

SPONTANEOUS LIFE-THREATENING HEMOBILIA DURING ACUTE LIVER FAILURE SUCCESSFULLY TREATED WITH TRANSARTERIAL EMBOLIZATION

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A 28-year-old patient admitted with jaundice, vomiting and deteriorating coagulopathy was diagnosed with acute liver failure. After listing for urgent transplantation, he developed Boerhaave's syndrome and massive hemobilia, two life-threatening complications. Massive hemobilia secondary to a fistula between the right hepatic artery and the right bile duct occurred several days after transjugular biopsy and was controlled with fluid resuscitation, transfusion and arterial embolization. Two days later he was transplanted successfully, and is currently doing well after more than 72 months. Aggressive treatment of potentially reversible complications during acute liver failure whilst awaiting transplantation is mandatory to allow survival of these patients.

Key-words: Hemobilia – Arteribiliary fistula – Boerhaave's syndrome – Acute liver failure – Embolization.

Acute liver failure is defined as a syndrome resulting from a massive and rapid impairment of all crucial liver functions in patients with previously unknown liver disease. The syndrome of acute liver failure includes signs directly related to the liver cell damage, altered liver function and the consequences on other organs. These manifestations essentially include jaundice, elevated serum transaminase levels, encephalopathy, coagulopathy, severe infections, and renal, cardiovascular, pulmonary, metabolic disorders. The prognosis depends on the extent of the liver cell damage, the liver's regenerative capacity, associated comorbidities and possible therapeutic interventions as liver transplantation. The first objective in the management of patients with acute liver failure is to maintain an optimal condition for liver regeneration together with taking the decision with appropriate timing for emergency transplantation.

Here, we present and discuss the case of an acute liver failure in a young patient who developed a rupture of the oesophagus and spontaneous massive hemobilia. Both these conditions were life-threatening but treated successfully, allowing a consecutive successful liver transplantation.

Case presentation

A 28-year-old patient was referred to our transplantation centre because of asymptomatic jaundice, regurgitations, increasing dyspepsia and

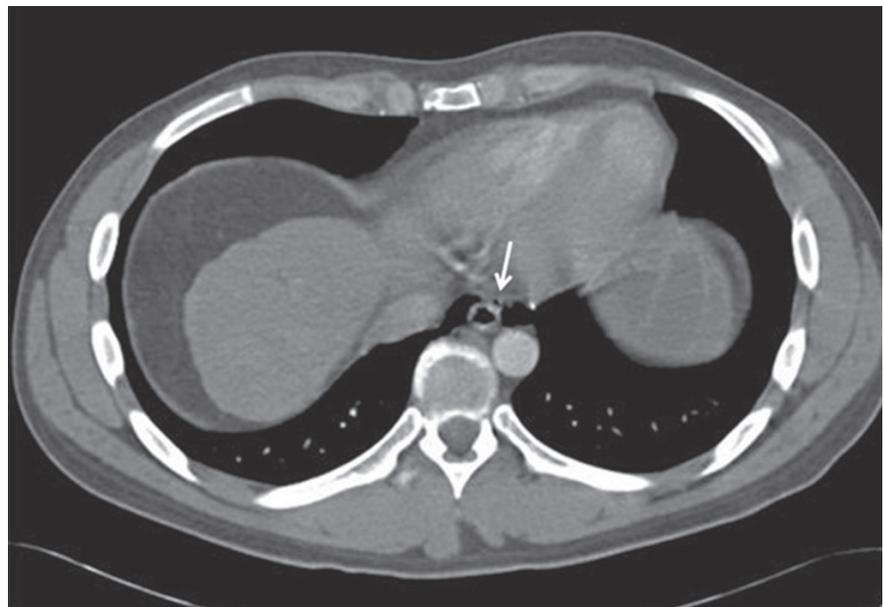


Fig. 1. – Representative image from a CT scan displaying free air around the distal oesophagus (arrow) as diagnosed in Boerhaave's syndrome.

vomiting since 2 weeks in combination with deteriorating coagulopathy. His previous medical history was uneventful. There was no family history of liver disease, no recent travelling or contact with toxic agents. He did not take any medication or illicit drugs but there was occasional excessive alcohol intake during weekends. On admission, physical examination revealed jaundice, without other signs of liver failure or long-standing liver dysfunction.

