

Case Report

Bronchobiliary fistula: a rare complication after biliodigestive surgery for bile duct injury

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ABSTRACT

A bronchobiliary fistula (BBF) is an abnormal communication between the biliary and bronchial systems. It is a rare condition with an unclear etiology. The principal causes are hepatic hydatid cysts, obstructive and iatrogenic processes. Presenting symptoms are variable and range from productive cough to recurrent pneumonia. The finding of bilioptisis is pathognomonic. This case report presented a 22-year-old female patient who underwent a Roux-en-Y hepaticojejunostomy (RYHJ) for a bile duct injury and who later on, developed a bronchobiliary fistula.

Keywords: Bronchobiliary fistula, Bilioptisis, Hepaticojejunostomy, Bile duct injury

INTRODUCTION

A BBF is an uncommon condition with only 68 reported cases until 2011.¹ It was initially described in 1850 by Peacock as a complication of a hepatic hydatid cyst.^{2,3} In 1938, Oschsner observed a 4% incidence of BBF in a case series of 453 patients with pyogenic liver abscesses and an incidence of 10.5% in 3608 patients with subphrenic abscesses.³

This process is a consequence of an abnormal communication between the abdominal and thoracic cavities, which is created by a fistulous tract from the biliary tree, pleural space and lung parenchyma to the respiratory tract.⁴ Bilioptisis is pathognomonic for BBF and it is usually accompanied by productive cough and fever.^{1,2}

The causes can be congenital or acquired, the latter include multiple etiologies that can be divided in

infectious, obstructive, traumatic or iatrogenic processes.^{2,3}

The diagnosis is clinical, although it often includes additional workup as hepatobiliary scintigraphy (HBS), computed tomography (CT), magnetic cholangio-resonance, endoscopic retrograde cholangio-pancreatography (ERCP) and transhepatic percutaneous cholangiography (TPC).^{3,5}

Treatment of this condition is complex and until now, there is not a clear recommendation about the optimal management.⁵

CASE REPORT

A 22-year-old woman presented to the emergency department complaining of productive cough and yellowish sputum. Her past medical history was relevant for an open cholecystectomy four years before

presentation. In the early postoperative period, she presented with abdominal pain and hyperbilirubinemia. An ERCP was performed and a Strasberg E3 bile duct injury was diagnosed.

The patient was referred to our tertiary center where a biliodigestive procedure consisting of a roux-en-Y hepaticojejunostomy was performed.

Two years after the RYHJ, she visited the emergency department complaining of fever, productive purulent cough and jaundice. A magnetic cholangioresonance was ordered, documenting anastomotic stricture of the hepaticojejunostomy. The patient was taken to surgery for reconstruction of the anastomosis and discharged without further complications.

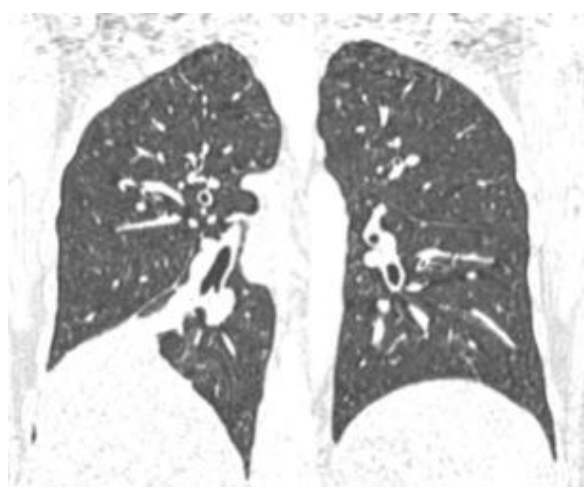


Figure 1: CT scan of the chest; CT scan shows a biliary fistula into the right lung cavity in the coronal plane.

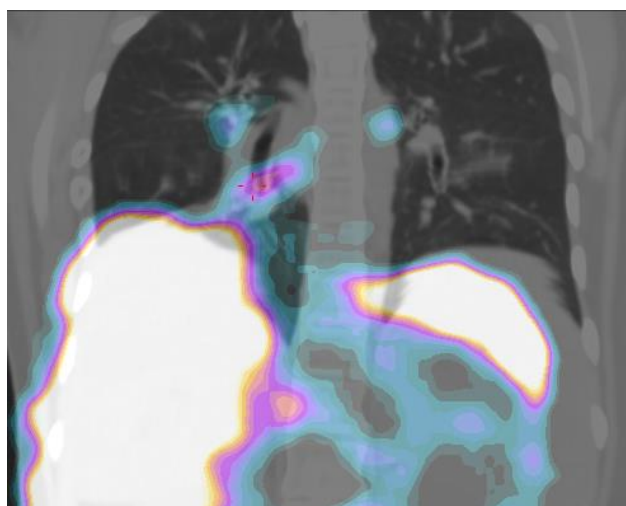


Figure 2: 99mTc-mebrofenin SPECT/CT is identified area of pathological uptake of the radiopharmaceutical arising from the liver dome and extending to the inferior bronchus of the right lung.

