The Aftermath of a Hepatic Artery Aneurysm—A Rare Etiology of Biliary Obstruction!

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ABSTRACT

BACKGROUND: Hepatic artery aneurysms (HAAs) constitute 14% to 20% of visceral artery aneurysms. Most HAAs are asymptomatic. Although rare, obstructive jaundice due to external bile duct compression or rupture of the HAA into the biliary tree with occlusion of the lumen from blood clots has been reported.

CASE PRESENTATION: A 56-year-old white man presented to an outside hospital with symptoms of obstructive jaundice, including abdominal pain and yellowing of the skin. Imaging showed a large HAA. Patient was transferred to our hospital where an endoscopic retrograde cholangiopancreatography with biliary stenting was performed. This was followed by coil embolization of the HAA with improvement in symptoms and liver chemistries.

CONCLUSIONS: Most clinicians agree that management of HAA is highly variable and depends on clinical presentation and anatomic location. Biliary stenting provides temporary relief for patients with obstructive jaundice. Definitive options include open aneurysmal repair versus endovascular therapy. Hepatic artery aneurysms represent a significant risk for hemorrhage and therefore must be addressed promptly once discovered.

KEYWORDS: Hepatic artery aneurysm, obstructive jaundice, biliary stenting, Quincke triad

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Introduction

Hepatic artery aneurysms (HAAs) are relatively rare; as stated above, they only comprise 20% of all visceral artery aneurysms. Eighty percent of aneurysms are extrahepatic and usually occur in the common hepatic artery (60%). Thirty percent of aneurysms occur in the right hepatic artery, whereas only 5% occur in the left hepatic artery. Hepatic artery aneurysms are more common in men (male:female = 2:1).[1] The mean age of presentation is in the sixth decade but have been documented in patients of all ages. The following case represents a classic presentation of obstructive jaundice secondary to a surprising anatomic defect.

Case Report

A 56-year-old white man with history of chronic obstructive pulmonary disease and hypertension presented to an outside hospital with complaints of yellowing of the skin and eyes, right-sided abdominal pain, and dark urine for 3 days. Relevant physical examination was significant for scleral icterus, jaundice, and diffuse abdominal tenderness to palpation, worst in the right upper quadrant. Pertinent laboratory work revealed white blood cell count of 9000/mm^3, total bilirubin of 24 mg/dL, alkaline phosphatase (ALP) of 1104 U/L, aspartate transaminase (AST) of 200 U/L, and alanine transaminase (ALT) of 236 U/L. Computed tomography (CT) of the abdomen and pelvis revealed intra- and extrahepatic biliary dilatation with an HAA compressing the common bile duct (CBD). He was transferred to our hospital. Patient underwent an endoscopic retrograde cholangiopancreatography (ERCP) with cholangioscopy. This revealed a CBD stricture approximately 2.5 cm in length at the level of cystic duct takeoff with massive biliary dilation proximally (Figure 1). A plastic stent was deployed traversing the stricture. Biopsies were obtained, and pathology only identified inflammation. Computed tomographic angiography with three-dimensional reconstruction confirmed location of an extrahepatic fusiform HAA compressing the CBD (Figures 2 and 3). He then underwent coil embolization of the hepatic artery (Figure 4A and B). Stent grafting could not be attempted secondary to discrepancy in the size of proximal (10 mm) and distal (5 mm) entry into the aneurysm. Patient tolerated the procedure well. On follow-up at 2 weeks post discharge, patient was asymptomatic with remarkable improvement in hepatobiliary chemistries—AST, 59 U/L; ALT, 52 U/L; ALP, 239 U/L; and total bilirubin, 2 mg/dL. Patient underwent a repeat ERCP (6 weeks after the initial

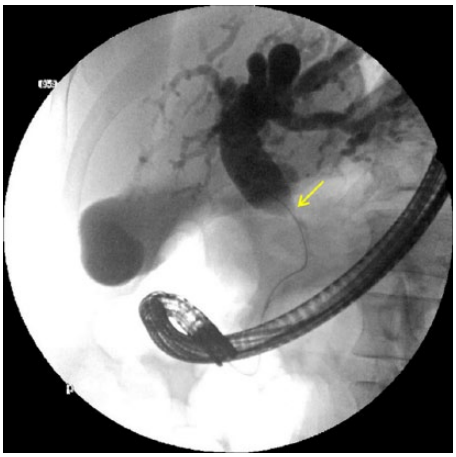

Figure 1. Endoscopic retrograde cholangiopancreatography image shows severe intra and extrahepatic biliary ductal dilatation starting at the proximal common bile duct with tapered narrowing (arrow) due to extrinsic compression from the hepatic artery aneurysm.

