

CASE REPORT

Open Access



Management of massive bilothorax post-percutaneous trans-hepatic biliary drainage

Thomas G. Vrachliotis¹, Stavros Spiliopoulos^{2*}  and Dionisios Voros³

Abstract

Background: Iatrogenic bilothorax is an infrequent major complication of percutaneous trans-hepatic biliary drainage (PTBD) and optimal treatment remains under-reported. The authors herein describe a case of PTBD complicated by a massive bilious effusion.

Case presentation: An 80-year-old male underwent PTBD due to malignant obstructive jaundice (total bilirubin 20.0 mg/dL). Following discharge, the patient was readmitted with severe dyspnea and recurrence of jaundice (total bilirubin 15.0 mg/dL). CT depicted the drainage catheter traversing the right costophrenic sulcus, complete obliteration of the right pleural space, no aerated right lung parenchyma and left mediastinal shift. The pleural effusion was successfully drained, and the biliary drainage catheter tract was sealed with Gelfoam pledgets and coils, to prevent bilopleural fistula formation and recurrence of the effusion.

Conclusions: In this case, a successful totally percutaneous management of iatrogenic bilothorax following PTBD is described. Due to the potential of rapid clinical deterioration and empyema formation, immediate evaluation and treatment are warranted. Prompt pleural drainage with antibiotic therapy and bile flow diversion from the pleural cavity are required.

Keywords: Bilothorax, Dyspnea, Percutaneous trans-hepatic biliary drainage

Background

Iatrogenic bilothorax after percutaneous trans-hepatic biliary drainage (PTBD) is rare and only sporadically reported in case reports. It represents, however, a serious complication as it can progress to thoracic empyema. A high degree of clinical suspicion is needed. Sealing the bilopleural fistula is mandatory, and this may prove very challenging technically. The authors present an educative case of bilothorax post-PTBD in an oncologic patient,

providing insight in the treatment steps successfully taken to overcome this fearful complication.

Case presentation

An 80-year-old male presents with obstructive jaundice (total bilirubin 20.0 mg/dL). Prior CT and MRI of the abdomen showed a 4,7 × 3,4 cm pancreatic head adenocarcinoma completely obstructing the common bile duct and possibly infiltrating the duodenum. There was moderate right-sided biliary dilatation and left hepatic lobe metastases. Endoscopic retrograde cholangiopancreatography (ERCP) to place a stent had not been successful. At this point, the patient was referred to our institution for further management. The patient had already been on antibiotic therapy. Due to the patients advanced age, overall status, and poor initial prognosis, the initial decision was to initially place a catheter and proceed

*Correspondence: stavspiliop@med.uoa.gr

² Interventional Radiology Unit, 2nd Department of Radiology, Division of Interventional Radiology, Medical School, National and Kapodistrian University of Athens, "Attikon" University General Hospital, 1st Rimini St, 12461 Chaidari, Athens, Greece

Full list of author information is available at the end of the article

with stenting in case of clinical improvement allowing a better prognosis. After written informed consent was obtained and under monitored assisted care (MAC), the right biliary system was uneventfully accessed and an 8,5Fr internal–external drainage catheter (Cook Medical, Bloomington, IN) was placed (Fig. 1). Within five days, the total bilirubin dropped to 12.0 mg/mL. On the sixth post-procedural day, the patient complained of mild dyspnea. Chest X-ray showed a moderate right pleural effusion, retraction, and downward course of the drainage catheter with angulation of its proximal part.



Fig. 1 Image of the right upper quadrant showing the 8,5Fr internal-external drainage catheter (white arrowheads) placed with its tip in the distal duodenum

Ultrasound of the liver showed no biliary dilatation. The catheter was repositioned over the wire and the patient was discharged. On the tenth post-procedural day, the patient returned complaining of severe dyspnea. The total bilirubin was 15 mg/dL upon readmission. Chest and upper abdominal CT clearly showed the drainage catheter traversing the right costophrenic sulcus, complete obliteration of the right pleural space, no aerated right lung parenchyma and left mediastinal shift (Fig. 2a). A right pleural drainage catheter was immediately placed, which drained 2000 mL of bile. Samples were sent for culture. The patient was subsequently brought to the Interventional Radiology suite. Under MAC, a hydrophilic super-stiff guide wire (Terumo Europe, Leuven, Belgium) was slowly and tediously advanced through the drainage catheter to the level of the ligament of Treitz. The catheter was removed and a 35 cm 7Fr metal Arrow sheath (Arrow, Super Arrow Flex, USA) was advanced over-the-wire with the tip into the duodenum. An Amplatz 0,035” wire (Cook Medical, Bloomington, IN) was then advanced through the sheath. A self-expandable 8 × 80 mm metal stent (Bard, E-Luminexx, Angiomed GmbH & Co., Karlsruhe, Germany) was deployed with the proximal end at the hepatic bifurcation and the distal end in the 2nd portion of the duodenum. There was excellent biliary drainage to the duodenum as seen with fluoroscopy (Fig. 2b). The guidewire was then removed. Prior to removing the Arrow sheath, the hepatic parenchymal tract was sealed with several pledgets of Gelfoam and three coils (Cook Medical, Bloomington, IN) (Fig. 3a,

