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Case report

A Case Report: Hepatic artery pseudoaneurysm causing life-threatening haemobilia

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ABSTRACT

Introduction & importance: Hepatic artery aneurysms (HAA) are rare and it accounts 20% of all visceral artery aneurysms. Commonly HAAs are autopsy findings, but rupture and bleeding carrying significant morbidity and can manifest as haemobilia.

Case presentation: A 63-year-old Sri Lankan male presented with severe melaena upper abdominal pain and features of obstructive jaundice was found to have a giant pseudoaneurysm at the right hepatic artery with the possible arterio-biliary fistula. The etiology for the pseudoaneurysm was not identified. Despite massive transfusion, the patient died before the endovascular intervention.

Clinical discussion: Atherosclerosis is the leading cause of HAA formation but can be associated with connective tissue disorders and arteritis. Most of the HAA are asymptomatic. Aneurysms can be managed with surgical or endovascular interventions.

Conclusion: Life-threatening haemobilia is a notorious complication of the rapture of HAA into the biliary system. The incidents of hepatic artery aneurysms and pseudoaneurysms due to percutaneous transhepatic interventions and minimal invasive hepatobiliary surgeries are in the rising trend. Nonleaking VAA can be best treated with endovascular treatment. The knowledge on this topic is important for the early detection and intervention of this rare entity.

1. Introduction

Visceral artery aneurysms (VAA) are rare and it is defined as aneurysms involving the celiac artery, superior mesenteric artery (SMA), inferior mesenteric artery and/or their branches with an estimated prevalence of 0.1 to 2%. Among VAA hepatic artery aneurysms (HAA) and pseudoaneurysm represents approximately 20% next to splenic artery aneurysm which is about 60%. Rupture and bleeding of the VAA carry significant morbidity and mortality, but most of the aneurysms are found incidentally when performing abdominal imaging for other reasons [1–2]. The definitive cause for HAA is unclear. Unlike the early twentieth century, mycotic aneurysms are seldom seen nowadays due to early diagnosis and antibiotic treatment of infective endocarditis. Iatrogenic and traumatic causes, atherosclerosis, vasculitis, as sequelae of *peri* inflammation such as cholecystitis and pancreatitis are the most reported causes in the recent articles [3–5].

Here we report a case of ruptured idiopathic hepatic artery pseudoaneurysm into the biliary system and presented as haemobilia.

This case report has been prepared according to the SCARE 2020 guideline [6].

2. Case presentation

A 63-year-old Srilankan male presented to the Emergency department of Teaching hospital-Jaffna, Sri Lanka, with the features of obstructive jaundice and severe melaena with symptoms of anemia for two days duration. He is a known patient with hypertension for sixteen years and was diagnosed with recurrent glioblastoma for two years. He underwent surgical excision of glioblastoma twice and completed radiotherapy and is now on oral chemotherapy. On examination, he was deeply icteric, not febrile to touch, pale and his heart rate was 128/min, Blood pressure was 100/70mmHg. He had mild upper abdominal tenderness with no mass. Digital rectal examination revealed melaena. After initial fluid resuscitation, 4 units of blood, fresh frozen plasma, and platelets were transfused for this patient. Intravenous Ceftriaxone 1g, IV tranexamic acid 1g, and vitamin K 10mg were given to the patient.

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Investigations on admission revealed Hemoglobin of 4.2 g/dL with a hematocrit of 12.3 and normal white cell and platelet count. His Total bilirubin was 204 μmol/L(0-17) with a predominant direct component of 170 μmol/L (0-3). Alkaline phosphatase was 948 (46-116) with mildly elevated transaminases Alanine Aminotransferase (ALT): 116 U/L (16-63) Aspartate Aminotransferase (AST). Prothrombin time was 11.9 with an INR value of 1.1. His inflammatory markers were normal with the erythrocyte sedimentation rate (ESR) of 40 mm/1st hour, and other blood investigations were not significant. After initial resuscitation, the patient improved symptomatically with the static Hb of 10.3 g/dL. He subsequently transferred to the Professorial Surgical Unit, the University of Jaffna-Sri Lanka for further evaluation and management.

An ultrasound scan (USS) of the abdomen showed a cystic structure measuring 6.2 × 6.9 cm with the bi-directional colour flow (Ying-Yang sign) near the pancreatic head suggestive of a hepatic artery aneurysm. The common bile duct and the intrahepatic ducts were dilated due to compression by the aneurysm. Heterogenous material filling the gall bladder suggestive blood. There was no free fluid in the abdomen. Upper GI endoscopy revealed there was a pulsatile mass within the gastric body and the first part of the duodenum. The second part of the duodenum is filled with blood (Fig. 1). The findings of the Contrast-enhanced Computed tomography of the abdomen and Angiogram was compatible with hepatic artery pseudoaneurysm (Fig. 2).