Initial laboratory tests revealed elevated transaminases (AST

2,456 IU/L, normal < 38 IU/L), direct hyperbilirubinaemia (26.08 mg/dl, normal range < 1 mg/dl) and coagulopathy (INR 2.3). Biochemical screening for viral and immunological causes of acute liver failure was negative; levels of ceruloplasmin, ferritin and α_1 -antitrypsin were normal. Ultrasound of the abdomen revealed a relatively small liver and mild splenomegaly without signs of biliary obstruction. Transjugular liver biopsy via right hepatic vein was performed uncomplicated and was suggestive for toxic hepatitis with more than 50% necrotic liver parenchyma. In accordance with the King's College criteria for acute liver failure (1), he was listed for urgent liver transplantation.

Within 12 hours after listing and following a vomiting episode, his clinical condition suddenly deteriorated,

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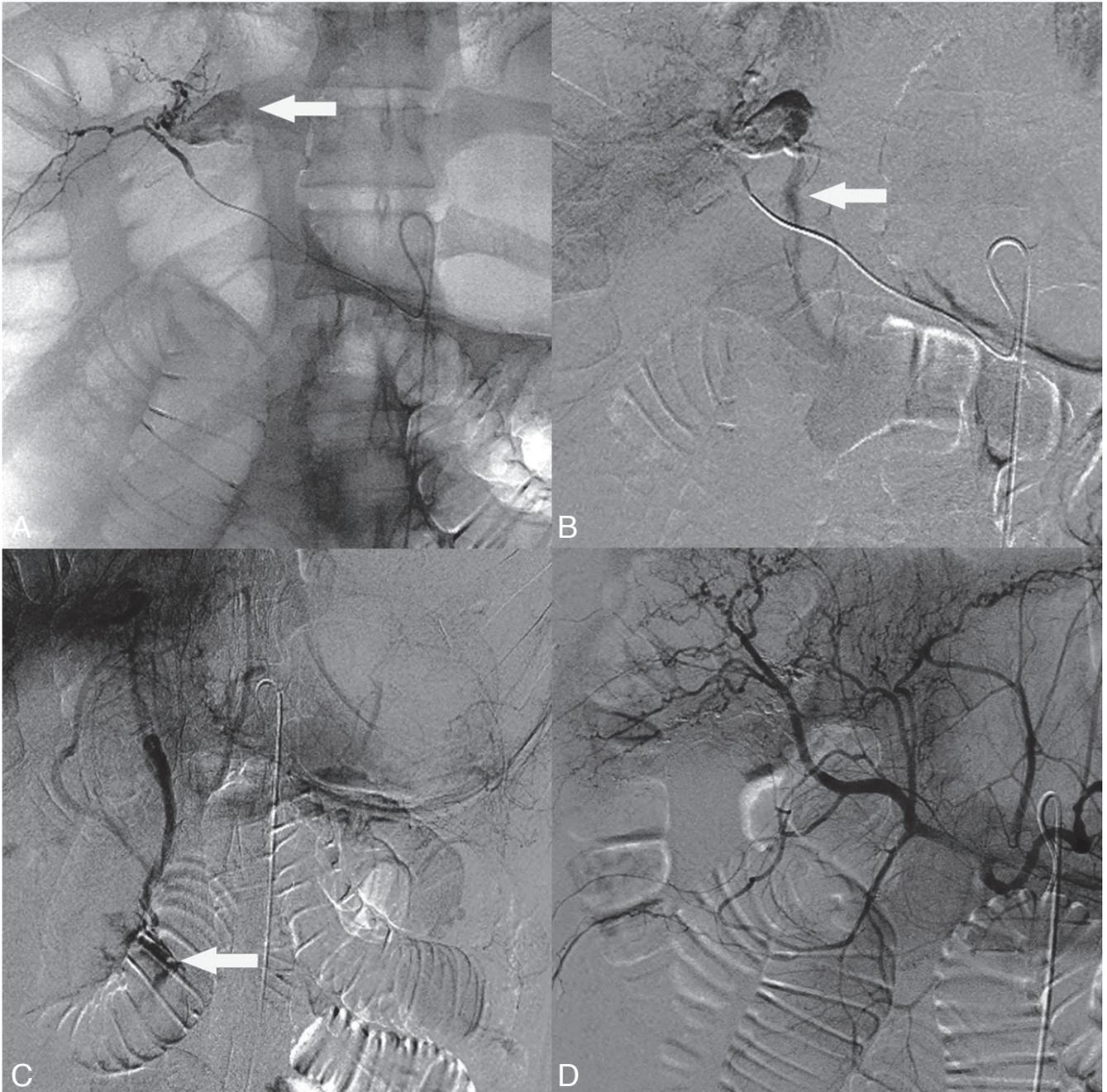