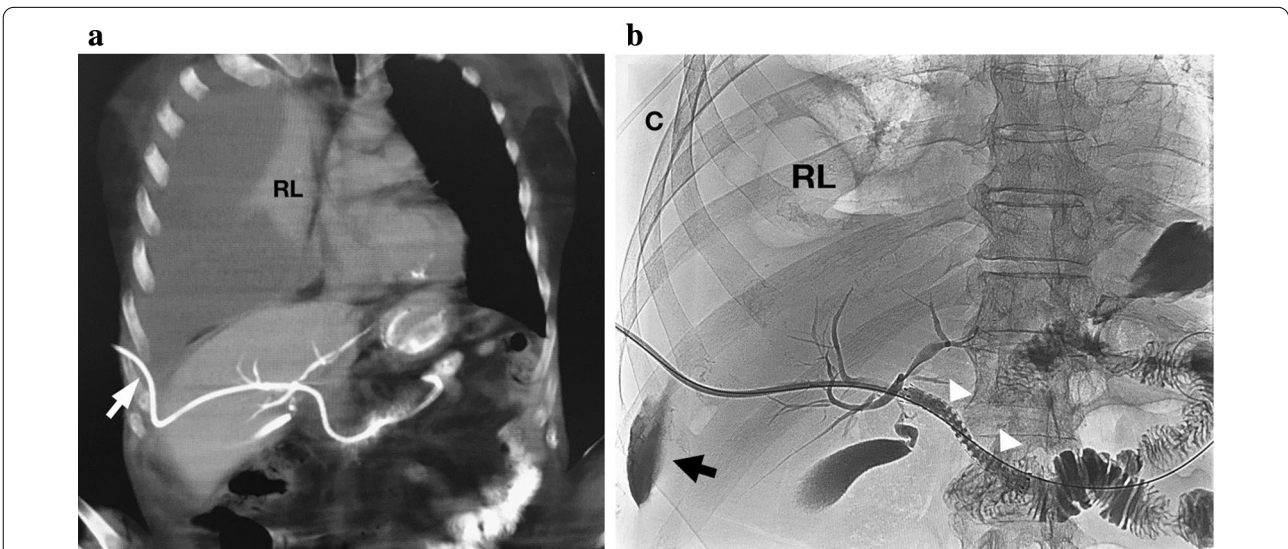


Fig. 2 a Coronal CT MPR reconstruction of the chest and upper abdomen. There is complete obliteration of the right pleural space by the bilious effusion, complete right lung collapse (RL) with left mediastinal shift. The drainage catheter (white arrow) clearly courses through the right costophrenic sulcus. **b** Image of the right upper quadrant immediately after stent deployment (white arrowheads) and prior to tract embolization. Black arrow points at the opacified right pleural sulcus. The right lung (RL) has partially re-expanded. C: pleural drainage catheter

