Published online: 2019-09-24

Case Report

# Upper Gastrointestinal Bleed: Do not Forget a Look at Ampulla

*Avinash Bhat Balekuduru, Abhinav Kumar, Athish Shetty, Satyaprakash Bonthala Subbaraj*

Department of Gastroenterology, Ramaiah Memorial Hospitals, Bengaluru, Karnataka, India

**Abstract**

Hemosuccus pancreaticus (HP) and hemobilia (HB) are uncommon causes of upper gastrointestinal bleed. In this report, 4 cases of HP and 1 case of HB with intermittent bleeding are described. The causes of HP are rupture of splenic artery pseudoaneurysm in 3 and gastroduodenal artery aneurysm in one. The cause of HB is due to cystic artery bleed. HP can be secondary to chronic or acute pancreatitis with bleeding from pseudoaneurysm arising from peripancreatic arteries. Iatrogenic transhepatic techniques, trauma, operative injuries, malignancy, and inflammation in hepatobiliary system are the common causes of HB. All the cases are missed on first endoscopy and diagnosed on second-look endoscopy. A strong clinical suspicion is required at first endoscopy for early diagnosis and management. This case report compares presentation of HP with HB, diagnosis, and their management.

**Keywords:** *Hemobilia, hemosuccus pancreaticus, pseudoaneurysm*

## Introduction

Hemosuccus pancreaticus (HP) or wirsungorrhagia or pseudohemobilia is first described by 1931 by Lower and Farrel and the name HP is given by Sandblom in 1970 in which blood loss is due to communication of visceral artery with pancreatic duct.[1] Francis Glisson in 1654 first described and Philip Sandblom in 1948 first used the term hemobilia (HB) for hemorrhage into the biliary tree.[2] HP and HB are the two causes of bleeding from duodenal ampulla. The source of bleeding is usually apparent from the clinical context. Interventional radiology to identify the fistula between the duct and the bleeding source, with therapeutic embolism, is the mainstay of treatment.[3] HP and HB are intermittent upper gastrointestinal bleeds which can present as occult blood loss to massive bleed.[4] A strong clinical suspicion will avoid delay in diagnosis and improvement in outcome.

## Case Reports

### Case 1

A 53-year-old man with significant consumption of alcohol was admitted owing to epigastric pain and intermittent hematemesis for 2 months' duration. Laboratory workup revealed hemoglobin level of 9.1 g/dL (normal range: 12–14 g/dL) and normal amylase and lipase levels. His ultrasound abdomen done at an outside hospital revealed a mass in the body of pancreas and was referred for evaluation of the same. Endoscopic ultrasound (EUS) was performed which showed a 7.2 cm × 6.5 cm lesion adjacent to the body of the pancreas with outer hypoechoic wall consistent with aneurysm and a central anechoic area [Figure 1a] which showed flow on color Doppler evaluation, and a diagnosis of giant splenic artery pseudoaneurysm (SAP) was made [Figure 1b]. The surrounding pancreatic parenchyma was edematous, and minimal fluid collection was noted. A contrast-enhanced abdominal computed tomography (CECT) scan followed by successful angiography and coil embolization was performed and remained asymptomatic at 1-year follow-up.

### Cases 2–4

Three cases of ethanol-related chronic pancreatitis with pseudocyst presented with abdominal pain and intermittent transfusion requiring gastrointestinal bleeding. Two cases of SAP and in one gastroduodenal

Access this article online

Quick Response Code:

Website: www.jdeonline.in

DOI: 10.4103/jde.JDE_25_18

***Address for correspondence:*** Avinash Bhat Balekuduru, Department of Gastroenterology, Ramaiah Memorial Hospitals, Bengaluru - 560 054, Karnataka, India.
E-mail: avinashbalekuduru@gmail.com



**How to cite this article:** Balekuduru AB, Kumar A, Shetty A, Subbaraj SB. Upper gastrointestinal bleed: Do not forget a look at ampulla. J Dig Endosc 2018;9:134-7.

artery pseudo aneurysm were noted. All underwent successful angiography and coil embolization followed by cystogastrostomy [Figures 2 and 3]. There was a delay in diagnosis by 2 days as the initial endoscopic evaluation was negative. Second-look endoscopy confirmed bleeding from the ampulla and CT angiography identified the culprit vessel which was then embolized by conventional angiography.