Fig. 2. — Selective angiography via the right femoral artery and catheterisation of the celiac trunc. Following contrast injection, immediate contrast extravasation of the right bile ducts and gall bladder is observed (A, arrow). Within seconds, the contrast fluid emerges through the common bile duct (B, arrow) into the duodenum (C, arrow). After embolization the biliary system was no longer visible (D).

developing severe epigastric pain with signs of localised peritonitis accompanied by melaena. CT of the abdomen revealed free air around the oesophagus, consistent with a transmural tear (Boerhaave's syndrome) which was treated conservatively (Fig. 1). Within 24 hours, the patient was found unconsciously and in hypovolemic shock. Laboratory analysis showed Hb 2.9 g/dL

(normal range 12-16 g/dl) and INR 10. He was resuscitated aggressively with colloids, blood and plasma transfusion, and administration of recombinant factor VIIa. An urgent gastroduodenoscopy revealed a continuous blood flow from the Vater's ampulla, stating the diagnosis of massive hemobilia. Consecutive angiography revealed an arterio-biliary fistula between a distal branch

of the right hepatic artery and the right bile duct with contrast extravasation in the gallbladder, through the common bile duct and into the duodenum (Fig. 2A-C). This arterio-biliary fistula was treated successfully with arterial embolization, using a mixture of embucrylate (Histoacryl[®], B. Braun, Melsungen, Germany) and lipiodol (ethiodized oil, Laboratoires Guerbet, Aulnay-sous-Bois, France). Two days

after the hypovolemic shock episode, the patient underwent an orthotopic liver transplantation. His recovery was uneventful apart from an anastomotic biliary stricture which was treated endoscopically. He is currently doing well, more than 72 months after the transplantation.

Discussion

Management and stabilization of patients with acute liver failure is challenging but crucial to maintain a condition as optimal as possible for liver regeneration and/or urgent transplantation. Once liver failure is diagnosed, it is mandatory that patients are referred to a centre with a liver transplantation program and experienced interventional radiology. As discussed here, this patient developed – whilst awaiting an urgent transplantation – two major life-threatening conditions which were treated quickly and adequately enabling a live-saving liver transplantation.

This patient first suffered from a Boerhaave's syndrome. This relatively rare disorder is caused by a barogenic rupture of the oesophagus and is associated with a high mortality (20-40%). In normal conditions, conservative treatment and/or endoscopic intervention are indicated in the absence of systemic effects whereas urgent surgical repair is mandatory in case of e.g. sepsis (2). The patient was treated conservatively since at that time his clinical condition remained stable in the next hours after onset of the symptoms, hereby also taking into account a high mortality risk-related surgery during acute liver failure.