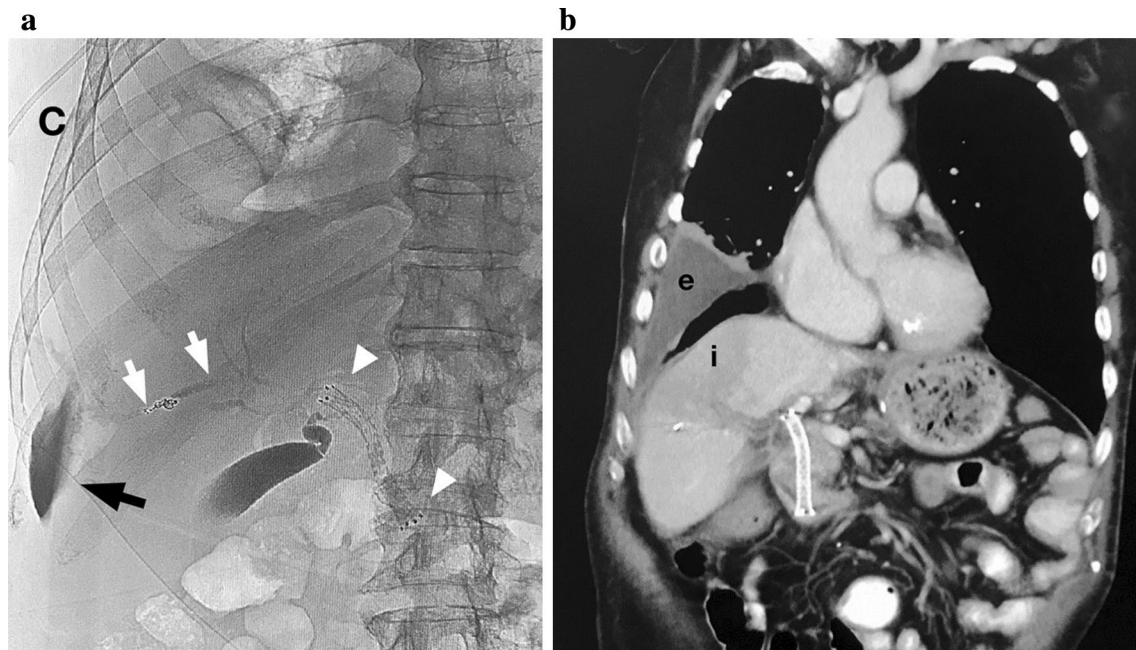


Fig. 3 **a** Final image post-stent placement (white arrowheads) and tract sealing (white arrows). Black arrow points at the opacified right pleural sulcus. C: pleural drainage catheter. **b** Coronal CT reconstruction of the chest and upper abdomen two months post-intervention shows stent in place and no biliary dilatation. There is residual right pleural effusion (e) and a right hepatic parenchymal infarct (i) from right portal vein thrombosis

b). The patient tolerated well the procedure, was discharged from hospital without symptoms one week later and underwent chemotherapy for the next five months during which time the bilirubin value had dropped to 2 mg/dL. He deceased a month later (six months from initial presentation) from metastatic liver disease progression and generalized malaise.

Discussion

Bilothorax or cholothorax is the accumulation of bile in the pleural cavity. It is the result of direct flow of bile from the biliary tree to the pleural space through a bilopleural fistula of traumatic, infectious or iatrogenic etiology [1]. In a review by Austin, et al., 59 cases of bilothorax were reported from 1960 to 2017 [2], 13 of which were related to percutaneous biliary drainage (PTBD). Karnik and Shair reported 14 cases from 2000 to 2019 [3], 4 of which were PTBD related. Bilothorax occurs almost exclusively on the right due to the anatomical location of the liver and biliary tree. It is a rare entity, which needs, nevertheless, immediate treatment and management due to the toxicity and inflammatory effects of bile in the pleural cavity that may result in pleural empyema.

Anatomically, each hemithorax is internally invested by parietal pleura. As the pleura changes direction caudally reflecting to cover the right hemidiaphragm, this reflection forms a continuous line, the right costophrenic

sulcus-pleural reflection. On the skin, from front to back, this pleural reflection corresponds to an imaginary line that connects the following spots: 6th rib-anterior mid-line, 8th rib-right mid-clavicular line, 10th rib-right mid-axillary line, 12th rib-scapular line. When accessing the right biliary system, classical teaching states that the fluoroscopically guided puncture, should be performed on the mid-axillary line below the 10th rib, with the tip of the needle angled 10 degrees cephalad and 10 degrees forward angulation. Staying below the 10th rib and just anterior to the mid-axillary line minimizes the chances of crossing the pleura [4]. While pleural complications are relatively rare (0,5% of PTBD cases performed) [5], it is not uncommon to traverse the pleural space [6] during PTBD. The exact rate, however, is unknown. In a series of 230 PTBDs, utilizing a right-sided intercostal approach only one case of bilothorax occurred [7]. In another series of 419 PTBD procedures, two cases of bilothorax were reported [8].

We believe that in our case, bilothorax occurred for the following reasons: first, and most important, the initial puncture was certainly high, and well through the pleural space as proven, although fluoroscopically it seemed acceptable. Second, the patient was sedated, thereby, full excursion of the lungs and hemidiaphragms was not done to fully appreciate the caudal extent of the right costophrenic sulcus. Third, there was possible malfunction of

the drainage catheter associated with the retraction. And, finally, the negative intrathoracic pressure within the pleural space [3] created a de-facto suction effect, thereby directing bile flow toward the pleural space instead of the duodenum.

Drainage catheters are usually secured on the skin with sutures and/or an adhesive locking device to prevent from external causes of dislodgment. It is important to allow for some slack to be present otherwise the catheter may coil in the intraperitoneal space (or in the pleural space). This is more likely to occur when the procedure is done under sedation, thus the liver is high within the thoracic cavity, which did occur in our patient [4]. Coiling of the catheter in our case was further exacerbated by the increased downward pressure on the liver, exerted by the massive bilothorax. That created an angled rigid catheter course resulting in very tedious guide wire advancement through the catheter lumen. Loss of access in this case would have been catastrophic.

