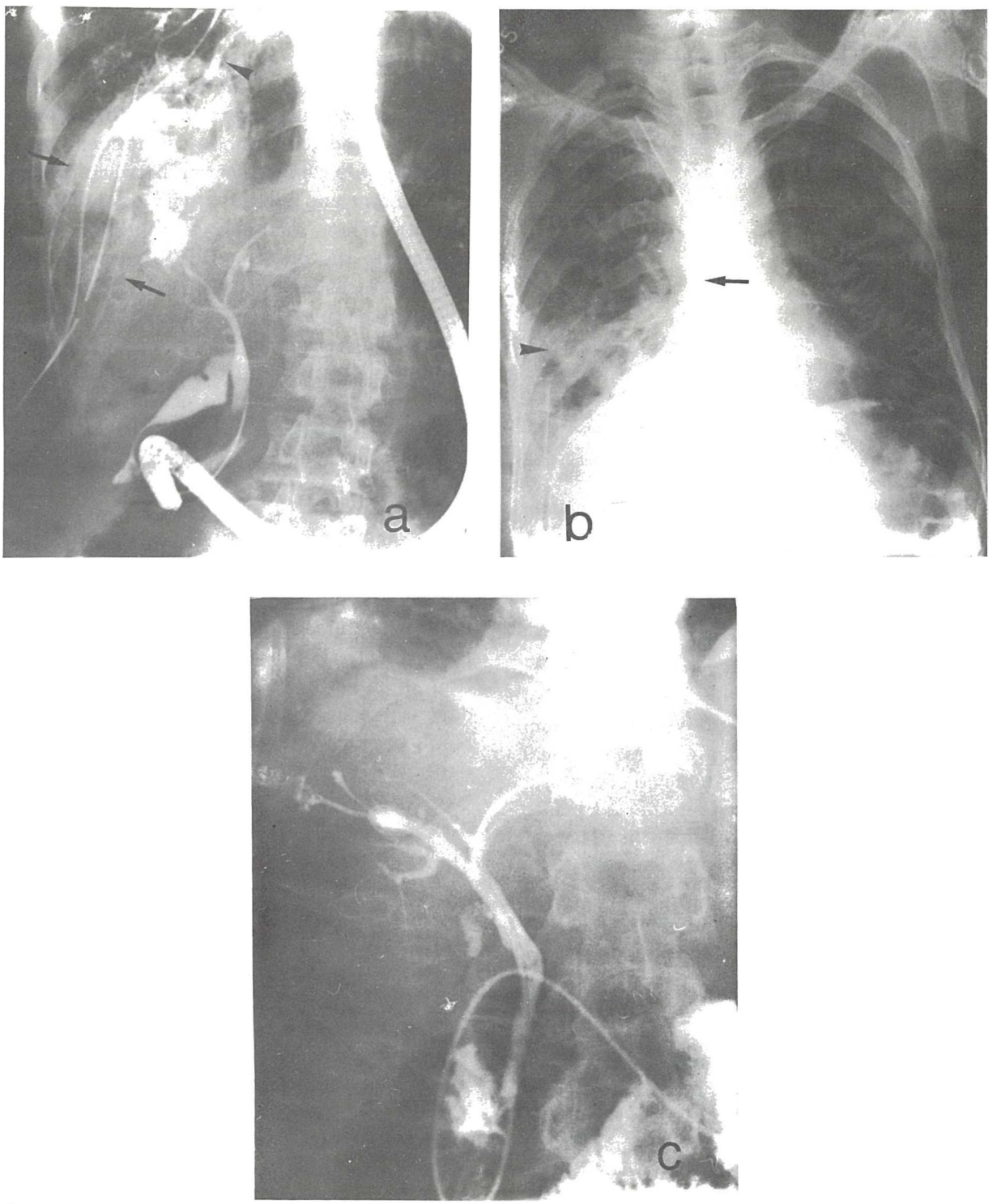

Figure 3: (case 8)

In this case thoracotomy and laparotomy were performed because of HDL and pulmonary hydatid cyst. **a:** The drains (arrows) are seen which were placed during the operation. On ERCP contrast material flowed into the bronchial tree (arrowhead). **b:** In the chest film chest drain (arrow) and the infiltration in the right lung secondary to bile extravasation (arrowhead) are seen. **c:** On the control cholangiogram following 4 week-treatment with NBD, bile extravasation is no longer seen.