A second complication was a massive hemobilia secondary to an arteriohepatic fistula. Arteriohepatic fistulas have been described previously as rare but potentially fatal complications, mainly after trauma or liver biopsy (3). The risk of an arteriohepatic fistula is higher after a transjugular liver biopsy (1.2%) compared to the percutaneous approach (0.006%) (4). Obviously, a transjugu-

lar biopsy was preferred here over a percutaneous approach because of the profound coagulopathy which is considered a contra-indication for percutaneous liver biopsy. In the presented case, however, it is unlikely that the massive hemobilia was directly related to the transjugular biopsy, since the hemobilia presented several days after the transjugular biopsy. Moreover, during arteriography there were several small bleeding points (distal branches of right hepatic artery) which were not located within the area of biopsy. However a facilitating effect of the transjugular biopsy can never be excluded. It is most likely that the fistula eroded slowly through the massive hepatocellular necrosis (as documented by liver biopsy) and on-going inflammation into small segmental branches of the right hepatic artery resulting in an arteriohepatic fistula.

In this particular case, we opted for glue embolization instead of coiling. Injection of glue is fast and a few droplets can immediately occlude all bleeding points. Glue-embolization of the hepatic artery has also been used for other indications, like treatment of unresectable neuroendocrine liver metastases without clear damage to the residual hepatocytes or biliary tract (5). In general, transarterial embolization is regarded an effective treatment for hemobilia and arteriohepatic fistulas (6-9). In case of failed transarterial haemostasis, surgical ligation of the feeding artery and/or liver resection (e.g. segmentectomy) may be the preferred method of salvage (10).

Conclusion

Aggressive treatment of potentially reversible life-threatening complications, even when occurring during acute liver failure whilst awaiting urgent liver transplantation, is essential. In this particular case, intensive medical treatment and interventional radiological techniques, performed in emergency setting, were crucial for the patient's survival until

a suitable donor liver became available.

References

- O'Grady J.G., Alexander G.J., Hayllar K.M., Williams R.: Early indicators of prognosis in fulminant hepatic failure. *Gastroenterology*, 1989, 97: 439-445.
- de Schipper J.P., Pull ter Gunne A.F., Oostvogel H.J.M., van Laarhoven C.J.H.M.: Spontaneous Rupture of the Oesophagus: Boerhaave's Syndrome in, 2008. Literature Review and Treatment Algorithm. *Dig Surg*, 2009, 26: 1-6.
- Gurakuqi G.C., Stadlbauer V., Portugaller H.R., Högenauer C., Trauner M., Stauber R.E.: Fatal hemobilia resulting from an iatrogenic arteriohepatic fistula as a rare complication of transjugular liver biopsy. *Eur J Gastroenterol Hepatol*, 2008, 20: 83-86.
- Gorriz E., Reyes R., Lobrano M.B., et al.: Transjugular Liver Biopsy: A Review of 77 Biopsies Using a Spring-Propelled Cutting Needle (Biopsy Gun). *Cardiovasc Intervent Radiol*, 1996, 19: 442-445.
- Loewe C., Schindl M., Cejna M., Niederle B., Lammer J., Thurnher S.: Permanent transarterial embolization of neuroendocrine metastases of the liver using cyanoacrylate and lipiodol: assessment of mid- and long-term results. *AJR Am J Roentgenol*, 2003, 180: 1379-1384.
- Xu Z.B., Zhou X.Y., Peng Z.Y., Xu S.L., Ruan L.X.: Evaluation of selective hepatic angiography and embolization in patients with massive hemobilia. *Hepatobiliary Pancreat Dis Int*, 2005, 4: 254-258.
- Srivastava D., Sharma S., Pal S., et al.: Transcatheter arterial embolization in the management of hemobilia. *Abdom Imaging*, 2006, 31: 439-448.
- Bloechle C., Izbicki J.R., Rashed M.Y., et al.: Hemobilia: presentation, diagnosis, and management. *Am J Gastroenterol*, 1994, 89: 1537-1540.
- Hidalgo F., Narváez JA., Reñé M., Domínguez J., Sancho C., Montanyà X.: Treatment of hemobilia with selective hepatic artery embolization. *J Vasc Interv Radiol*, 1995, 6: 793-798.
- Dousset B., Sauvanet A., Bardou M., Legmann P., Vilgrain V., Belghiti J.: Selective surgical indications for iatrogenic hemobilia. *Surgery*, 1997, 121: 37-41.