### Case 5

A 67-year-old male presented with unstable angina and abdominal pain. He underwent emergency angiogram and stenting of left main coronary artery. On the 3rd poststenting day, he had hematemesis, fever, worsening of abdominal pain, and jaundice. His total bilirubin was 3.7 mg/dL with direct being 3.02 mg/dL. Aspartate aminotransferase, alanine aminotransferase, alkaline phosphatase was 160, 125, and 587 IU/mL, respectively. Emergency CECT was performed which revealed perforated gallbladder and dilated common bile duct with its lumen filled with heterogeneous density contents without contrast enhancement [Figure 4]. Endoscopic retrograde cholangiopancreaticography (ERCP) was done which confirmed the diagnosis of HB and adequate clearance of bile duct, and stenting was performed [Figure 5]. HB settled and after 6 weeks underwent cholecystectomy without any recurrence of HB. The diagnosis of hemorrhagic cholecystitis was made as gallbladder revealed multiple hemorrhagic erosions on the mucosal side. Cholecystolithiasis and anticoagulant /antiplatelet agents might be the causation for HB in this case.

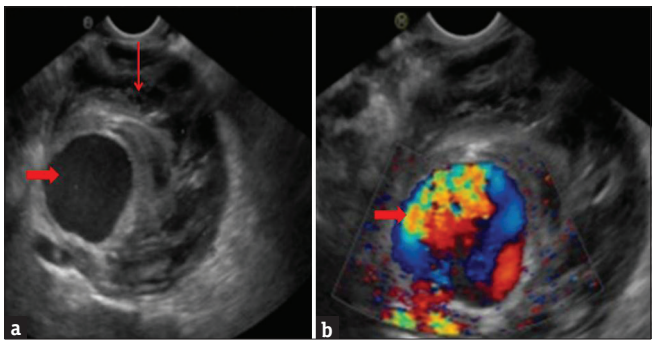


**Figure 1:** (a) Linear endoscopic ultrasound image (with transducer in stomach) showing outer hypoechoic and central anechoic area and (b) color Doppler image showing vascular fl ow in anechoic area. Response: part labelled. Response: : (a) Linear endoscopic ultrasound image (with transducer in stomach) showing outer hypoechoic (plain arrow) and central anechoic area (block arrow) and (b) color Doppler image showing vascular fl ow in anechoic area (block arrow)

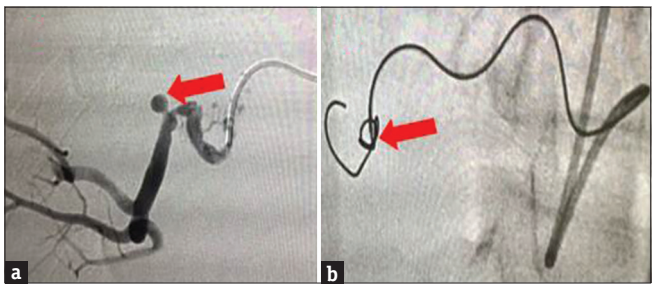


**Figure 3:** (a) Fluoroscopic image showing splenic pseudoaneurysms aneurysm (block arrow) and (b) showing coil embolization ( block arrow)

## Discussion

It is important to know about the two rare causes of extra luminal source of gastrointestinal bleeding into the duodenum which can be missed easily and

