



Cholangioscopic diagnosis of hemobilia: an unusual case of left hepatic portal hypertension by plasma cell tumor

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Hemobilia refers to potentially life-threatening bleeding, of diverse causes, into the biliary tree. Digital direct cholangioscopy could be a useful tool to identify the source of biliary bleeding in cases in which it cannot be clarified by imaging modalities.

A 68-year-old man with a history of diabetes mellitus was referred to our institution for evaluation of a bulky tumor in the head of the pancreas (Fig. 1A). Abdominal CT revealed a large hyperdense tumor (10.4 cm in diameter) obstructing the distal bile duct, with no evident dilation of the pancreatic duct. ERCP showed distal biliary compression, and a covered self-expandable metallic stent

was inserted for the relief of obstructive jaundice. EUS-guided fine-needle biopsy showed evidence of a malignant tumor (Fig. 1B and C). Histopathologic examination revealed positive immunostaining of the tumor cells for vimentin (+), CD45 (+), MUM-1(+), and CD138 (++) and other markers were negative, which led to the strong suspicion of a plasma cell tumor.

The patient then developed fever and chills and had an episode of cholangitis. He was admitted for evaluation of the cholangitis; abdominal CT showed that the mass, which had increased in size compared to 1 month earlier (Fig. 1D), compressed the horizontal portion of the

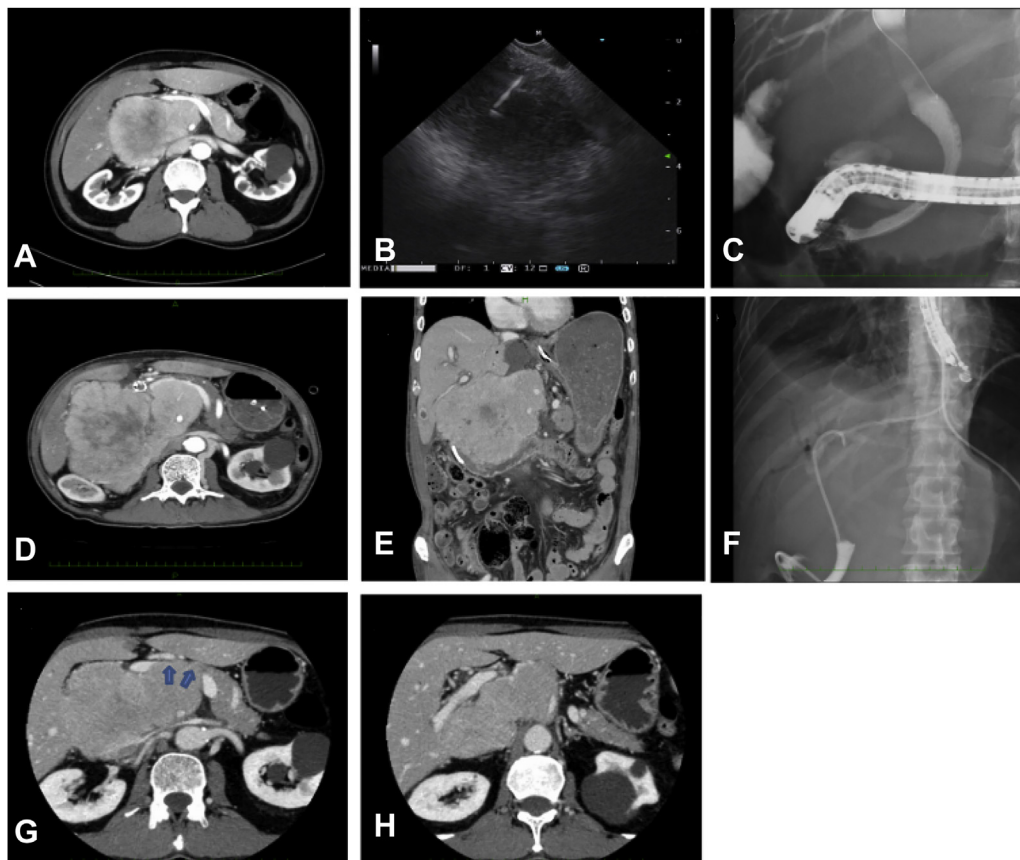


Figure 1. **A**, CT image showing a bulky tumor in the head of the pancreas. **B**, A 22-gauge EUS-FNA needle was used to puncture the bulky tumor in the head of the pancreas. **C**, Radiograph showing endoscopic transpapillary biliary drainage with a covered self-expandable metallic stent. **D**, CT image showing an increase in the size of the mass as compared with a month earlier. **E**, The mass was found to compress the horizontal portion of the duodenum. **F**, Radiograph showing EUS-guided hepaticogastrostomy with a plastic stent. **G**, CT image showing the bulky mass causing focal compression of the base of the left portal vein (*blue arrows*). **H**, Multiple varices formed in the left portal venous system.

