Hepatic Artery Aneurysm: An Unusual Cause of Upper Gastrointestinal Bleed in 49 Years Old Patient

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Abstract

Hepatic artery aneurysm (HAA) is a rare disease. HAA is generally asymptomatic disease when symptomatic, they usually present with abdominal pain, upper gastrointestinal (GI) bleeding and/or jaundice, hypovolaemia secondary to rupture or GI bleeding with normal GI endoscopy. Surgical repair and endovascular treatment are the two therapeutic options available at present.

Case report: A 49-year-old male presented at the emergency department with high gastrointestinal bleeding, abdominal pain and jaundice. Gastroscopy showed an ulcer with flat pigmented haematin on ulcer base (Forrest IIc) that was controlled by medical treatment. CT angiography was done and showed aneurysm of the proper hepatic artery almost totally thrombosed measuring 100 × 59 mm associated with signs of contained rupture. Emergency surgery was indicated. The laparotomy objectified a rupture of the aneurysm in the biliary tree in per operative excision of aneurysm and ligation of the hepatic pedicle was carried out. After surgery, the evolution was favorable with a follow-up of 8 months.

Conclusion: HAA rupture is a rare cause of upper GI bleeding. The mortality rate after rupture is relatively high. CT angiography or MRI can diagnose a ruptured of HAA. Urgent surgery should be the first choice in patients with a ruptured HAA with active hemorrhage causing hemorrhagic shock.

Keywords

Hepatic Artery Aneurysm, Rupture, Upper GI Bleeding, Surgery
1. Introduction

Hepatic artery aneurysm (HAA) is a rare disease (0.002% - 0.4%) [1]. HAA represents approximately 20% of all visceral aneurysms [2]. The hepatic artery is the fourth common site of intraabdominal aneurysm from any cause following infrarenal aorta, iliac artery and splenic artery [3] [4]. 80% HAAs are extrahepatic and 20% are intrahepatic [3] [4]. HAA is generally asymptomatic disease when symptomatic, they usually present with abdominal pain, upper gastrointestinal (GI) bleeding and/or jaundice, hypovolemia secondary to rupture or GI bleeding with a normal GI endoscopy [5] [6]. The majority of aneurysms are diagnosed incidentally on imaging tests [6]. Imaging modalities like computed CT-angiography, MRI angiography have a valuable role in the early detection of HAA, its complications, and in selecting appropriate treatments depending on the size and location of the aneurysms [7]. Causes of HAA include atherosclerosis, inflammation (such as vasculitis) or iatrogenic (for instance during angiography). They may also be idiopathic [6]. Surgical repair and endovascular treatment are the two therapeutic options available at present [5]. We report the case of a 49-year-old man with upper GI bleeding secondary to a hepatic artery aneurysm.

2. Case Report

The patient was a 49-year-old patient man with a history of uncomplicated choledolithiasis awaiting surgery. There was no history of surgical procedures or trauma. The patient presented to the emergency department with paroxysmal hypochondrium pain associated with pruritis for two weeks and several episodes of hematemesis for the last 24 h. At admission the patient presents jaundice, sensitivity of the right hypochondrium and hematemesis with hemodynamic instability. Clinical examination found icteric sclera and paleness of the mucous membranes. Her abdomen was soft, not distended and slightly tender to palpation. Rectal exam was positive for melenic stool.

Lab results revealed a decreased of hemoglobin at 5.6 g/dL.

Total bilirubine, alkaline phosphatase (ALP), gamma-glutamyltranspeptidase (GGT), Alanine aminotransferase (ALT), Aspartate aminotransferase (AST) were elevated at 56 mg/L, 43 mg/L, 378 U/L, 86 U/L, 102 U/L and 87 U/L respectively. AgHBs, anti-HCV antibodies, anti-mitochondrial antibodies, antinuclear antibody (ANA), smooth muscle antibody (ASMA), liver kidney microsomal antibody (LKM) were negative.

