

Pseudoaneurysm of the Right Hepatic Artery - A Rare Cause of Obscure GI Bleeding

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ABSTRACT

A 63 years old female with 1 month history of hematemesis and malena, severe weakness and lethargy, for which she had upper GI endoscopies multiple times, but the source of bleeding could not be identified. Base line investigations revealed Hb of 6.0g/dl. CT scan abdomen showed cholelithiasis with air inside the gallbladder. The selective celiac axis / Hepatic artery angiogram revealed a right hepatic artery aneurysm (pseudoaneurysm). After resuscitation with blood transfusion and fluids, the patient underwent surgical exploration, revealing an aberrant right hepatic artery aneurysm bleeding inside the gall bladder with a cholecystoduodenal fistula (Mirrizi type III) into the 1st part of the duodenum from where the blood was leaking into the gastrointestinal tract and causing severe hematemesis and malena. A cholecystectomy, dissection of sleeve of liver bed, ligation of the aneurysmal bleeding vessel, repair of the cholecystoduodenal fistula and placement of the T-Tube done. Post operative the patient remained stable and was discharged on 7th post operative day. Biopsy revealed acute on chronic cholecystitis and cholelithiasis. Biopsy of the aneurysmal wall revealed inflamed granulation tissue.

Key Words: hepatic artery aneurysm, cholecystoduodenal fistula, obscure GI bleeding, Mirrizi syndrome.

INTRODUCTION

This condition is rare but may present as a diagnostic challenge. It accounts for 20% of the entire splanchnic artery aneurysms. Most commonly associated are common hepatic artery proximal to gastroduodenal artery, in the hepatic artery distal to the gastroduodenal artery, in the right and left hepatic arteries or in the hepatic parenchyma in order of frequency¹. Etiological factors include atheroma, infection (bacterial endocarditis, fungal infections) trauma, radiological interventions, needle biopsies and biliary surgery (cholecystectomy etc)^{1,2}. Rupture may occur in the peritoneal cavity, in the bile duct, in the portal vein or in the duodenum (as in this case) leading to massive upper GI bleeding.

Most of the hepatic artery aneurysms are seen during middle age and are more common in males than females with a ratio of 2:1 whereas some

aneurysms are asymptomatic and are discovered on imaging tests done for other disorders. The patient may present with non specific right upper quadrant pain usually diagnosed by ultrasound and confirmed by CT angiography. It can also present as biliary colic, jaundice and evidence of upper gastrointestinal hemorrhage with sudden collapse and peritonitis.

Even asymptomatic hepatic artery aneurysms should be treated because of the risks of rupture¹⁻³. Ideally they should be managed by percutaneous embolization but sometimes technical difficulties make open surgery inevitable.

If the aneurysms arise from the common hepatic artery, isolation with exclusion or excision and ligation is safe provided the liver function is normal, otherwise, an aorto-hepatic bypass is needed in addition to the excision. Percentage of patients who present with intra peritoneal rupture carries a grave prognosis with a mortality

approaching approximately 100%. However, most patients with aneurysmal rupture into the biliary tract are salvageable after achieving control³.

CASE REPORT

A 63 years old female with no known comorbidities, admitted with 1 month history of intermittent hematemesis and malena, exacerbated over the past few days with 4-5 episodes of vomiting (hematemesis) accounting to approximately 100 ml each, with passage of tarry stool.

On examination the patient had a pulse rate of 110/min, regular with a systolic blood pressure of 100 mmHg and postural drop of >10 mmHg. R/Rate was 25/mm with 36.6°C temperature.

Clinically the patient looked pale and dehydrated. No jaundice or cyanosis. Chest was clear to auscultation with no adventitious sounds. Abdomen was tender in right upper quadrant, no visceromegaly and gut sounds were audible. There was no focal neurological deficit.

The investigations done at that time included.

CBC:

Hb:	6 gm/dl
TLC:	11500 with neutrophils of 90%
Platelets:	348,000
PT:	13
APTT	29
INR	1.0

LFT's:

ALP:	453
ALT:	83
AST:	94
T. Bill:	0.78

RFT's:

Bun:	11
Creatinine:	0.8

SIE:

Na+:	144
K+:	3.4

Hepatitis Screening:

HBsAg	Negative
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Anti HCV Negative

USG Abdomen:

- Done at that time was normal.
- Shows contracted gall bladder

GI Endoscopy

A series of upper GI Endoscopies were done (total 4 times) without identifying the source of GI bleed.

