

# Non-Traumatic Right Hepatic Artery Pseudoaneurysm: An Unusual Cause of Hemobilia and Obstructive Jaundice

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## ABSTRACT

Most hepatic artery pseudoaneurysms (HAPA) are post traumatic, and non-traumatic pseudoaneurysm is rarely reported. It is a potentially life threatening vascular disorder and difficult to diagnose before rupture. Early diagnosis and prompt nonoperative intervention of this lesion could be life saving. The authors report the case of a patient with hemobilia caused by ruptured right hepatic artery pseudoaneurysm and subsequently developed right hepatic duct stricture, which has not been reported previously. This patient was successfully treated with endovascular stent graft of pseudoaneurysm and endoscopic stenting of right hepatic duct stricture.

**Key words:** Pseudoaneurysm. Hemobilia. Endovascular stenting. Endoscopic stenting.

## INTRODUCTION

Hepatic artery pseudoaneurysm (HAPA) is an unusual but potentially life threatening vascular entity, mainly caused by acute or chronic hepatic artery injuries. There are many causes of HAPA. It usually follows blunt or penetrating injuries and interventional radiological procedures.<sup>1-3</sup> Nontraumatic pseudoaneurysm of hepatic artery is reported very rarely, caused by arteriosclerosis, polyarteritis nodosa, acute pancreatitis, cholecystitis, bacterial endocarditis and liver abscess,<sup>4-7</sup> Most of the patients present with hemobilia, after rupture of HAPA and very rarely, a patient presents with obstructive jaundice due to compression on hepatic duct.<sup>3,8</sup>

The authors report the case of a patient with hemobilia caused by ruptured right hepatic artery pseudoaneurysm who subsequently developed right hepatic duct stricture, which has not been reported previously.

## CASE REPORT

A 20-year-old woman was in her usual state of health with no known comorbid medical conditions, when she developed initial symptoms of high grade fever for two weeks, yellow discoloration of sclera for ten days, passing black stool for 6 days, abdominal pain and two episodes of vomiting fresh blood. There was no history of trauma. The rest of her history was unremarkable, except that she was empirically treated for enteric fever

by the family physician with Ciprofloxine 500 mg twice a day orally for 7 days. She was brought to the emergency room of this hospital because of her increasing fever, jaundice and dizziness.

Her blood pressure was 88/48 mm of Hg, pulse rate was 120/minute and she had a core temperature of 38.6°C. Clinically, she was jaundiced and systemic examinations were unremarkable except that her digital rectal examination was positive for malenotic stool. She became stable hemodynamically after fluid resuscitation with 2000 cc of normal saline 0.09%.

Laboratory investigations performed in the emergency room showed: hemoglobin of 5.7 gm/dl (11-14.5) with haematocrit of 16.45 (35.4-42), white cell count of 17.9 x 10E9/L (4.0-10.0), platelets of 594 x 10E9/L (150-400) and normal coagulation profiles with I.N.R of 1.09. Her liver function tests showed serum total bilirubin of 9.8 mg/dl (0.25-1.0) with 6.0 mg/dl, alkaline phosphatase of 225 IU/L, LDH of 799 IU/L and hepatocellular enzymes were within normal limits. Upper gastrointestinal endoscopy revealed hemobilia and subsequent angiography confirmed right hepatic artery pseudoaneurysm of size 4 cm in transverse diameter with narrow neck.

Later on, using a 9fr guided catheter, a 6 x 28 mm size balloon mounted, covered metallic stent graft was placed leading to complete exclusion of pseudoaneurysm. She received 4 units of packed cells in the postangiography period and then continued on broad spectrum intravenous antibiotics. A CT scan was performed before discharge from the hospital to look for the cause of pseudoaneurysm, which was normal except mild biliary dilatation and clots in gall bladder and common bile duct. She was discharged on the 7th day of admission and laboratory investigations showed hemoglobin of 12.3 gm/dl, white cell count of 10.9 x 10E9/L and improving liver function tests at that time.

