Published online: 2019-09-16

Case Report

Endoscopic Biliary Stenting for Portal Biliopathy Perforating Paracholedochal Collateral: A Rare Complication

Kartik Goyal, Sabir Hussain, Pawan Kumar Garg[1], Narender Bhargava, Vaibhav Kumar Varshney[2]

Department of Gastroenterology, Mathura Das Mathur Hospital, Departments of [1]Diagnostic and Interventional Radiology and [2]Surgical Gastroenterology, All India Institute of Medical Sciences, Jodhpur, Rajasthan, India

Abstract

Extrahepatic portal venous obstruction (EHPVO) usually presents with upper gastrointestinal bleed in the first decade. Symptomatic portal hypertensive biliopathy is seen in minority of patients with EHPVO. With the use of endoscopic intervention, biliary drainage is maintained in these patients. Various procedural complications have been linked while performing endoscopic retrograde cholangiography and stenting; however, these are managed conservatively. Here, we are highlighting a case of EHPVO with symptomatic portal biliopathy who bled from paracholedochal collateral after biliary stenting and managed successfully with multidisciplinary approach.

Keywords: *Extra-hepatic portal venous obstruction, paracholedochal plexus, portal biliopathy, stent*

Introduction

Extrahepatic portal venous obstruction (EHPVO) is characterized by a chronic blockage of portal vein, usually presented in childhood with signs of portal hypertension (PHT) and its complications.[1] The diagnosis of EHPVO is mainly clinical – features of PHT without any evidence of liver dysfunction and presence of portal cavernoma on Doppler ultrasound (US).

Portal biliopathy is seen in 80%–100% of patients with EHPVO, however, remains asymptomatic in 62%–95% patients.[2] The patient usually present with features of jaundice with cholestatic features, biliary colic, and recurrent cholangitis, presence of bile duct stones and abnormal liver function test.

Endoscopic retrograde cholangiopancreatography (ERCP) with stenting is usually required in case with obstructive jaundice and cholangitis.[2] ERCP and stent-induced bleeding from venous plexus in portal hypertensive biliopathy (PHB) is extremely rare.[3,4] Here, we are discussing a case of EHPVO with symptomatic PHB who underwent biliary stenting which led to torrential bleeding from paracholedochal collateral and its management.

Case Report

A 31-year-old male patient presented with painful progressive jaundice from the last 1 month associated with intermittent episodes of high-grade fever. He was icteric with moderate splenomegaly and tenderness in the right hypochondrium on abdominal examination. He was diagnosed as a case of EHPVO ~15 years back and on secondary prophylaxis with beta blockers for variceal bleeding. He also underwent open cholecystectomy 4 months back for symptomatic cholelithiasis. With the background of EHPVO and current clinical scenario; he was worked up for portal biliopathy.

Biochemical investigations were suggestive of raised leukocyte count (19,700/mm^3) and deranged liver function test: bilirubin (27.5 mg/dl) with direct component (13.57 mg/dl); alkaline phosphatase (311 IU/L). Ultrasound abdomen was suggestive of portal cavernoma and dilated common bile duct (CBD) with echogenic sludge within. Magnetic resonance imaging abdomen with magnetic resonance cholangiopancreatography reported stricture at lower end CBD with proximal dilation of CBD (21 mm) and intrahepatic biliary

Address for correspondence: Dr. Vaibhav Kumar Varshney, Department of Surgical Gastroenterology, All India Institute of Medical Sciences, Basni Industrial Area, Phase-II, Jodhpur - 342 005, Rajasthan, India.
E-mail: drvarshney09@gmail.com

How to cite this article: Goyal K, Hussain S, Garg PK, Bhargava N, Varshney VK. Endoscopic biliary stenting for portal biliopathy perforating paracholedochal collateral: A rare complication. J Dig Endosc 2019;XX:XX-XX.

Access this article online

Quick Response Code:

Website: www.jdeonline.in

DOI: 10.4103/jde.JDE_37_18

radicals (IHBR) with few soft calculi (2–4 mm). Upper gastrointestinal (GI) endoscopy revealed eradicated esophageal varices and no gastric varices. With these findings, he underwent ERCP under sedation. Selective CBD cannulation done and cholangiogram revealed dilated IHBR with grossly dilated CBD except narrowed distal segment. Endoscopic papillotomy was done using pull type papillotome, and minor bleed was seen which resolved spontaneously. Stones were extracted with the help of extraction balloon and over the wire and 10 fr × 10 cm straight plastic stent was placed. Immediately, a gush of blood was noted from ampulla into the duodenum with stent *in situ*. The procedure was abandoned as patient developed hypotension.

The patient was shifted to the Intensive Care Unit (ICU) for resuscitation, and nasogastric tube was placed and intravenous fluids, inotropes, and terlipressin were started. Injectable antibiotics with injection tranexamic acid were also supplemented. Two units of packed red blood cells were transfused over 6 h.