CT Enteroclysis

On initial CT Enteroclysis done it was reported that the gall bladder had diffuse wall thickening and multiple large calculi compressing upon the adjacent 1st part of duodenum with hazy surrounding fat planes. Some air was also noted in the gall bladder. A patent portal vein and minimal prominence of intra hepatic biliary channels. No ascities or lymphadenopathy. Pancreas, spleen, kidneys, adrenals and bladder were unremarkable.

On the basis of above findings a repeat CT Abdomen / CT hepatic artery angiogram was done which showed a distended gall bladder containing multiple calculi and air with significant wall thickening, compressing upon the CBD in supra pancreatic part and causing mild intrahepatic biliary dilation. Wall of the first part of the duodenum, the celiac trunk, SMA, common hepatic artery and gastroduodenal arteries were normal. The right hepatic artery was tortuous and small in caliber passing in closed proximity between fundus of gall bladder and adjoining part of the duodenum showing a 5 mm dilatation at the corresponding level - pseudoaneurysm??).

After a review of relevant investigations a decision to rescope the patient was made which showed the stomach full of blood which was sucked out and a gush of fresh blood from the duodenum without localization of the source. So an emergency exploration was done with an inverted L-Shaped incision in right hypochondrium. The intra operative findings included.

- A thick walled distended gall bladder with multiple large stones 5-6 in numbers largest 3cm in diameter.
- Gall bladder densely adherent to the liver, omentum, duodenum and transverse colon.

- A cholecystoduodenal fistula into the first part of the duodenum and CBD (Mirizzi type III).
- An aberrant Right Hepatic artery forming a 5mm aneurysm inside the gall bladder and actively bleeding inside the gall bladder.

After acknowledging the above findings, a cholecystectomy and dissecting a sleeve of liver bed along with repair of the duodenal fistula, ligation of the bleeding pseudoaneurysm and CBD exploration plus placement of T-Tube was done.

The specimen was sent for histopathology the patient was reversed and shifted to recovery in stable condition and after 24 hrs stay in ICU (intensive care unit) the patient was shifted to the general ward from where she was discharged on her 7th post operative day along with T-Tube in situ.

On her follow up visit as an out patient a T-Tube cholangiogram was done which was normal and was removed after clamping. Her post operative investigations included complete blood counts, Liver function tests, renal function test and serum electrolytes, were all unremarkable. The biopsy report turned out to be acute on chronic cholecystitis with cholelithiasis, wedge resection specimen of liver revealed mild to moderate triditis. Wall of the vessel and aneurysm revealed fragments of blood clot, fibrin necrotic tissue and inflamed granulation tissue.

DISCUSSION

Aneurysm of the hepatic artery contribute approximately 20% of all visceral aneurysm out of these 80% of HAA are extra-hepatic and 20% are Intrahepatic. In 63% of the cases the Left hepatic is involved while the anomalies of Right Hepatic artery and left hepatic artery is 28% and 5% respectively in 4% of the cases both hepatic artery¹⁻⁴.

True aneurysms are either segmental or diffuse dilation of arteries. On the other hand pseudoaneurysms are dilatations which don't involve all the three layers. True aneurysm occurs more frequently (4 times) in extra hepatic artery and involves the common hepatic artery and is associated with atherosclerosis⁶⁻⁸.

Pseudoaneurysm arises as a consequence of inflammation adjacent to the arterial wall damaging the adventitia. This causes thrombosis of the vasa vasorum leading to weakness of the vessel wall.

Hepatic artery aneurysm also results from abdominal trauma, transhepatic procedures (TIPS), pancreatitis, liver transplantation, percutaneous biopsies, choledochal cysts and cholecystectomy^{5,9-12}. Most patients of hepatic artery aneurysm are asymptomatic while some presents with right upper quadrant pain, jaundice or symptoms associated with rupture. Risk of rupture of hepatic artery into the adjacent viscous (stomach, duodenum and colon) peritoneal, retroperitoneal area or biliary tract leads to GI hemorrhage, hypovolumic shock, biliary colic, hemobilia or obstructive jaundice^{1,2,14-17}. In this case the rupture of an aneurysm in the aberrant right hepatic artery into the gall bladder which was forming a cholecystoduodenal fistula led to massive GI bleed (Mirizzi type). This is very rare presentation of erosion and rupture of hepatic artery aneurysm presenting as massive GI bleed²¹⁻²².

