CASE REPORT

Unexpected Noninvasive Procedure in the Treatment of Bronchobiliary Fistula (BBF): Case Report

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Introduction
Bronchobiliary fistula is an abnormal connection between the bronchial tree and biliary tract and it is one of the rare diseases [1], but with high morbidity and mortality [2]. BBF is either congenital [3] or acquired [4]. Acquired BBF may be a consequence of local infections (amoebic liver abscess, complicated liver hydatid disease or pyogenic hepatic abscess), neoplasm, biliary tract obstruction, and trauma [5-62]. In a systematic review of 68 cases, Liao et al [4], 2011, reported that BBF cause was tumors (32.3%; 19.1% primary tumors; 13.2% metastatic tumors), followed by biliary stenosis (18%), cholangiolithiasis (13%), hepatic hydatidosis (12%), trauma (10%), multiple primary or single primary disease (6% for each) and chronic pancreatitis (3%). In 2012 Cao et al [1], reported a case in 48 years old male with hepatocellular carcinoma. In 2015, Dai et al [63] reported a case of BBF in a 65-year-old male with a hepatic abscess. In 70-year-old male, 2016, Hay et al [64] reported that BBF was developed following heptectomy of a recurrent hepatocellular carcinoma. Other cases of BBF was reported [65], 2016, in 77 year old female with hepatocellular carcinoma; in 2017, in 55 year old female with hydatid cyst [66]; In 2018, a 70-year-old male after pancreaticoduodenectomy [67], a 61-year-old male after radiofrequency ablation for HCC [68] and in 53-year-old female with breast cancer that was metastasized to liver [69]. While in 2015, BBF case reported in 3.5-year-old child after liver abscess rupture [70]. In Iraq, one review [71], in 2014, 14 cases of BBF were reported for the period from 2004 to 2010, four of them are with previous surgery to liver or right pulmonary hydatid cyst and 10 cases were with lower thoracic or penetrating abdominal injuries. Although the first case of BBF was reported by Peacock in 1850 [55], however, the incidence of BBF still a rare clinical condition.

Case report
In January, 2000 as a general surgeon on call, I was consulted at night to see a patient with clinical features of acute abdomen. The patient was sixteen year old thin female admitted to the medical ward a day before complaining of shortness of breath and right upper abdominal pain. At admission, patient was afebrile, not jaundiced, nor pale, nor cyanosed but anxious with mild dyspnea. Chest examination revealed basal right chest dullness and diminished air entry with mild abdominal distension .PA chest x-ray showed elevated right dome of the diaphragm. She is single, last menstrual period ended few days before admission with normal cycle menarche at age of thirteen.

On examination the patient was feverish (38.7 °C), pulse rate was 104 bpm, and Respiratory rate was 20/min. Blood Pressure was 115/80, anxious distressed by abdominal pain, not pale, nor jaundiced, nor cyanosed. Abdomen distended tender and
rigid with negative bowel sound. Digging back in her medical history she was diagnosed few days ago to have right pleural effusion by somebody and trial for pleural aspiration was done, but no urticarial manifestations noted. Laboratory investigation showed leukocytosis and normal liver function test (LFT). Ultrasound of the abdomen showed free fluid in Douglas pouch and subhepatic space the pelvis with the unilocular sub diaphragmatic right lobe hepatic cyst of 20x20 cm size, no dilatation of intrahepatic nor extrahepatic ducts (IHD&EHD), mildly distended intestinal loops with normal other viscera

According to the clinical features, laboratory findings, X ray and ultrasound, the case diagnosed as acute abdomen possibly due to infected hepatic hydatid cyst and a decision for urgent laparotomy was made. After rapid and short course of resuscitation and preparation urgent exploratory laparotomy through upper right paramedian incision was performed. More than 1.5 liters of turbid, non-bile stained, peritoneal fluid was aspirated, turbid non bile stained cystic fluid aspirated and 10% betadine solution injected intracystic as scolicidal agent, endocystectomy done as usual cavity washed with saline, left as such without obliterating suturing, no evident biliary communication could be identified, cyst closed with intracystic drain, common bile duct was not dilated, consistent with ultrasound finding. Peritoneal lavage with warm saline done and abdomen closed with the subhepatic drain.