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She required readmission after 48 hours of discharge. This time she was febrile with core temperature of 38.8°C, dehydrated, deeply jaundiced and tender on deep palpation in epigastrium and right hypochondrium. Laboratory investigations showed hemoglobin of 11.2 gm/dl, white cell count of 17.4 x 10E9/L with 90% neutrophils, total bilirubin of 8.8 mg/dl with direct component of 6.3 mg/dl and alkaline phosphatase of 489 IU/L. Ultrasonogram showed dilated intrahepatic biliary system and subsequent endoscopic retrograde cholangio-pancreaticography (ERCP) revealed stricture in right hepatic duct close to the site of pseudoaneurysm. Balloon dilation of stricture with plastic biliary stenting was done and her post ERCP period was uneventful. Patient was followed in the clinic with serial liver function tests, which showed total bilirubin of 0.4 mg/dl, alkaline phosphatase was 96 IU/L and the biliary stent was removed on 6th week.

### DISCUSSION

Hepatic artery pseudoaneurysm is a rare and potentially life threatening vascular entity, with reported mortality rate of 50-75% or more, in patients requiring surgery for ruptured aneurysm.<sup>2,5</sup> Upto 60% of HAPA are clinically silent and found incidentally and even symptomatic patients can have a non-specific presentation requiring a high index of suspicion.<sup>5,6</sup> Forty percent of HAPA will present with rupture. Sandblom was the first to apply the term "hemobilia" to the syndrome of gastrointestinal bleeding of biliary tract origin and the classical triad of abdominal pain, jaundice and upper gastrointestinal bleeding.<sup>6,7</sup> The patient we reported had fever followed by jaundice and abdominal pain. Upper gastrointestinal endoscopy demonstrated blood coming out of duodenal papilla of Vater. Many cases of hemobilia are reported to be treated for unexplained upper gastrointestinal bleeding, and treatment tends to be delayed. Therefore, hemobilia should be considered in the differential diagnosis when other sources are excluded.

Historically, mycotic aneurysms were the most common cause of HAPA, but in current literature they account for only 4% and the major underlying pathology is infected endocarditis.<sup>3,8-10</sup> Most of the mycotic HAPA will present with fever and ruptured aneurysm and there was only one case report of mycotic aneurysm, who presented with obstructive jaundice.<sup>8</sup> The present patient had two weeks history of high grade fever and was empirically treated for enteric fever, before presentation.

Radiological imaging provides the best tool for early diagnosis. Selective angiography is the gold standard in the diagnosis of HAPA and at the same time provides a therapeutic opportunity. CT angiography, MRI angiography and Doppler sonography offer promising alternatives to conventional angiography for the diagnosis and treatment planning of HAPA.<sup>2-5</sup> Treatment

options for this HAPA include surgical repair, endovascular repair including super selective micro-coil embolization and stent-grafting. Endovascular therapy stent-grafting is being increasingly utilized in the treatment of visceral aneurysms with optimal outcome.<sup>2,3,5</sup> Placement of stent-graft in an infected area can provoke late graft disintegration and consequently, aneurismal rupture. This patient was not immuno-compromised, but the aneurysm was large with a narrow neck and the bleeding was massive. With hemorrhagic shock and hemoglobin of 5.2 gm/dl, the stent-graft was a more favourable option in this case and successfully controlled the bleeding. There have been two reported cases of HAPA causing obstructive jaundice because of compression on right biliary duct.<sup>3,8</sup> This patient had pictures of obstructive jaundice on first presentation but improved after stent-grafting of HAPA. She subsequently developed cholangitis due to stricture of right hepatic duct close to the site of aneurysm and the explanation for this could be compression leading to stricture. The right hepatic duct stricture was successfully treated with retrograde balloon dilation and stenting. The second admission could have been avoided by early ERCP, which could have been diagnostic and therapeutic on the 1st admission.

Two lessons are learned from this case; one is that stent-grafting of hepatic artery pseudoaneurysm is an effective treatment option; the second is that HAPA could cause right hepatic duct stricture and can be managed successfully through ERCP with balloon dilation and biliary stenting.

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