Hepatic artery aneurysms are better depicted by spiral or multisecton CT as it can be scanned during peak contrast enhancement¹⁹⁻²⁰.

An angiographic study may demonstrate the site of active bleeding and the sac of pseudoaneurysm. Once an aneurysm is identified, treatment is indicated regardless of bleeding¹⁴. Percutaneous embolization is the gold standard for intrahepatic artery aneurysm having a success rate of more than 80%²³⁻²⁶.

In the present case the initial CT Scan done did not reveal any aneurysm or fistula formation. However, subsequent CT angiography following a massive episode of GI bleeding revealed an aneurysm.

Although primary surgery has largely been replaced by selective angiographic embolization of the intrahepatic artery aneurysm. Surgery is still reserved for extrahepatic artery aneurysms of greater than 2 cm and embolization failure^{2, 4, 13,25, 27}. The surgery was undertaken in this case because of the patient developing massive GI bleeding, requiring emergency exploration.

Surgical ligation carries a complication rate of 20-29% and a mortality rate of 50% in hemodynamically unstable patients^{27,28}. The surgical

approach involving ligation for hepatic artery distal to celiac trunk in patients with delayed hemorrhage is preferred as this will preserve hepatic circulation and ensure homeostasis.

Proximal control of hepatic artery should be attempted first but in the present case due to easy access of the aneurysm this was not required. Post operatively the patient should be closely monitored with liver function test and serial ultrasounds to check the liver perfusion.

CONCLUSION

Right Hepatic artery aneurysm bleeding into the gall bladder having a cholecystoduodenal fistula and causing gastrointestinal bleeding is a rare cause of massive upper GI Bleeding.

Since the bleeding vessel is obscured from the GI tract, it cannot be picked up with endoscopies which was the case in our patient, in which multiple endoscopies did not reveal the bleeding point.

Radiological contrast studies (Angiogram) are ideal investigations for obscure GI bleed, which also offers the advantage of therapeutic angioablation/embolization, where the expertise are available.

Surgical intervention is indicated when all the other modalities (Endoscopy, angioablation) fail to control the source of bleeding or there is immediate life threatening haemorrhage.

REFERENCES

1. Okuno A, Miyazaki M, Ito H, Ambiru S., Yoshidome H, Shimizu H, Nakagawa K, Shimizu Y, Nukui Y, Nakajima N. Non-surgical management of ruptured pseudoaneurysm in patients with hepatobiliary pancreatic diseases. *Am J Gastroenterol*: 2001; 96: 1067-71
2. Pasha SF, Gloviczki P, Stanson AW, Kamath PS. Splanchnic artery aneurysms. *Mayo Clin Proc*: 2007; 82:472-79
3. Green MH, Duell RM, Johnson CD, Jamieson NV: Haemobilia. *Br J Surg*: 2001; 88: 773-86.
4. Abbas MA, Fowl RJ, Stone WM, Panneton JM, Oldenburg WA, Bower TC. Hepatic artery aneurysm: factors that predict complications. *J Vasc Surg*: 2003; 38:41-5.
5. Enns R, Schmidt N, Harrison P, Chipperfield P, Skarsgaard P, Brown JA. Endoscopic Diagnosis of a right hepatic artery pseudoaneurysm. *Endoscopy*, 2002;34:337-40.
6. Kibbler CC, Cohen DL, Cruickshank JK, Kushwaha SS, Morgan MY, Dick RD. Use of CAT scanning in the diagnosis and management of hepatic artery aneurysm. *Gut*; 1985;26:752-56.
7. Shanley CJ, Shah NL, Messina LM. Common splanchnic artery aneurysms: splenic, hepatic, and celiac. *Ann Vasc Surg*; 1996;10:315-22.
8. Stanley JC, Wakefield TW, Graham LM, Whitehouse WM Jr, Zelenock GB, Lindenauer SM. Clinical importance and management of splanchnic artery aneurysms. *J Vasc Surg*: 1986;3:836-40.
9. Poon RT, Tuen H, Yeung C. GI hemorrhage from fistula between right hepatic artery pseudoaneurysm and the duodenum secondary to acute cholecystitis. *Gastrointest Endosc*: 2000; 51:491-93.
10. Finley DS, Hinojosa MW, Paya M, Imagawa DK. Hepatic artery pseudoaneurysm: A report of seven cases and a review of the literature. *Surg Today*; 2005, 35:543-47.
11. Saluja SS, Ray S, Gulati MS, Pal S, Sahni P, Chattopadhyay TK. Acute cholecystitis with massive upper gastrointestinal bleed: a case report and review of the literature. *BMC Gastroenterol*: 2007; 7:12.
12. Briceño J, Naranjo A, Ciria R, Sánchez-Hidalgo JM, Zurera L, López-Cillero P. Late hepatic artery pseudoaneurysm: a rare complication after resection of hilar cholangiocarcinoma. *World J Gastroenterol*; 2008; 14:5920-3.
13. Dolapci M, Ersoz S, Kama NA. Hepatic Artery Aneurysm. *Ann Vasc Surg*: 2003;17: 214-16.
14. Reber PU, Baer HU, Patel AG, Wildi S, Triller J, Büchler MW. Superselective microcoil embolization: treatment of choice in high-risk patients with extrahepatic pseudoaneurysms of the hepatic arteries. *J*