After maintaining hemodynamic stability, contrast-enhanced computed tomography (CT) abdomen with angiogram and portal venography was performed. Noncontrast CT abdomen [Figure 1a] showed contrast within dilated intrahepatic biliary radicles which was injected during ERCP. Biliary stent was away from hepatic artery branches with no active extravasation or pseudoaneurysm seen [Figure 1b].

Portal venous phase [Figure 2a and b] showing multiple paracholedochal collaterals with dilated CBD. Tip of the biliary stent was misplaced and seen within one of the large paracholedochal collateral. Interventional radiologist and surgical gastroenterologist opinion sought. Meanwhile, inotropic support was gradually tapered and terlipressin was continued for 72 h.

Percutaneous transhepatic biliary drainage (PTBD) was done to decompress biliary system and with the fear of displacing hematoma around collateral puncture site; external catheter was placed at hilum [Figure 3]. He responded to treatment well with the stabilization of hemoglobin and decrement in bilirubin and improvement in liver function test.

After 7 days, ERCP was repeated, and *in situ* stent was removed, and 8 fr × 10 cm double pigtail stent was placed into the right ductal system. The procedure was uneventful. Later, PTBD was clamped and with continued decreasing level of bilirubin; it was removed. The patient was subsequently discharged with an advice of follow-up after 14 days. After a follow-up of 3 months, the patient was symptomatically improved with no further episodes of GI bleed and decrement in the level of bilirubin.

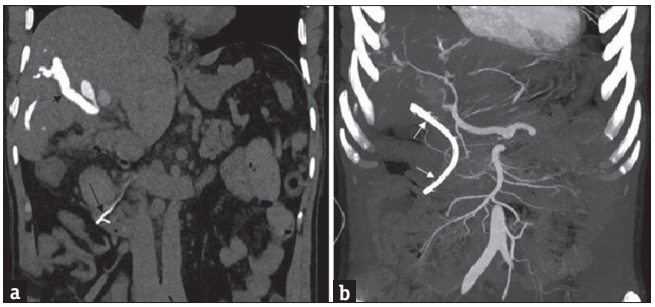

Figure 1: (a) Noncontrast computed tomography abdomen in coronal plane showing contrast filled dilated intrahepatic biliary radicles (small black arrow). The lower end of common bile duct stent (large black arrow) is also seen; (b) Coronal maximum intensity projection in arterial phase showing biliary stent (white arrow) away from hepatic artery branches with no active extravasation or pseudoaneurysm seen

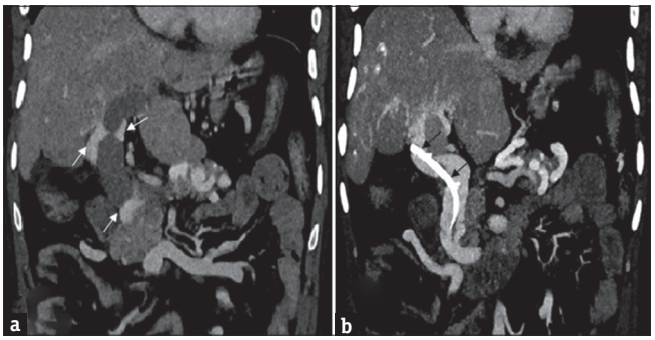

Figure 2: (a and b) contrast-enhanced computed tomography abdomen during portal venous phase in coronal plane showing multiple paracholedochal collaterals (white arrow) with dilated common bile duct. Tip of biliary stent (black arrow) is misplaced and seen within one of the larger paracholedochal collateral

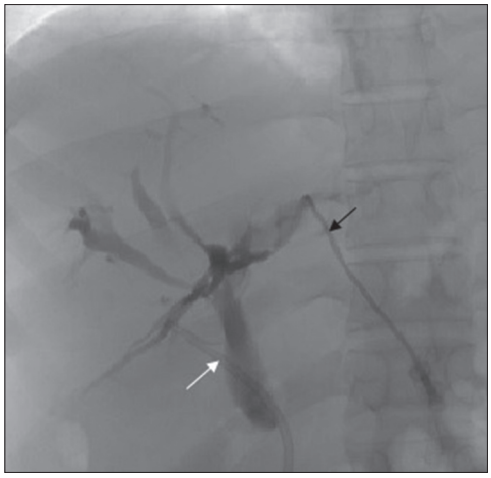

Figure 3: Cholangiogram taken through an external biliary catheter (black arrow) placed from the left side biliary system into the right side showing mild dilatation of intrahepatic biliary radicals and common bile duct. Biliary stent seen within common bile duct in lower part and misplaced out of common bile duct (white arrow) in the upper part

Discussion

Portal biliopathy is reported in 80%–100% cases of EHPVO; however, it manifests with symptoms of biliary obstruction in only 5%–14% of cases.[2] The mechanisms postulated to cause peribiliary plexus are dilated epicholedochal and paracholedochal venous plexuses causing compression on the bile ducts; the formation of new vessels and connective tissue resulting in fibrosis around the ducts and ischemic changes in bile duct.[5] These processes may lead to stasis, stricture, cholangitis, and choledocholithiasis.