Table II: The results of the endoscopic treatment of the patients with BBF

Case	Treatment modality	Duration of the treatment (week)	Result	Follow up
1	NBD	4	Healed	Asymptomatic since years
2	NBD	4		Recurrence after months
3	NBD	5		Asymptomatic since years
4	EST + stone extr + NBD	4		Recurrence after year
5	EST + debris clearance + NBD	6		No recurrence since years
6	EST + NBD	4		No recurrence since years
7	EST + stone extr + NBD	4		Asymptomatic since 1.5 years
8	NBD	4		Asymptomatic since years
9	EST + stone extr + NBD	4		Recurrence after one week

EST= Endoscopic sphincterotomy, NBD= Naso-biliary drain

ment bilioptysis disappeared and the fistula tract could not be visualized at the control cholangiogram. The patient is now symptom free for a follow-up period of 4 years. In case 9 BBF recurred 1 week after removal of the first NBD placement. We placed another NBD into the CBD which remained 3 months. The patient is now symptom free since one year.

During these procedures no complication was observed.

DISCUSSION

BBF is a rare condition. It was first described by Peacock (1) in a patient with Hydatid disease of the liver. Echinococcal and amoebic disease of the liver account for the majority of instances of BBFs. The other causes are biliary tract diseases, trauma, tuberculosis, hepatic abscesses, syphilis, Hodgkin's disease and Ascaris lumbricoides. Congenital BBFs are extremely rare and were reported only in 11 cases throughout the literature (2-4).

There are two mechanisms through which a BBF may develop in cases of hepatic abscess or Hydatid disease of the liver. Firstly, an injury to the diaphragm may occur during operations related to the above mentioned cases, resulting in a communication between the biliary system and pleural cavity (pleurobiliary fistula). When intrapleural collection penetrates into the bronchi, BBF occurs (5). Secondly, in cases in which adhesions are present between the diaphragm and lung, an abscess or cyst may erode the diaphragm and perforated directly into the bronchial system (5). As in case 4. development of the BBF in biliary tract diseases require almost always extrahepatic obstruction and infection (5).

Clinically, the production of bile-stained sputum is virtually pathognomonic of BBF. Patients may have empyema, bronchiolitis, pneumonia and bronchiectasis of the right lower lobe and jaundice. Colvee (6) described a patient with gallstones in bile-stained sputum. In patients with long lasting BBF, secondary sclerosing cholangitis may develop as a result of biliary tract suppuration. Such patients suffer from intermittent cholangitis attacks.

The diagnostic methods of BBFs are ERCP, percutaneous transhepatic cholangiography (PTC), scanning, bronchoscopy and bronchography. ERCP should the procedure of choice in diagnosing BBFs when complete obstruction is not present (5,7). During ERCP it is not always possible to identify the fistula tract completely because the tract is usually too small. But contrast material extravasation into the pleural space can often be shown and it should be considered as an indirect sign for the presence of BBF in a symptomatic patient. In presence of extrahepatic biliary obstruction, PTC should be used to detection of BBFs. Bronchoscopy and bronchography have proven disappointing in demonstrating the fistula (5). Scanning has been used with some success

but should be further studied on (8,9). Abdominal ultrasound and/or computerized tomography can also help to delineate the abdominal anatomy and the extent of localized abscess formation.

In general, BBFs have been treated surgically. But surgery carries high risk and reoperation may be needed in some cases (10,11). Tube thoracostomy (10) and percutaneous abscess drainage (12) have been used in treatment, though rarely, but require further evaluation. There are many reports pointing to successful treatments of biliary-cutaneous fistulas using NBDs (13-15), with endoscopically sphincterotomy (16,18) and with endoscopically insertion of endoprothesis (17,18).

In the cases reported here, both BBF and BCF were treated successfully by endoscopically placed NBD. No complications were confronted.

In summary, BBF is a rare condition. Surgical treatment carries high risk and reoperation may be needed in some cases. Other therapy modalities should be further evaluated. Inserting a NBD into the CBD, we treated successfully nine patients with BBFs, 6 of which also had BCFs. It appears that in the future, surgery will be replaced by this conservative method in the treatment of BBFs, as well as BCFs.

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