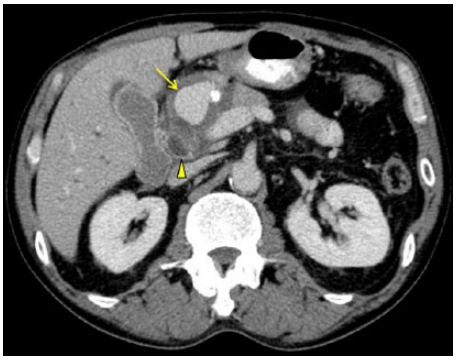

Figure 2. Contrast-enhanced axial computed tomographic image of the abdomen shows a contrast-filled rounded structure (arrow) adjacent to the hepatic hilum compressing the common bile duct (arrowhead). This finding is consistent with a hepatic artery aneurysm. There are intra- and extrahepatic biliary ductal dilatation secondary to an extrinsic compression from the hepatic artery aneurysm.

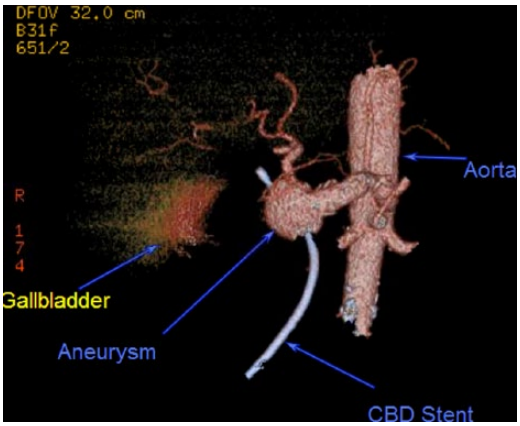

Figure 3. Three-dimensional reconstruction of Figure 2. CBD indicates common bile duct.

ERCP) for removal of the biliary stent. This revealed improvement in the biliary tree dilatation (Figure 5). A CT scan performed 18 months after aneurysmal coiling was limited by metallic artifacts, in its assessment of vasculature. However, poorly characterized biliary system dilatation was improved (Figure 6). Ultrasound performed at the same time revealed a normal caliber (0.47 cm) CBD (Figure 7).

Discussion

Hepatic artery aneurysms are being encountered more frequently by physicians likely secondary to improved diagnostic imaging modalities. Common causes include surrounding inflammation, trauma, vasculitis, arteriosclerosis, and infections, including tuberculosis and syphilis.[2] Hepatic artery aneurysms are also observed as a consequence of percutaneous biliary procedures and liver transplantation. Rarely, congenital diseases are the principal cause; such diseases include Osler-Weber-Rendu/hereditary hemorrhagic telangiectasia, Marfan syndrome, and Ehlers-Danlos syndrome.

Most clinicians agree that management of HAA is highly variable and depends on clinical presentation and anatomic location. Abbas et al[3] expound on the fact that some clinicians recommend repair at any size or anatomic location due to the risk of rupture, whereas other sources only recommend repair once the lesion is >2 cm, unless there are other complicating factors. Risk of rupture ranges between 20% and 80%.[4] Patients at increased risk of rupture include those with HAA of nonatherosclerotic origin and multiple HAA.[3] The risk of mortality is 35% or higher in patients who present with rupture.[1,3] Ongoing research by Longo et al[6] suggests that increased enzymatic activity of matrix metalloproteinases (MMP) may contribute to expansion and rupture of aneurysms. Matrix metalloproteinases contain collagenase, which aids in the degradation of elastin and collagen, key components in the matrix that composes the vascular wall; these destructive proteins are upregulated during states of inflammation. Pathologic differences exist between early-stage aneurysms, which exhibit decreased (though abundant) amounts of collagen as compared with normal vascular tissues, and late-stage or ruptured aneurysms, which are composed of mostly fibrin and inflammatory infiltrate on close inspection. These findings have been directly correlated with rupture of abdominal aortic aneurysms (AAAs), and research has only recently begun to extrapolate these findings to include aneurysms of other vessels.[5,6]