Symptomatic biliopathy is a definite indication for intervention–endoscopic or surgical. Endoscopic interventions include sphincterotomy, stone extraction, biliary stenting, stricture dilatation, and mechanical lithotripsy.[6] Portosystemic shunt is done primarily to release pressure on CBD and if symptoms persist, endotherapy is utilized. However, surgical biliary drainage either choledocho-duodenostomy or hepaticojejunostomy may be needed for persistent stricture.[7] In our case, apart from EHPVO, the patient also had splenic vein and superior mesenteric vein thrombosis with no shuntable varix and in view of cholangitis, we had to rely on endoscopic management primarily.

The overall success rate of endotherapy in portal biliopathy was 70%–100%.[2,8] Various complications such as hemobilia and pancreatitis have been reported with ERCP and stenting in portal biliopathy and managed conservatively.[8] The bleeding has been encountered in portal biliopathy while performing ERCP either during sphincterotomy, balloon dilatation, or while removing stent.[3,4] We encountered bleeding after stent insertion which has not been reported before.

Usually, ERCP- and stent-induced bleeding do not cause significant hemodynamic changes and stop spontaneously with conservative management. In this case, the patient developed hemorrhagic shock, managed with inotropes, splanchnic vasoconstrictors, and blood transfusion. In view of cholestatic liver with associated coagulopathy, risk of massive bleeding due to the formation of bilio-portal collateral fistula on CT scan and also risk of bilhemia due to associated obstructive jaundice with high level of jaundice, stent was not removed initially. PTBD was performed and that helped in following ways: decrease the biliary pressure, decrease the level of bilirubin, and improve cholestasis and coagulopathy. Similar approach by endoscopic nasobiliary drainage (ENBD) has been utilized in the past for bridging period of acute crisis.[9] Later, the culprit stent was removed and replaced with another plastic stent.

To the best of our knowledge, this is the first incidence of stent-induced bilioportal collateral fistula leading to massive hemobilia. The mode of injury was different in our case although the culprit lesion was same, i.e., paracholedochal varices. This case highlights various important aspects in managing such patients: (a) utmost care while performing ERC stenting in symptomatic PHB without portosystemic shunt; (b) importance of CT portal venography and endoscopic ultrasound to delineate the biliary plexus collaterals in such patients; (c) biliary drainage with either PTBD or ENBD to improve cholestasis if hemobilia occurs; (d) managing such patients in ICU with multidisciplinary approach.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Sarin SK, Agarwal SR. Extrahepatic portal vein obstruction. Semin Liver Dis 2002;22:43-58.
2. Dhiman RK, Behera A, Chawla YK, Dilawari JB, Suri S. Portal hypertensive biliopathy. Gut 2007;56:1001-8.
3. Al-Akwaa AM, Elsadig M, Al-Fayaa AE, Al-Shehri MD. Portal hypertensive biliopathy presents with massive bleeding during ERCP after balloon sphincteroplasty in a noncirrhotic Saudi sickler patient. Case Rep Med 2017;2017:4163919.
4. Layec S, D'Halluin PN, Pagenault M, Bretagne JF. Massive hemobilia during extraction of a covered self-expandable metal stent in a patient with portal hypertensive biliopathy. Gastrointest Endosc 2009;70:555-6.
5. Dhiman RK, Puri P, Chawla Y, Minz M, Bapuraj JR, Gupta S, *et al.* Biliary changes in extrahepatic portal venous obstruction: Compression by collaterals or ischemic? Gastrointest Endosc 1999;50:646-52.
6. Chandra R, Kapoor D, Tharakan A, Chaudhary A, Sarin SK. Portal biliopathy. J Gastroenterol Hepatol 2001;16:1086-92.
7. Agarwal AK, Sharma D, Singh S, Agarwal S, Girish SP. Portal biliopathy: A study of 39 surgically treated patients. HPB (Oxford) 2011;13:33-9.
8. Oo YH, Olliff S, Haydon G, Thorburn D. Symptomatic portal biliopathy: A single centre experience from the UK. Eur J Gastroenterol Hepatol 2009;21:206-13.
9. Mutignani M, Shah SK, Bruni A, Perri V, Costamagna G. Endoscopic treatment of extrahepatic bile duct strictures in patients with portal biliopathy carries a high risk of haemobilia: Report of 3 cases. Dig Liver Dis 2002;34:587-91.