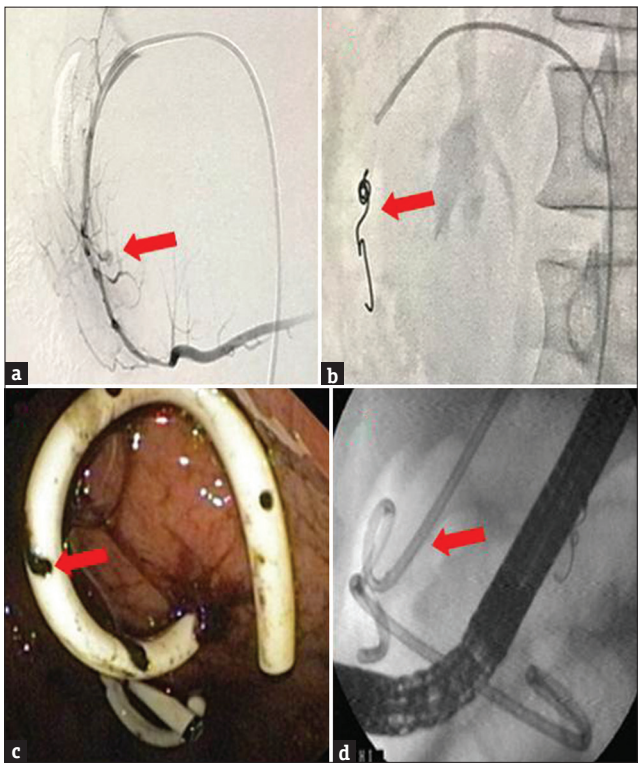


**Figure 2:** (a) Fluoroscopic image showing gastroduodenal artery aneurysm (block arrow) and (b) showing coil embolization of gastroduodenal artery (block arrow) (c) Endoscopic image showing 2 cystogastrostomy stents draining fl uid stained with blood (block arrow) and (d) fl uoroscopic image of the same (block arrow)

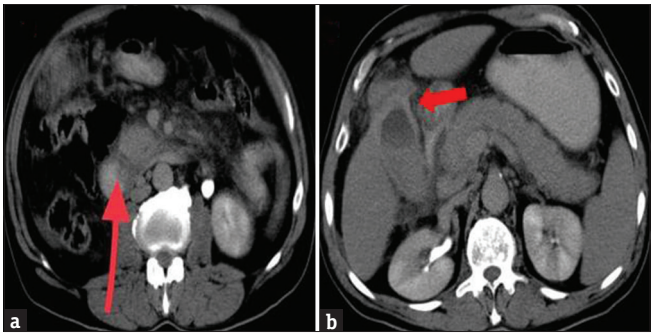


**Figure 4:** (a) Contrast-enhanced abdominal computed tomography image showing dilated common bile duct with intraluminal fi lling defects (plain arrow) and (b) computed tomography image showing perforated gallbladder and pericholecystic collection (block arrow)

**Table 1: Comparison of both the causes of bleeding from ampulla**

| | | |
|---|---|---|
| Bleeding from ampulla via | Pancreatic Duct → Hemosuccus Pancreaticus[6] | Bile Duct/gall bladder → Hemobilia[5] |
| Predisposition | Pancreatic diseases (80% of all cases)[7] | Iatrogenic (70%) |
| | Chronic pancreatitis/Pseudocyst communicating with pericyst artery → pseudoaneurysm | 14% percutaneous biliary drainage |
| | Intraductal stone or dilatation → vascular ulceration | 4% percutaneous |
| | Neuroendocrine tumor/ductal adenocarcinoma of pancreas | Transhepatic cholangiography, needle liver biopsy 1%, ERCP, 40-85% - hepatobiliary surgeries |
| | Acute pancreatitis | Others - gallstones, malignancy, inflammatory diseases, ascaris, and vascular etiologies |
| | Ectopic pancreas | Blunt or penetrating trauma |
| | Pancreas divisum | |
| | Post-ERCP | |
| | Vascular anomaly in visceral arteries (20%) | |
| | True aneurysm of visceral arteries | |
| | Pseudoaneurysm of visceral arteries (nonpancreatic origin) | |
| | Dissection of visceral arteries | |
| Culprit arteries involved | Splenic 40% | Right hepatic artery - 80% 8 hepatic, cystic, gastroduodenal, and supra- and infra-pancreatic arteries |
| | Gastroduodenal 30% pancreaticoduodenal 20% gastric 5% and hepatic arteries 2% | |
| Pathophysiology | Pancreatic enzymes → necrosis/skeletonisation of the peripancreatic vessels → pseudoaneurysm | Bile → damages blood vessels → delayed healing of an injured artery → pseudoaneurysm.[8] |
| | Scarring and granulation on the pseudocyst → traction on the vessel → GI hemorrhage | |
| Clinical features | Intermittent abdominal pain and gastrointestinal hemorrhage with hyperamylasemia | Quinckes triad (22%)[8] |
| | | Biliary colic-70%/Melena-90%/ Hematemesis-60%/Jaundice- 60% |
| Diagnosis | Endoscopy- 30%- Intermittent bleed- spend time at ampulla | Endoscopy 12%- first test |
| | CECT - roadmap for therapy | CECT - possible etiology |
| | Angiography - Gold standard for diagnosis and treatment | ERCP- in significant hemobilia and ascending cholangitis |
| | | Angiography - most accurate- 90% |
| | | MRCP- differentiates- clot and stone[8] |
| | | Technetium red cell scan |
| Treatment | Radiological: prosthetic material, balloon tamponade and stent placement in case if risk of distal infarction. | Transcatheter arterial embolization of hepatic artery: (first approach). Surgery if TAE fails; bile duct tumor, hemorrhagic cholecystitis |
| | Surgical: Uncontrolled bleeding, persistent shock, failure/rebleed after embolization → distal pancreatectomy and splenectomy, central | Endoscopic management is the preferred initial strategy for decompression of the biliary tree |
| | pancreatectomy, intracystic ligation of the blood vessel, aneurysm ligation and bypass graft | |