Bilothorax should be suspected in any patient post-PTBD who develops dyspnea, ipsilateral pleuritic chest pain, or septicemia, especially when the biliary drainage catheter has been dislodged. Pleural drainage must be performed with fluid culture/analysis and prompt initiation of empiric antibiotic treatment. At the same time, bile should be prevented from further entering the pleural cavity. Upsizing the drainage catheter to 12Fr or 14Fr was not considered, as the on-going clinical scenario could easily repeat itself. A new puncture was also not an option as there was no biliary dilatation. After documenting that the stent we placed was widely patent, the tract was successfully sealed with Gelfoam pledges and coils to prevent further leakage of bile. The use of compressed gelatin foam pledget in a pre-loaded delivery device (Hep-Plug™) along the intrahepatic tissue tract has also been reported [9].

Conclusions

Due to the potential of rapid clinical deterioration and empyema formation, immediate evaluation and treatment are warranted in cases of suspected bilothorax following PTBD. A high degree of clinical suspicion is needed, prompt pleural drainage with antibiotic therapy and bile flow diversion away from the pleural cavity.

Abbreviations

ERCP: Endoscopic retrograde cholangiopancreatography; MAC: Monitored assisted care; PTBD: Percutaneous trans-hepatic biliary drainage.

Acknowledgements

Not applicable.

Authors' contributions

TGV was a major contributor in writing the manuscript and contributed with conception, literature review and analysis, drafting, critical revision and editing.

SS contributed to this paper with writing, literature review and analysis, drafting, critical revision and editing. DV contributed to this paper with writing, literature review and analysis, drafting, critical revision and editing. All authors read and approved the final manuscript.

Funding

This case report received no funding.

Availability of data and materials

Will be available upon reasonable request.

Declarations

Ethics approval and consent to participate

Ethics approval was waived for this case report by the Hospital's Ethics Committee. Written informed consent was obtained by the patient before each procedure described.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests

The authors declare that they have no competing interest.

Author details

¹Department of Interventional Radiology, Henry Dunant Hospital Center, Athens, Greece. ²Interventional Radiology Unit, 2nd Department of Radiology, Division of Interventional Radiology, Medical School, National and Kapodistrian University of Athens, "Attikon" University General Hospital, 1st Rimini St, 12461 Chaidari, Athens, Greece. ³Department of Surgery, Henry Dunant Hospital, Athens, Greece.

Received: 27 July 2021 Accepted: 21 February 2022

Published online: 03 March 2022

References

- Jenkinson MRJ, Campbell W, Taylor MA (2013) Bilothorax as a rare sign of intra-abdominal bile leak in a patient without peritonitis. *Ann R Coll Surg Engl* 95(12–13):10
- Austin A, Fox N, Huggins JT, Chopra A (2017) The green pleural effusion: a comprehensive review of the bilothorax with case series. *Pleura* 4:21–31
- Karnik NP, Shair KA (2019) Iatrogenic bilothorax: a rare complication of orthotopic liver transplant. *ACG Case Rep J* 6:1–4
- Kim SH, Zangan SM (2015) Percutaneous transhepatic biliary drainage complicated by bilothorax. *Semin Intervent Radiol* 32:54–56
- Saad WE, Wallace MJ, Wojak JC, Kundu S, Cardella JF (2010) Quality improvement guidelines for percutaneous transhepatic cholangiography, biliary drainage, and percutaneous cholecystostomy. *J Vasc Interv Radiol* 21(6):789–795
- Strange C, Allen ML, Freedland PN, Cunningham J, Sahn SA (1988) Biliopleural fistula as a complication of percutaneous biliary drainage: experimental evidence for pleural inflammation. *Am Rev Respir Dis* 137(4):959–961
- Nichols DM, Cooperberg PL, Golding RH, Burhenne HJ (1984) The safe intercostal approach? Pleural complications in abdominal interventional radiology. *AJR Am J Roentgenol* 142(5):1013–1018
- Weber A, Gaa J, Rosca B et al (2009) Complications of percutaneous transhepatic biliary drainage in patients with dilated and nondilated intrahepatic bile ducts. *Eur J Radiol* 72:412–417
- Kortes N, Gnutzmann D, Konietzke P et al (2017) Occlusion of a Long-Term Transpleural Biliary Drainage Tract Using a Gelatin Pledget (Hep-Plug™). *Cardiovasc Intervent Radiol* 40:1800–1803

Publisher's Note

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.