She presented a year after her last surgery complaining of productive cough with greenish sputum, thoracic pain, fever and jaundice. Physical exploration was significant for generalized jaundice and decreased breath sounds at the right lung base. Laboratory markers showed mild leucocytosis and hyperbilirubinemia with a cholestatic pattern. The thorax X-ray showed discrete right pleural effusion. A thorax CT scan showed hepatosplenomegaly and a fistulous tract from the hepatic segment VII to the 8th segment of the right lung (Figure 1).

The workup was completed with 99mTc-mebrofenin HBS combined with single photon emission computed tomography (SPECT/CT), where the presence of a bronchobiliary fistula was confirmed (Figure 2). Because anastomotic stricture of the hepaticojejunostomy was still suspected, a magnetic cholangioresonance was ordered and reported surgical changes and dilated intrahepatic biliary ducts, findings suggestive of a HJ stricture.

Once the diagnosis of BBF was confirmed, the pneumology department was consulted to evaluate the possibility of a bronchoscopy as non-operative initial management; however, the localization of the fistula at the 8th segment of the bronchial tree, precluded such treatment. As definitive treatment, a surgical procedure by gastrointestinal surgery alongside cardiothoracic surgery was proposed but the patient refused the intervention.

During her stay, she was managed with antibiotics (piperacillin/tazobactam and levofloxacin), ursodeoxycholic acid and cholestyramine. Resolution of the fever and biliptisis was achieved and the patient was discharged home.

DISCUSSION

A bronchobiliary fistula is a rare condition characterized by an abnormal communication between the biliary and bronchial trees.^{2,6} This communication was maintained by a pressure gradient between the positive pressure of the biliary ducts and the negative pressure of the thoracic cavity. The bile that acts as a corrosive agent and can generate both an inflammatory process and necrosis of the diaphragm.³

Among its various etiologies, a BBF can be congenital or acquired.^{1,3} Ferguson et al observed that the main cause were infectious processes; Morton et al found that obstructive biliary tract disease was the second most common cause.³

Crnjac et al proposed the classification of these etiologies in 5 groups (Table 1).

In the past this complication was frequently associated to infectious diseases, most commonly, hepatic hydatid disease. Since the inception of diagnostic and antimicrobial treatment, the disease's burden and its

complications had reduced considerably. In the literature, the most actual cases reported associate the presence of a BBF to hepatobiliary tumors.⁷ Tumors were accounted for 32% of the BBF cases, followed by biliary obstruction in 31%, infectious process in 12%, trauma in 10%, chronic pancreatitis in 3% and other etiologies in 12%.⁴

Table 1: Crnjac et al classification.

BBF etiologies	Examples
Congenital	
Infectious	Hepatic hydatid, cyst pyogenous, liver absces
Obstructive	Tumors, biliary tract stricture, hepatolithiasis and choledocholithiasis, chronic pancreatitis
Traumatic	Trauma, open or closed
Iatrogenic	Hepatic surgery, RFA (radiofrequency ablation), postoperative biliary tract stenosis

Table 2: Treatment of BBF.

Main categories	Types of treatment
Surgical	Laparotomy, thoracotomy MIS
Conservative	Bronchoscopy, ERCP±prothesis or DNB, transhepatic percutaneous drainage, fistula embolization, somatostatin analog
Combined	Abscess and fluid collection drainage+diversion of the biliary tract, surgery as a 2nd step procedure

For a BBF to be formed, there are some conditions that have to be met.⁷ They are a purulent or bile collection; any diaphragmatic injury; adhesions between the pleura and the lung. The clinical manifestations can be either acute or chronic, the former being the most common and with productive cough, fever and recurrent pneumonia as the predominant symptoms.^{2,3}

Bilious sputum or biliptysis is a pathognomonic finding.^{2,3} Not uncommonly, a yellowish-greenish sputum can be thought of as purulent, so an analysis was warranted as to determine the presence of bile.^{4,8} In the case of our patient, the second time she visited the emergency department, she had already complained of productive cough; it was highly probable that this was suggestive of the BBF a year before definitive diagnosis.