can be life threatening. Table 1 compares the clinical characteristics, diagnosis, and management of these two different conditions which present with bleeding from ampulla.

SAPs are rare with fewer than 200 cases reported in the English literature. In true aneurysm, wall is composed of intima, media, and adventitia, but in pseudoaneurysm, wall contains only intima and media. Pancreatitis is the most common cause for SAP secondary to autodigestion of splenic artery wall with pancreatic enzymes.[9] The other causes of SAP are abdominal trauma, postoperative/iatrogenic, and rarely peptic ulcer disease. All the four cases were of ethanol-related chronic pancreatitis complicated with peripancreatic pseudocyst. Abdominal pain, hematemesis, and melena are the common presenting features. Risk of rupture can be as high as 37%, with a mortality rate approaching 90% when untreated. Transarterial embolization in hemodynamically stable and pancreatectomy with splenectomy in unstable patients are standard treatment modalities.[10] EUS is emerging as a diagnostic modality for SAP.

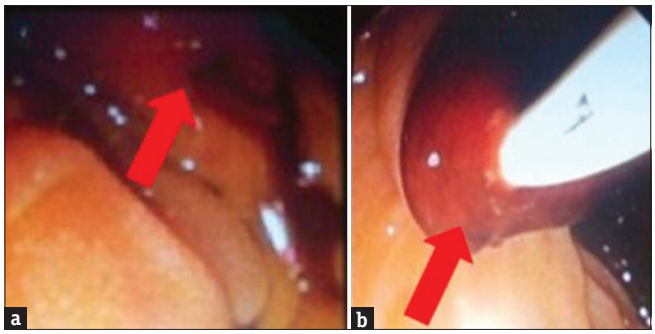


**Figure 5:** (a) Duodenoscopic image showing blood coming from ampulla (Block arrow) and (b) showing balloon sweep retrieving the blood clot from common bile duct (block arrow)

The goal of angioembolization in HB is always to preserve as much hepatic artery as possible. HB patients present with concomitant biliary obstruction from intraluminal bleeding and clot. ERCP is the preferred initial strategy for decompression of the biliary tree.[5]

All the five patients survived the gastrointestinal bleeding episodes, but there was a delay in diagnosis which could have been avoided with strong clinical suspicion.

## Conclusions

HB secondary to biliary or operative intervention and HP due to complication of acute or chronic pancreatitis are rare causes of gastrointestinal bleeding. CT angiography as the initial diagnostic evaluation followed by angiography with embolization in a hemodynamically stable or surgery in a hemodynamically unstable patient is the standard diagnostic and treatment approach in patients with clinical HB/HP.

### 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.

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