Postoperative period was smooth sub hepatic drain removed on third postoperative day, but the intracystic drain was draining between 750-1000 ml of bile stained fluid daily, the patient was discharged after stich removal with the intracystic drain and advised to record daily drain output and to be seen weekly. After two weeks the drainage is nearly the same bile stained otherwise she looked well. Live function tests were normal, so she was referred for ERCP and sphincterotomy (S-ERCP) which was done and patient sent back after a week but without the intracystic drain which was incidentally dislodged during her transport. After 5 days she presented with fever, jaundice and cough, and was readmitted for management and soon she got bilipysis and the daily sputum collected was 650 – 1000 ml. Thus diagnosed as Bronchobiliary fistula (BBF). Chest examination and chest x ray showed signs of right basal consolidation, abdominal US revealed residual hepatic cystic cavity with no dilatation of IHD nor of EHD. Supportive therapy started to prepare her for operation and Roux-en Y cysto-jejunostomy, clubbing of fingers started to develop during this preoperative week period.

For the second operation, while the patient on the operative table induction done and endotracheal tube inserted (ETT) and checked to be inside trachea as usual, once connected to the ventilator, abdomen started to distend rapidly with cyanosis and bradycardia. ETT checked again while a small supraumblical incision done appeared useless, and nasogastric tube was inserted urgently. Once in the stomach there was gush of air and distension decreased and the cyanosis reduced and the pulse return to normal. Operation deferred and patient admitted to the intensive care unit after recovery.

Next morning patient was well, fever subsided and the daily collected sputum was less than 625 ml. On follow up days sputum decreased markedly, became less bile stained with time till it became clear and less than 100 ml/day. Patient afebrile not jaundiced, with good appetite, chest clinically and radiological improved. The patient was discharged after 17 days, cough was occasional and no expectoration, BBF assumed closed. Follow up was weekly for one month, then monthly for three months, and then every three months, till 2002 patient was well but with clubbing of fingers.

Discussion
Diversity of BBF causes influences their management and outcomes. In literature, only 14 cases of BBF were reported [71]. Since 1983 till now I was involved in management of two cases of BBF, first case in 1983 a 25-year-old male, shrapnel war injury, with the chest, diaphragmatic and extensive liver injury, complicated by pleural empyema and biliptysis treated operatively. The present case was the second patient in 2000 due to infected hydatid cyst following inadvertent aspiration. Retrospectively, with the subdiaphragmatic position of the cyst, it was not easy to explore the whole cystic cavity for cyst-biliary tree communication but gastric and bowel "insufflation" that occurred immediately following connection to ventilator raise the possibility of such problem, with distal intrahepatic biliary obstruction by debris or sludge. Professor Khalid Naji, an Iraqi general surgeon, he always repeats his famous statement regarding pathogenesis of gastrointestinal fistulae "with fistula, think about distal obstruction, when I close the door you will get out through the window, the window is the fistula"[72].

The diagnostic technique such as HIDA and contrast enhanced MRCP were not available at that time, January, 2000 and S-ERCP was the diagnostic and possibly the best therapeutic choice. It may be possible to visualize the site and cause of obstruction by contrast study through the intracystic drain but the incidental dislodgement of intra cystic drain without releaving the obstruction by S-ERCP, gave no choice other than invasive operative procedure for internal drainage of the cyst with adhesolysis of diaphragmatic adhesions.

Hydatid disease is endemic in Iraq [73,74] and involve the liver in 80% of cases [75], one of the common complication of liver hydatid cyst (LHC) is rupture into biliary tree which occur in 5-10% of cases and gives clinical manifestation similar to choledocholithiasis [76,77]. Intraperitonial rupture may occur spontaneously or traumatic leading to acute abdomen with or without urticaria [78]. In prospective study for66 case of complicated LHC done in Basrah Iraq from 1990-2000 intrabiliary communication was the commonest operative finding (48/66), 26 with jaundice and 22 without. Intraperitoneal rupture in 10/66 which seemed high. Operative treatment were according to whether patient jaundiced or not and operative findings, closure of communication by suturing with external drainage with or without decompression of biliary tree by T-tube or choledochoduodenostomy or transduodenal sphincterotomy. External drainage in non jaundiced done in eight, five of them remain with low output external biliary fistula for 7-9 months before closed spontaneously, internal drainage done in three cases. When S-ERCP being available we think it is a good minimally invasive technique to solve the problem of external fistulae and BBF as well, before shifting to more invasive option.

In conclusion, the endotracheal intubation and respiratory ventilator did bowel insuffulation through the fistulous tract and free normal bile drainage became possible, the door opened so the fistula closed.

References


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