



Conservative treatment of bronchobiliary fistula evaluated with magnetic resonance imaging

Konzervativno lečenje bronhobilijarne fistule procenjivano magnetnom rezonancom

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Abstract

Introduction. Bronchobiliary fistula (BBF) is a pathological communication between the bronchial system and the biliary tree that presents with biliptysis. Many conditions can cause its development. There is still no optimal therapy for BBF. Conservative treatment is rarely indicated, as was published before in a few cases. **Case report.** We presented a 71-year-old Caucasian Serbian woman with BBF secondary to previous laparotomy due to multiple echinococcus liver cysts. The diagnosis was established by the presence of bilirubin and bile acids in sputum and magnetic resonance cholangiopancreatography (MRCP). A repeat MRCP performed after conservative procedure, did not reveal fistulous communication. **Conclusion.** We suggest that in small and less severe fistulas between the biliary and the bronchial tract, conservative treatment may be used successfully, and invasive treatment methods are not needed in all patients.

Key words: bronchial fistula; biliary fistula; diagnosis; magnetic resonance imaging; drug therapy.

Apstrakt

Uvod. Bronhobilijarna fistula (BBF) predstavlja patološku komunikaciju između bronhijalnog sistema i bilijarnog trakta koja se prezentuje biliptizijama. Mnoga stanja mogu izazvati njen nastanak. Još uvek ne postoji optimalna terapija za BBF. Konzervativno lečenje je retko indikovano i predhodno je objavljeno samo kod nekoliko bolesnika. **Prikaz bolesnika.** Prikazali smo ženu, staru 71 godinu, sa BBF koja je nastala nakon predhodne laparotomije zbog multiplih ehinokoknih cisti jetre. Dijagnoza je postavljena na osnovu prisustva bilirubina i žučnih kiselina u sputumu i na osnovu holangiopankreatografije primenom magnetne rezonance (MRCP). Ponovljenim MRCP nakon konzervativnog lečenja nije potvrđeno prisustvo fistulozne komunikacije. **Zaključak.** Kod manjih i nekomplikovanih fistula između bilijarnog i bronhijalnog trakta, konzervativno lečenje može biti uspešno. Stoga, invazivne metode lečenja nisu neophodne kod svih bolesnika.

Ključne reči: fistula, bronhijalna; fistula, bilijarna; dijagnoza; magnetna rezonanca, snimanje; lečenje lekovima.

Introduction

Bronchobiliary fistula (BBF) is defined as the passage of bile in the bronchi and the presence of bile in sputum (biliptysis). The most common causes are trauma, bile duct obstruction, and liver infections such as hydatid cysts, echinococcosis, or amebic abscess¹. It can occur, rarely, in the congenital form². Therapeutic options include surgical repair or minimal invasive treatments such as nasobiliary drainage, endoscopic retrograde cholangiopancreatography (ERCP) (endoscopic sphincterotomy and biliary stenting) and percu-

taneous drainage. Herein, we described a patient with BBF after liver echinococcosis treated conservatively.

Case report

A 71-year-old Caucasian Serbian woman was admitted to our hospital with a 2-month history of biliptysis. The patient had the history of laparotomy one year previously because of multiple echinococcus liver cysts. Pericystectomy was done with omentoplasty and cholecystectomy. Eight months after the operation an abdominal ultrasound revealed fluid