As evidenced in this case, HAA can present as obstructive jaundice, as well as gastrointestinal hemorrhage with hypovolemic shock or vague abdominal pain with or without a pulsatile mass. One-third of patients present with abdominal pain, obstructive jaundice, and hemobilia (Quincke triad). However, hemobilia alone can be the presenting feature in up to one-third of patients as described by Parmar et al,[7] mostly with intrahepatic aneurysms or pseudoaneurysms. Intraperitoneal bleeding is the most common manifestation of extrahepatic aneurysmal rupture or oozing.

Most HAAs are diagnosed incidentally on abdominal imaging or even during surgical procedures. Some estimate that between 30% and 50% of all HAAs are found during autopsy.[8]

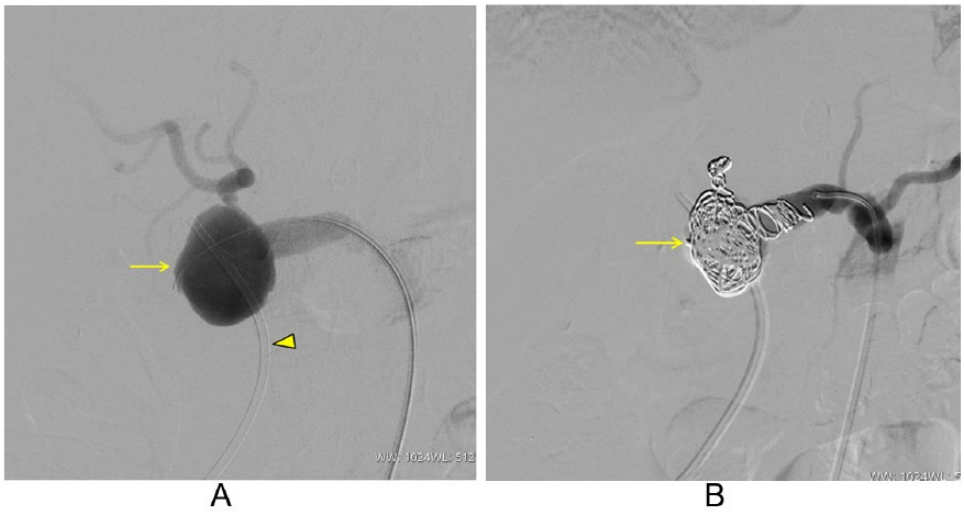

Figure 4. Selective digital subtraction angiography of the hepatic artery (A) before and (B) after embolization. The aneurysmal dilatation of the common hepatic artery is clearly delineated (A, arrow). Incidentally noted a plastic biliary stent (A, arrowhead). Follow-up image after multiple metallic coil placement (B, arrow) shows no contrast material filling the aneurysmal sac.

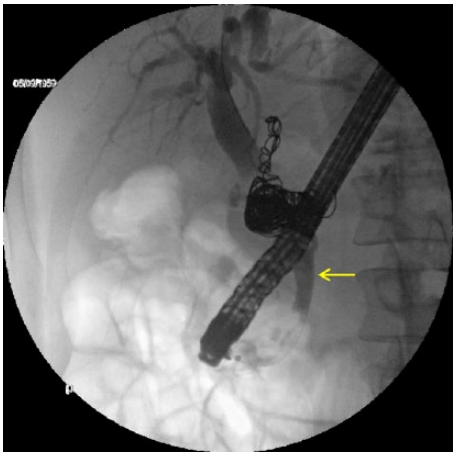

Figure 5. Endoscopic retrograde cholangiopancreatography (ERCP) sagittal view approximately 6 weeks after first ERCP and hepatic artery embolization demonstrates improvement of the intra- and extrahepatic biliary ductal dilatation (arrow).