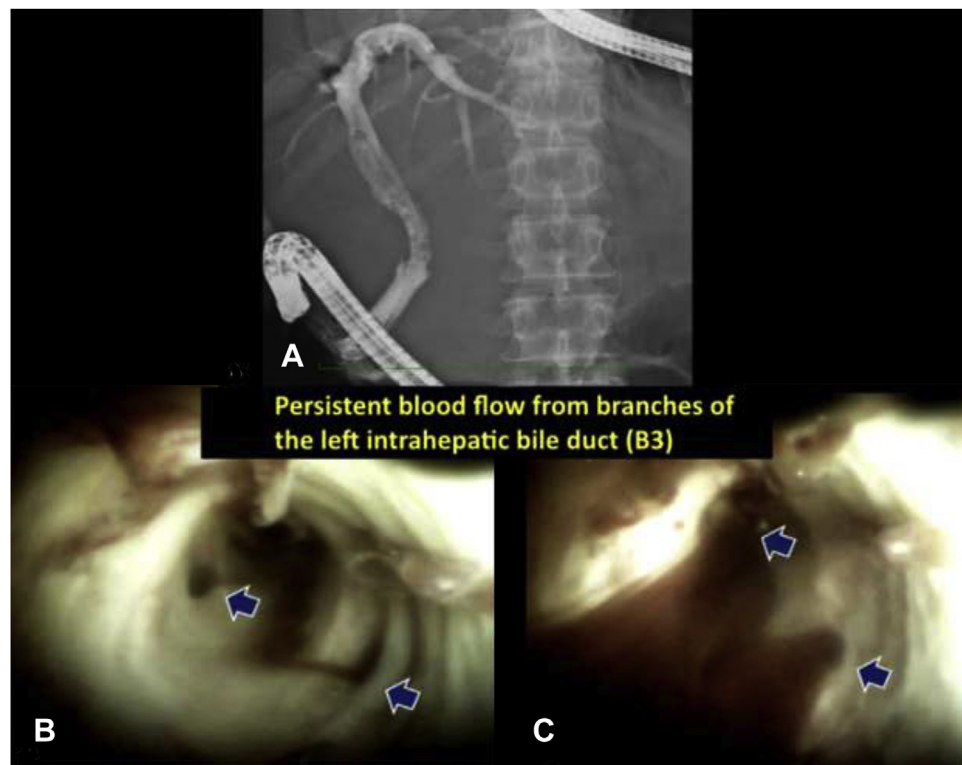


Figure 2. **A**, Radiograph showing the findings of cholangiography performed using a digital cholangioscope (Spyglass). **B** and **C**, Cholangiographic images showing persistent blood flow from branches of the left intrahepatic bile duct (B3) (blue arrows).



Figure 3. **A**, Angiograph showing the bulky mass causing focal compression of the main portal vein. **B**, Angiographic images showing the main portal venous stent placement by interventional radiology.

duodenum, causing duodenobiliary reflux (Fig. 1E). We alternated the biliary drainage route by EUS-guided hepatogastrostomy (HGS) with a plastic stent to facilitate biliary drainage into the stomach and prevent transpapillary duodenobiliary reflux (Fig. 1F).

Three days after the EUS-HGS, the patient had multiple episodes of hematemesis and went into shock. Upper GI endoscopy showed no evidence of any bleeding lesions in the esophagus, stomach, or duodenum. There was a stigma of bleeding from the transpapillary biliary self-

expandable metallic stent, which led to the suspicion of hemobilia. Triple-phase contrast-enhanced CT showed no obvious extravasation or pseudoaneurysm formation in the biliary tract. The tumor mass caused focal compression of the base of the left portal vein, with the formation of multiple varices in the left portal venous system (Fig. 1G and H).

We performed peroral digital cholangioscopy with Spyglass (SpyGlass™ DS System, Boston Scientific) to investigate the source of the hemobilia. We advanced the

cholangioscope into the left biliary duct system and were able to identify persistent blood flow from branches of the left intrahepatic bile duct (B3) (Fig. 2A, B, and C). Cholangiography revealed biliary bleeding from a peripheral branch of B3 and not B2, which had a plastic stent inserted by EUS-HGS (Video 1, available online at www.giejournal.org). Taking this into consideration, we determined that the hemobilia originated from the varices in the left portal venous system. The patient was referred for interventional radiology and underwent main portal venous stenting to reduce the portal venous pressure (Fig. 3A and B). He had no further episodes of bleeding after the procedure. He was discharged soon after and started on chemotherapy for plasma cell tumor.

Hemobilia is defined as bleeding into the biliary tree, varying in severity from mild to life-threatening, depending on the cause of bleeding.^{1,2} Etiologies include biliary tract tumor, cholangiopathy secondary to cirrhosis, prosthesis, or invasive procedures associated with endoscopic biliary interventions.³ Determination of the bleeding source is usually made by arterial- or portal-phase CT findings.

Our patient had a bulky mass compressing the bile duct, duodenum, and portal vein. While we awaited the final diagnosis, the large mass rapidly grew even larger and caused severe compression of the portal vein, as evidenced by sequential CT imaging. Eventually, the marked increase in portal venous pressure resulted in the formation of varices in the left venous portal system, causing persistent hemobilia. We suspected several possible etiologies to explain the hemobilia, from iatrogenic biliary interventions to portal hypertensive cholangiopathy or biliary ulceration from the tumor. Eventually, digital cholangioscopy proved to be of great value to determine the source of bleeding.

Portal biliopathy causes various symptoms of bleeding, cholangitis, and obstructive jaundice.⁴ Therapeutic options are surgical shunt, endoscopic therapy, and interventional

radiology, depending on the pathogenesis.⁵ We have highlighted the first cholangioscopic case to identify hemobilia associated with portal hypertension in the literature. We controlled the persistent hemobilia of unusual causation (portal hypertension resulting from a large plasma cell tumor) with appropriate intervention.

DISCLOSURE

All authors disclosed no financial relationships.

Abbreviation: HGS, hepaticogastronomy.

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