After resuscitation with crystalloid solutions and blood transfusions, an upper digestive endoscopy was performed, reporting a 15 mm round ulcer with flat pigmented haematin on ulcer base (Forrest IIc) on the anterior side of the duodenal bulb. The diagnosis of acute bleeding of peptic ulcer was done and treatment with proton pump inhibitor was beginning. The evolution was marked by the persistence of the hematemesis with hemodynamic shock.

After a second resuscitation another upper digestive endoscopy was per-
formed and showed clean ulcer base on the anterior side of the duodenal bulb (Forrest III), there was no luminal blood or stigmata of recent bleed, the major duodenal papilla was normal without signs of bleeding. The evolution of the ulcer (Forrest IIC to III) had not explaining the bleeding. The association of pain in the right hypochondrium, jaundice and upper GI bleeding, constituting the classic triad Quincke’s suggested a haemobilia. CT angiogram was done and showed aneurysm of the proper hepatic artery almost totally thrombosed measuring 100 × 59 mm associated with signs of contained rupture compressing the hepatic artery in distally, the portal trunk and the superior biliary confluent responsible for a diffuse ischemia of the liver with dilatation of the intrahepatic bile ducts and lithiasic gallblader without signs of cholecystitis.

Emergency surgery was indicated. The laparotomy objectified a rupture of the aneurysm in the biliary three; in per operative an excision of aneurysm and ligation of the hepatic pedicle was carried out.

The postoperative evolution was marked by a regression of the hematemesis and a decrease in the intensity of the jaundice, the abdominal pain and a good hemodynamic stability.

The patient was discharged fifteen days after admission, maintaining an outpatient follow-up. At follow-up 8 months later, the patient was asymptomatic. He has not present new bleeding episodes or other complications derived from the procedure thus far (Figure 1).

3. Discussion

Hepatic artery aneurysm was first reported by the English anatomist Wilson in 1819 during an autopsy [8]. The first symptomatic hepatic artery aneurysm was reported by Quincke [9].

The first surgical treatment was performed by Kehr in 1903 and the first successful revascularization was performed by Paul in 1951 [10] [11].

Hepatic artery aneurysm is a rare disease (0.002% - 0.4%) [1].

HAAs are the second most common visceral artery aneurysm, accounting for 4% - 30% [12]. The hepatic artery is the fourth common site of intraabdominal aneurysm from any cause following infrarenal aorta, iliac artery and splenic artery. It is more frequent in men, and the incidence is highest between the ages of 40 and 60 years old. 80% HAAs are extrahepatic and 20% are intrahepatic [13].

63% of HAAs involve the common hepatic artery, 28% involve the right hepatic artery, 5% involve the left hepatic artery, and 4% both the left and right hepatic arteries [3] [4]. The clinical manifestations depend on the size of the aneurysm. Although small HAAs are often asymptomatic, the natural history is for progressive enlargement with increasing risk of rupture and death [7].

The frequency of rupture is reported as 20% - 30%, and the mortality rate after rupture is relatively high, at 35% [14].

In 80% of the cases rupture of the aneurysms is the first clinical manifestation.

The aneurysms can rupture with equal frequency into the biliary tree or abdominal cavity [2].
Axial view CT angiography demonstrating aneurysm of the proper hepatic artery almost totally thrombosed associated with signs of contained rupture.

In the symptomatic patients HAA may cause abdominal pain, jaundice, GI bleeding, hypovolemia, and abdominal mass. In our patient, GI bleeding and jaundice were the first symptoms of the HAA.

Several clinical cases have reported the same clinical manifestations as circumstances of discovery of HAAs [1] [2] [5] [6].

The classic triad of hemobilia—Quinke’s triad, i.e., obstructive jaundice, abdominal pain and GI bleeding, is seen in 30% of the patient [7].