- Am Coil Surg: 1998; 186:325-30.
15. Clay RP, Farnell MB, Farnell. Lancaster JR. Hemosuccus pancreaticus. Ann Surg; I 1985;202:75-9.
16. Wagner WH, Allins AD, Treiman RL, Cohen JL, Foran RF, Levin PM, Cossman DV. Ruptured visceral artery aneurysms. Ann Vasc Surg; 1997;11:342-47.
17. Zachary K, Geier S, Pellicchia C, Irwin G. Jaundice secondary to hepatic artery aneurysm: radiological appearance and clinical features. Am J Gastroenterol: 1986;81:295-98.
18. Gopanallikar A, Rathi P, Sawant P, Gupta R, Dhadphale S, Deshmukh HL. Hepatic artery pseudoaneurysm associated with amebic liver abscess presenting as upper GI hemorrhage. Am J Gastroenterol: 1997; 92: 1391-93.
19. Hsieh CH. Comparison of hepatic abscess after operative and nonoperative management of isolated blunt liver trauma. MT Surg: 2002; 87: 178-84.
20. Szentkereszty Z, Peter M, Erdelyi G, Sapy P: Results of surgical treatment of liver abscess, with special emphasis of percutaneous and drainage. Magy Seb: 2000; 53: 259-62.
21. Balthazar EJ: "Hemobilia: Calcified hepatic artery aneurysm presenting with massive gastrointestinal bleeding. Gastrointest Radiol; 1977; 2:71-4.
22. Narula HS, Kotru A, Nejjim A. Hepatic artery aneurysm: an unusual cause for gastrointestinal haemorrhage. Emerg Med J; 2005;22:302.
23. Romano L, Giovine S, Guidi G, Tortora G, Cinque T, Romano S. Hepatic trauma: CT findings and considerations based on our experience in emergency diagnostic imaging. Eur J Radiol: 2004; 50: 59-66.
24. Jaeckle T, Stuber G, Hoffmann ME, Jeltsch M, Schmitz BL, Aschoff AJ. Detection and localization of acute upper and lower gastrointestinal (GI) bleeding with arterial phase multi-detector row helical CT. Eur Radiol: 2008; 18:1406-13.
25. Sasaki K, Ueda K, Nishiyama A, Yoshida K, Sako A, Sato M, Okumura M. Successful utilization of coronary covered stents to treat a common hepatic artery pseudoaneurysm secondary to pancreatic fistula after Whipple's procedure: report of a case. Surg Today; 2009; 39:68-71.
26. Kim SY, Kim KW, Kim MJ, Shin YM. Lee MG, Lee SG. Multidetector row CT of various hepatic artery complications after living donor liver transplantation. Abdom Imaging; 2007; 32:635-43.
27. Moore E, Matthews MR, Minion DJ, Quick R, Schwarcz TH, Loh FK, Endean ED. Surgical management of peripancreatic arterial aneurysms. J Vasc Surg; 2004; 40:247-53.
28. Bender JS, Bouwman DL, Levison MA. Pseudocysts and pseudoaneurysms: Surgical Strategy. Pancreas; 1995; 10:143-47.

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