The 2D ECHO cardiogram revealed the left ventricular ejection fraction was 45% and no vegetations in the valves. Blood cultures were negative for the bacteria and fungi. The patient had not been undergone any hepatobiliary procedures in the past. He did not have a past medical history of infective endocarditis, vasculitis, or connective tissue disorders.

The multidisciplinary discussion was carried out including Vascular surgeons, Hepatobiliary surgeons, interventional radiologists, and family members. The possibilities of surgical correction, coil embolization, and angioembolization were discussed. Considering the size of the aneurysm and the broad neck, and the patient's preexisting glioblastoma, endovascular coil embolization had been proposed as the best option than open surgery. The patient was decided to be managed conservatively as the patient and the family members refused any sort of surgical endovascular treatment (EVT). On day five of admission unfortunately patient developed sudden onset of severe bleeding per rectum. Despite massive blood transfusion and active resuscitation, the patient succumbed to hemorrhagic shock.

3. Clinical discussion

HAA is potentially fatal as the frequency of rupture is more common in HAA than in other VAA. It is reported as 20–30%, and the mortality rate after rupture is relatively high, at 35%. HAA can be intrahepatic (20%) or extrahepatic (80%). Approximately 60% of the extrahepatic artery aneurysms are involving the common hepatic artery. HAAs are found more commonly in men than women [7]. True aneurysms are much commoner than pseudoaneurysms, but in the case of HAA, both are equal. Approximately, half of the pseudoaneurysms are located in the parenchyma of the liver, which reflects the increasing incidence of iatrogenic etiology.

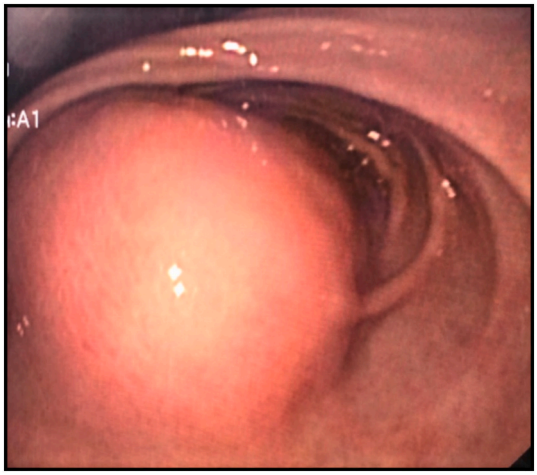

Fig. 1. Budging mass in the first part of the duodenum.

Atherosclerosis is the leading cause of HAA formation which accounts for more than half. Compare to other VAA, the higher incidence of Hepatic artery pseudoaneurysms are due to diverse complications of invasive transhepatic procedures and surgeries. These interventions are the second most common etiology. The less common causes include fibromuscular dysplasia, Segmental arterial mediolysis (SAM), polyarteritis nodosa, infection, trauma, and biliary diseases. Rarely HAA has been associated with disorders such as Takayasu arteritis, Kawasaki disease, von Recklinghausen neurofibromatosis, and Wegener granulomatosis. Congenital causes of HAA include Marfan syndrome, Ehlers-Danlos syndrome, Osler-Weber- Rendu syndrome, and hereditary hemorrhagic telangiectasia. Only a few patients are diagnosed to have VAA due to infective causes [8–10].

Most HAAs are asymptomatic and are found incidentally during screening for other diseases. Large aneurysms rarely can cause mass effects and manifest as obstructive jaundice. If the rupture occurs into the biliary system usually patients may present with bloody vomiting and Malena or bleeding per rectum as well as anemia. Contrast-enhanced CT angiography, magnetic resonance angiography, and the abdominal US help to identify the morphology and features of the aneurysm, and it facilitates a definitive diagnosis [1].

The relationship between aneurysm size and risk for rupture has been difficult because of the rarity of HAA. However, the risk of rupture is more in non-atherosclerotic aneurysms [9]. Treatment is generally indicated for all symptomatic HAAs, pseudoaneurysms and aneurysms larger than 20 mm in diameter. Multiple HAAs and nonatherosclerotic aneurysms should also be treated, regardless of size, since these HAAs have a high risk of rupture [11].