collection in the liver. Percutaneous ultrasound-guided drainage was performed with daily drainage of 150–200 mL of green-yellowish content. Intravenous antibiotic therapy was applied in a few days followed with oral therapy. Two months after catheter droppage, the patient noticed green-yellowish sputum similar to previously drained content. Physical examination revealed fever (38°C), and decreased breath sound over the right basal lung fields. The sputum output range was 50–75 mL/day. Laboratory tests findings showed the elevated sedimentation rate of 70 mm/h (normal less than 30 mm/h) increased levels of aspartate aminotransferase (AST) 39 U/L (normal < 37 U/L), alkaline phosphatase 370 U/L (normal 40–120 U/L) and gamma glutamyl transferase (GT) 736 U/L (normal < 55 U/L). A chest radiograph showed right pleural effusion (Figure 1). Fiberoptic bronchoscopy was normal. Sputum samples detected the presence of bilirubin and bile acids in concentration of 14 $\mu\text{mol/L}$ (normal serum total bilirubin < 17.1 $\mu\text{mol/L}$, direct < $\mu\text{mol/L}$ 5, bile acids < 12 $\mu\text{mol/L}$) (Figure 2), while sputum and blood cultures were negative. ERCP was not successful in selective cannulation of common bile duct, so we could not visualize biliary system. Magnetic resonance cholangiopancreatography (MRCP) showed micro fistula between the intrahepatic bile ducts and the right pleural space (Figure 3). The patient was treated with antibiotics (cephalosporin of the third generation with metronidazole) and saline solutions intravenously in seven days. Bilioptysis disappeared after 5 days and the patient was discharged from the hospital in good clinical condition. Follow-up MRCP performed nine months later did not reveal any fistulous communication (Figure 4). Control chest radiograph made two years after was normal.



Fig. 1 – Chest radiograph showed right pleural effusion.

Discussion

This case is interesting to clinicians for showing that conservative antibiotic treatment after percutaneous drainage could be successful in the treatment of BBF, alleviating the need for more invasive management.

BBF represents an abnormal communication between the biliary system and bronchial tree. The most common causes for BBF in Western countries are bile duct obstruction due to cho-

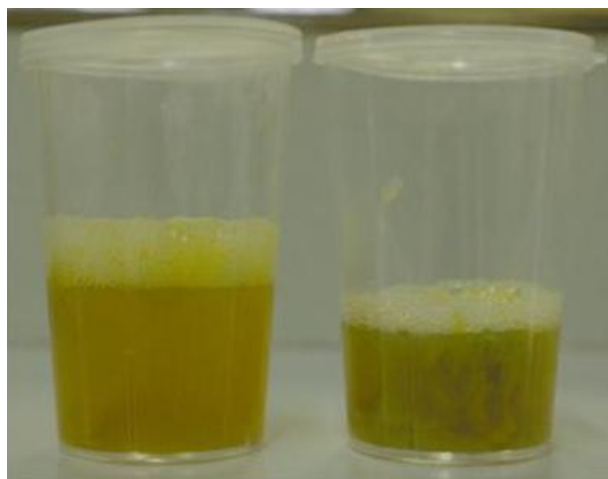


Fig. 2 – Sputum samples.

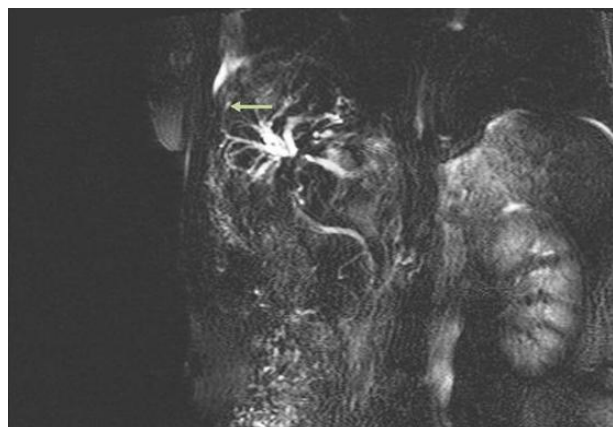


Fig. 3 – Magnetic resonance cholangiopancreatography finding showed a fistula between the intrahepatic bile ducts and the right pleural space.