Other frequent symptoms were pleuritic pain, abdominal pain in the upper right quadrant, jaundice and fever.⁵ Some patients can even show a condition called cholelithoptysis which refers to the expectoration of gallstones.⁹ The diagnosis of a BBF was clinical.³ Still, a complete workup was recommended and often necessary to define the fistula characteristics as anatomy and localization. Since this condition was rare, a golden standard for its diagnosis had not been defined.⁴

The radiology studies that had been described to analyze and confirm this diagnosis were HBS, magnetic cholangioresonance, ERCP and percutaneous transhepatic cholangiography.⁵ According to what had been observed in several cases, the ERCP is the study with the highest sensibility, nevertheless, this study was not feasible in patients with a hepatojejunostomy.⁸

Several reports described the usefulness of HBS in the detection of even small BBF, had high specificity, low cost and was minimally invasive. Other advantages of HBS included localizing the fistulous tract, differentiating BBF from bile reflux and evaluating the effectiveness of therapy.¹⁰⁻¹² The treatment of a BBF is divided in three categories (Table 2).³

The most reported treatment of a BBF was surgery.² In 1966, Yilmaz et al published the first case series of 11 patients managed with a conservative approach and all of them presented complete resolution of the fistula. Since then, patients with acquired fistulas, especially those with a length <3 mm, were candidates for non-operative management.^{2,6} Singh et al reported a successful outcome in 62.5% of patients with BBF with non-operative management; this included chest tube, percutaneous drainage of subdiaphragmatic abscesses and fluid collections and ERCP drainage of the biliary tract.¹

The main goal of non-operative treatment was to reduce the pressure inside the biliary tract, gain control of the infectious source by draining abscesses and fluid collections and with these two interventions, prompt the closure of the fistula. In conditions that were contraindicated for conservative management or after its failure, the definitive treatment was surgery.⁸

Surgical treatment can be open or minimal invasion by laparoscopy or thoracotomy. A 2-step approach was recommended. The first consisted of source control by percutaneous biliary drainage and/or abscess drainage and then, this was followed by the definitive surgical correction of the underlying etiology.⁷ The critical steps that had been described for a successful surgery were fluid drainage, pleuro-pulmonar decortication, resection of the fistula tract, repair of the diaphragmatic defect and as key of the definitive treatment, the correction of the initial insult. In some instances, a lobectomy may be required if there was extensive lung damage.⁸ A systematic-review reported that the resolution rate for

interventionist and surgical management are of 97% and 85%, respectively.⁷

CONCLUSION

A BBF is a unusual condition that requires high clinical suspicion in patients that present with biliptysis and have medical history of hepatobiliary disease. In this article, we presented the case of a patient with a BBF posterior to a hepaticojejunostomy in the setting of a post-cholecystectomy bile duct injury. The treatment of this pathology can be conservative or surgical, depending on its etiology and anatomic characteristics.

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REFERENCES

1. Mauduit M, Rouze S, Turner K, Latour B, Verhoye JP. Combined thoracic and hepatobiliary surgery for iatrogenic bronchobiliary fistula. *Asian Cardiovasc Thorac Ann.* 2018;26(1):63-6.
2. Marcano L, Endis M, Siavichay M. Bronchobiliary fistula secondary to hepatic hydatid cyst in thoracic transit. clinical case. *Cirugía Pediátrica.* 2021;34:130-3.
3. Murcia A, Rivera J, Mejía G. (2017). Thoracoabdominal approach to bronchobiliary fistula. *Revista Colombiana de Cirugía.* 2017;32(3).
4. Odufalu FD, Zubairu J, Silverman W. Bronchobiliary fistula: a rare complication after pancreaticoduodenectomy. *BMJ Case Rep.* 2018.
5. Galindo P, Rengifo J, López C, Hernández G, Alfredo O, Galvis B, et al. Biliopleurobronchial fistula: a case report. *Revista Gastroenterología de Peru.* 2017;37(4):6-12.
6. Varela M, Durán F, Geribaldi N. Hepatobronchial fistula: a rare complication of a liver abscess. *Cirugía Espanola.* 2017;95(7):410-1.
7. Shim JR, Han SS, Park HM, Lee EC, Park SJ, Park JW. Two cases of bronchobiliary fistula: Case report. *Ann Hepato Bil Pancreat Surg.* 2018;22(2).
8. Crnjac A, Pivec V, Ivanecz A. Thoracobiliary fistulas: literature review and a case report of fistula closure with omentum majus. *Radiol Oncol.* 2013;47(1):77.
9. Bade BC, Janech MG, Ravenel JG, Stenbit AE. Bronchobiliary fistula presenting with recurrent pneumonia and cholelithoptysis. *Am J Med Sci.* 2015;350(1):72-3.
10. Alessio A, Viceconte G, Romano L, Sciuto R, Maini CL. detection of a suspected bronchobiliary fistula by hepatobiliary scintigraphy. *Ann Nucl Med.* 2008;641-3.
11. Fatma B, Corapcioglu F, Guvenc BH. Bronchobiliary fistula detected with hepatobiliary scintigraphy. *Clin Nucl Med.* 2006;31(4):237-9.
12. Bird R, Fagen K, Taysom D, Silverman ED. A case of bronchobiliary fistula in the setting of adult polycystic kidney and liver disease, with a review of the literature. *Clin Nucl Med.* 2005;30(5):326-8.

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