The Quinke’s triad is present in our patient. Only about 10% of cases of hemobilia are secondary to a ruptured aneurysm of the hepatic artery. Portal hypertension may exist due to compression of the portal trunk by the aneurysm.

Endoscopy is the first investigation for upper gastrointestinal bleeding; in patients with HAA GI endoscopy is usually normal or inconclusive.

In our patient, the GI endoscopy did not show signs suggestive of rupture of HAA. Rizo and al also report the same aspect [5].

Before, the diagnosis was made post mortem during the autopsy or after an emergency laparotomy. Nowadays, with the progress of imaging, the diagnosis is easy and relies essentially on CT angiography, MRI angiography, angiography and sometimes abdominal doppler ultrasound. Endoscopy is the first investigation for upper gastrointestinal bleeding in patients with HAA; endoscopy is usually normal or inconclusive.

Plain film of the abdomen may occasionally show a curvilinear calcification representing the wall of aneurysm in the right upper quadrant of the abdomen [15]. Despite lower sensitivity, ultrasonography can reveal the presence of an aneurysm as a mixed echogenic mass with variable proportions of cystic and solid components, depending on the extent of thrombosis. Calcifications may occasionally be seen in the wall [16] [17].

Doppler ultrasound can help differentiate vascular masses from other types of
masses. Color Doppler shows arterial or turbulent flow in the lesion, suggesting that it is a mass of vascular origin [2] [3].

In addition, color Doppler ultrasound can differentiate aneurysms from other vascular anomalies, such as fistulas or arteriovenous malformations [18].

CT angiography is an excellent examination for evaluation of HAA, but it exposes patients to high radiation doses. The CT angiography is useful for detecting small aneurysms and assessing anatomical details and is being used instead of angiography. The technique can confirm the diagnosis of HAA, delineates the feeding vessels, depicts collateral blood flow, reveals any other aneurysms and shows anatomic variations of the vessels [19].

MRI angiography is another non-invasive technique for the diagnosis and visualization of HAA. It is a rapid, accurate, and noninvasive technique for the diagnosis and visualization of visceral artery aneurysms. It provides a good representation of anatomy and arterial variations and can guide the choice of appropriate treatment [20].

The selective angiography is the most valuable investigation modality for diagnosis of HAA which has a sensitivity of 100% [21].

The main etiology of hepatic artery aneurysm is atherosclerosis. Other common causes include fibrodysplasia, intra-abdominal surgery (including liver transplantation, which is increasing), vasculitis, mycotic aneurysms, trauma, polyarteritis nodosa, lupus, pancreatitis and tuberculosis. Aneurysms related to bacterial endocarditis are disappearing. Rare causes are Takayasus disease, Kawasaki disease, Von Recklinghausen neurofibromatosis, Wegener granulomatosis, prolonged amphetamine use, pregnancy, and congenital diseases (Marfan, Ehers-Danlos, Rendu-Osler syndrome) [22].

The management of HAA is based on endovascular, surgical methods. Surgery has been supplanted in the treatment of HAA and should only be considered in unstable patients, aneurysms larger than 5 cm or after failure of endovascular treatment. Surgical treatments include excision of the aneurysm with saphenous vein grafting or excision and ligation of the hepatic artery [23].

In endovascular methods, thrombin, vascular adhesives, onyx, or spirals are administered to the aneurysm causing the aneurysm to clot or the stents can be inserted into the artery to cut off the blood supply to the aneurysm and preserve blood flow [24].

In our patient, it was decided to perform surgery and the evolution was favorable with a follow-up of 8 months.

4. Conclusion

HAA rupture is a rare cause of upper GI bleeding. The mortality rate after rupture is relatively high. The association of jaundice, abdominal pain, and gastrointestinal bleeding suggests hemobilia and should motivate the performance of CT angiography or MRI angiography to diagnose HAA rupture. Urgent surgery should be the first choice in patients with a ruptured HAA with active hemorrhage causing hemorrhagic shock.
Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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