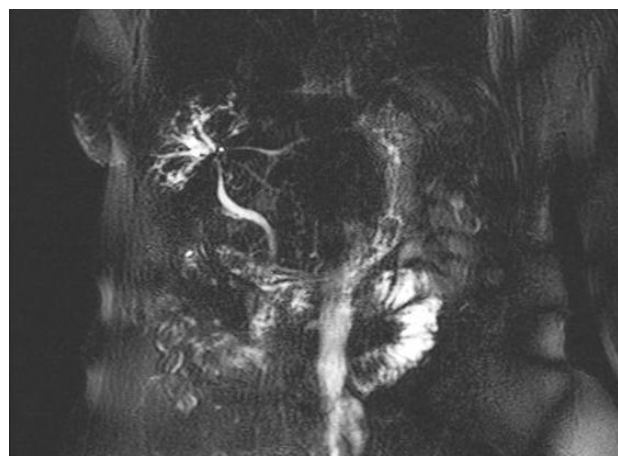


Fig. 4 – Magnetic resonance cholangiopancreatography repeated 9 months later showed no fistulous communication.

ledocholithiasis, malignancy, trauma or postoperative complications of hepatobiliary surgery³⁻⁵. In developing countries, echinococcal cysts, amebiasis, pancreatitis and tuberculosis are the most prevalent causes^{6,7}. BBF can be iatrogenic after radiofrequency ablation for liver metastases or congenital^{2,8}. Pathogenesis of BBF includes two mechanisms, biliary obstruction that produces the inflammatory process in the subdiaphragmatic space and rupture towards the bronchial tree⁹. The diag-

nosis of BBF is established by the presence of bile in sputum. Bilioptysis is pathognomonic of BBF. The radiographic finding includes pleural effusion with pneumonic consolidation as in the presented patient. Different diagnostic imaging investigations are useful in visualization of fistula such are percutaneous transhepatic cholangiography ERCP, MRCP, T-tube cholangiography, fistulography, bronchoscopy or bronchography, hepatobiliary iminodiacetic acid scyntygraphy^{10,11}. The main aim of BBF is to reduce the increased intrabiliary pressure in order to facilitate bile flow to the duodenum. Conservative, minimal invasive and surgical methods are used in the treatment of BBF. Conservative treatment can be used in small, not complicated cases. It includes antibiotic therapy and somatostatin or its analogues. Somatostatin or analogues, reducing the pancreatic and bile secretion, are the most commonly used in the treatment of acute or chronic pancreatitis complicated by the pancreatic-pleural fistula. The use of octreotide in BBF management has been described in the literature¹². Biliary decompression may be obtained by the following methods of the minimal invasive treatment such as nasobiliary and percutaneous transhepatic drainage^{13,14}. In our patient after liver collection occurred, we decided to use ultrasound-guided percutaneous drainage. Percutaneous drainage was successful with daily drainage of 150–200 mL of green-yellowish content. Two months after catheter dropped and a new one was not replaced. Until the presence of bilioptysis, the patient did not visit the doctor. On the other hand, in cases of large, persistent fistulas and non-effective non-invasive treatment, surgery is needed. The type of surgical pro-

cedure depends on etiology, location and complications of BBF. The following surgical procedures such as drainage of right subphrenic or hepatic abscess, closure of the fistula, resection of hydatid cyst, biliary drainage using T-tube and biliointeric anastomoses are performed in BBF^{3,5,8}. In cases of diaphragmatic, pleural, bronchial or pulmonary damage, closure of the diaphragm, pleural drainage, decortication or different pulmonary resections are used¹⁵.

In the presented patient ERCP was performed without successful selective cannulation of biliary tree, therefore we decided to treat our patient conservatively. Reduced biliary pressure after bilioptysis associated with antibiotic therapy could facilitate healing and closing of BBF. It is probably, because the described fistula was small. Somatostatin was not used in the presented patient, as was published before¹².

The clinical course of our patient improved. MRCP performed nine months later was without fistulous communication.

Early diagnosis is mandatory because of serious complications such are chemical pneumonitis, pneumonia or necrotizing bronchitis¹⁶.

Conclusion

We suggest that in small and less severe fistulas between the biliary and the bronchial tract, conservative treatment may be used successfully, and invasive treatment methods are not needed in all patients. Accurate therapeutic approach should be a key factor of management.

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