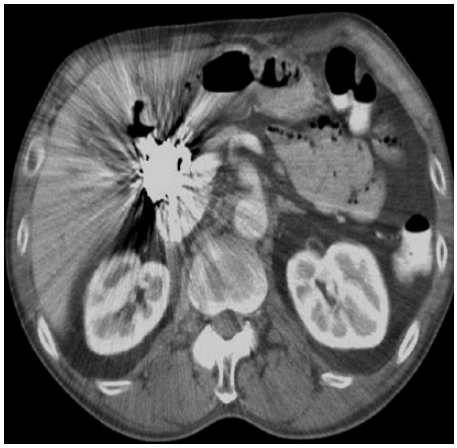

Figure 6. Contrast-enhanced axial computed tomographic image of the abdomen after hepatic artery aneurysm embolization (arrow). Coiling material produces metallic artifacts limiting the evaluation of the surrounding structures. Poorly characterized biliary system dilatation is improved.

Abdominal ultrasound with Doppler is a common initial modality to identify any vascular masses within the right upper quadrant. Ultrasound may also be used intraoperatively during excision to evaluate vascular patency. The criterion standard of radiographic diagnosis is CT angiography, which is required for confirmation of HAAs. Prior to widespread use of CT, aortography was the most common imaging study used for diagnosis and/or aneurysm characterization prior to surgical intervention.

Patients that present with obstructive jaundice are usually candidates for biliary stenting. Biliary stenting prior to surgical management of aneurysm is often attempted in patients without hemobilia. However, risk of the stent entering the aneurysm and precipitating major hemorrhage presents a significant risk. The popularity of endovascular stent grafting is growing among physicians. Current case reports indicate this technique is more commonly used in the treatment of intrahepatic artery pseudoaneurysms.[9] Our patient was not eligible for stent grafting secondary to a discrepancy in the size of proximal and distal entry points of the aneurysm.

In patients with HAAs presenting with hemobilia or Quincke triad, biliary obstruction should be dealt with as part of the definitive excision of the aneurysm and repair of the damaged bile duct. Extrahepatic aneurysms, including those of the common hepatic artery, are often candidates for aneurysmectomy or aneurysmal exclusion. Revascularization is required for aneurysms involving the common hepatic artery, as the liver is at high risk of ischemic injury. On the contrary, percutaneous embolization by interventional radiology remains the most common treatment approach for intrahepatic HAAs. Case series by Lumsden et al[9] suggests

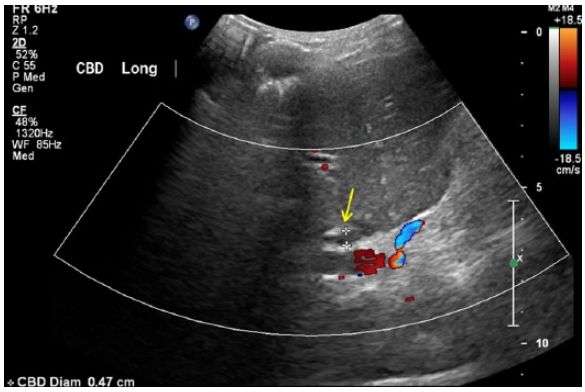

Figure 7. Ultrasound image of the common bile duct (CBD) performed 18 months after hepatic artery embolization shows a normal caliber (0.47 cm) CBD (arrow).

this approach presents a much lower risk of hepatic devascularization and overall morbidity compared with surgical repair. Collateral circulation rapidly develops after occlusion, decreasing the risk of both acute and chronic ischemia.[10] However, patients occasionally require repeat embolization, where repeat surgical excision is less commonly required.

Conclusions

Although rare, HAAs represent a significant cause of morbidity and mortality. Most often, these lesions are found incidentally on abdominal imaging, although they may present symptomatically, as in our patient. All sources agree that treatment strategies are highly variable and should be individualized for each patient. Biliary stenting is a common practice in patients with evidence of obstructive jaundice and without hemobilia. Definitive management with percutaneous embolization or surgical repair is often undertaken due to the high risk of rupture and mortality if left untreated.

Author Contributions

A Seth contributed to design of the article. A Seth, KS, A Sheth, MB, PJ, RR, JB, and GS collected data. Drafting the article, as well as critical revision, completed by CL, A Seth, and JB. All authors approved the final revision for publishing.

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