Like other VAAs, treatment options for HAAs depend on the anatomy, morphology, and location of the aneurysm, as well as the patient's clinical status and comorbidities. Treatment options can be surgical such as aneurysmectomy, ligation, and bypass with a venous graft or prosthetic vascular graft, laparoscopic surgery (mainly ligation), EVT (embolization and endo grafting), or a combination of treatments. Though there are no randomized control trials that have been able to identify EVT as being superior to open repair, considering the surgical trauma associated with open procedures and mortality rate, non-leaking VVA favor endovascular treatment using covered stents or stent-grafts [12].

If an aneurysm develops in the common hepatic artery, surgical ligation or endovascular coil embolization can be performed because the Gastroduodenal arteries (GDA) facilitate collateral circulation to the liver. When the aneurysm is in the proper hepatic artery or the left or right hepatic artery, peripheral to the GDA branch, arterial reconstruction is generally required to maintain hepatic blood flow. In practice, the right and left hepatic arteries have intrahepatic communication; thus, major complications arising from the use of coil embolization via EVT or ligation of one of the hepatic arteries (right or left) are rare. Most intrahepatic artery aneurysms are treated with embolization using EVT [11]. Image-guided human thrombin injection has been widely used in peripheral false aneurysms. The recurrence rate is high after thrombin treatment for pseudoaneurysms when the neck is wide [13].

Though the open surgery was the better option for our patient as he had an aneurysm at the terminal right hepatic artery with the wide neck. Considering his life expectancy due to the pre-morbid condition and intraoperative mortality, Endovascular coil embolization was thought to be the better treatment option for this giant Hepatic artery pseudoaneurysm.

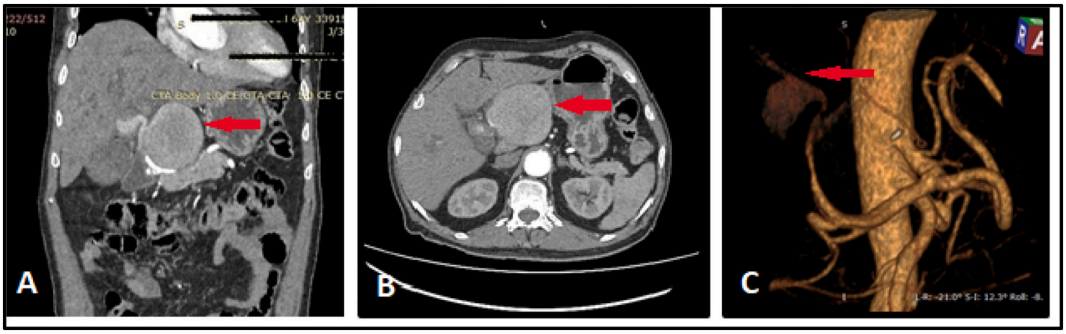

Fig. 2. Contrast-enhanced Computed tomography of the abdomen and Angiogram- A: Coronal view B: Axial view C: 3D Reconstructed view. Enhancing lesion is seen adjacent to the hepatic artery measuring 6.7 × 6 × 5.6 cm (red arrow). It appears to be arising from the right hepatic artery favoring an aneurysm. No focal lesions in the liver. Intrahepatic bile ducts are dilated. High-density areas are seen in the gall bladder suggestive of blood. No growth in the gall bladder. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

4. Conclusion

A rare cause of obstructive jaundice and Malena can be due to rupture of giant Hepatic artery pseudoaneurysm into the biliary system. Life-threatening haemobilia is a notorious complication of this rare entity. Early intervention is necessary to prevent mortality. The incidents of hepatic artery aneurysms and pseudoaneurysms due to percutaneous transhepatic interventions and minimal invasive hepatobiliary surgeries are in the rising trend. Nonleaking VAA can be best treated safely and effectively with endovascular treatment. The knowledge on this topic is important to general and hepatobiliary surgeons for the early detection and intervention of this rare entity.

Informed consent

Informed written consent was obtained from the patient's next of kin for publication of the data and clinical images. A copy of the written consent is available for review by the Editor in chief of this journal on request.

Authors' contributions

Authors JS and SR (Clinical supervisor) have equally contributed to the concept, design, data collection, and writing the case report.

Declaration of competing interest

Both authors disclose any financial and personal relationship with other people or organizations that could inappropriately influence their work.

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Guarantor

Professor S Raviraj-Board-certified consultant surgeon and professor in surgery, Department of Surgery, Faculty of Medicine, University of Jaffna, Sri Lanka.

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Ethics approval

All procedures followed were in accordance with the ethical standards of the institution (University surgical unit, Teaching hospital Jaffna). Institution exempts ethics approval for reported cases.

Registration of research studies

Not applicable.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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