Management of hemodynamically unstable, ruptured multiple hepatic artery pseudoaneurysms by open surgery combined with endovascular embolization

Meng-Hsuan Chung¹, Yi-Szu Wen¹,², Hsiuo-Shan Tseng³, Hsin-Chin Shih¹,²

¹Department of Emergency, Division of Trauma, Taipei Veterans General Hospital, ²Faculty of Medicine, National Yang-Ming University, ³Department of Radiology, Taipei Veterans General Hospital, Taipei, Taiwan

Abstract

Hepatic artery pseudoaneurysm is a type of visceral artery aneurysm, which may cause life-threatening situation if the diagnosis and treatment are delayed after rupture. Increased clinical awareness and aggressive, definitive management are necessary in obtaining optimal outcomes. In general, surgery is the preferred treatment for extrahepatic lesions, whereas embolization is appropriate for intrahepatic aneurysms. However, exploratory laparotomy for vascular ligation or anastomosis is the only way in emergency if the hemodynamic status is unstable. Since the cases of ruptured pseudoaneurysm in both intra- and extra-hepatic artery is rare, we present a case of ruptured hepatic artery pseudoaneurysms with hemorrhagic shock rescued by surgery to stabilize the patient, followed by angioembolization to manage the multiple hepatic aneurysms and provide a review of the current literature on this topic, focusing on appropriate decision-making under multidisciplinary management.

Keywords: Angioembolization, endovascular, hepatic artery aneurysm, liver, pseudoaneurysm

INTRODUCTION

Hepatic artery pseudoaneurysm (HAA) is a type of visceral artery aneurysm which may potentially cause life-threatening situation, especially when the diagnosis and treatment are delayed. However, the initial presentation of HAA in bleeding is usually non-specific clinically with right upper quadrant or epigastric pain only, but the mortality rate of ruptured HAA is estimated 33%–70%.[4] Since the cases of ruptured HAA with concomitant intra- and extra-hepatic artery is rare, we present a patient in this critical situation with hemorrhagic shock, who was then successfully rescued by both surgery and angioembolization under a multidisciplinary team approach.

CASE REPORT

A 66-year-old female with no systemic diseases came to our Emergency Department (ED) in the evening due to progressive epigastric pain after Tai Chi exercise in the morning on the same day. She could not recall any trauma history except a contusion by bamboo stick around upper abdomen in a week ago, but there was not
any ecchymosis or discomfort after the blunt trauma. In the ED, the vital signs showed respiratory rate: 20/min, heart rate (HR): 86/min, blood pressure (BP): 128/72 mmHg, body temperature: 37.5°, and Glasgow coma scale: E4V5M6. The physical examination found general malaise, mild cold sweating, and epigastric dull pain without radiating to back. Laboratory data showed normal white blood cell (WBC) count (8500/µl) with mild increase of segment percentage (89.3%) and C-reactive protein (CRP) (3.04 mg/dl). The liver enzymes and renal function were all within normal limits. Besides the diagnosis of gastroesophageal regurgitation disease and gastritis, diverticulitis or other infectious diseases were suspected, and computed tomography (CT) scan with contrast was arranged which found neither fluid nor active lesions in the abdominal cavity except a 0.9-cm hemangioma in the left hepatic lobe [Figure 1]. She was discharged with oral medication of H2-blocker for symptoms in the next early morning.

However, she still felt epigastric discomfort with nausea and vomiting. Hence, she was transferred back to our ED by ambulance for further diagnosis after visiting a local clinic in the afternoon. On arrival, suspicious seizure attack was noted, and vital signs showed respiratory rate: 21/min, HR: 99/min, and BP: 80/53 mmHg. Bedside sonography disclosed localized bowel wall thickening over the epigastric region with minimal ascites. Laboratory data showed decreased hemoglobin level (10.3 g/dl) with increased WBC level (12,300/µl), lactate: 62.6 mg/dl, and CRP: 4.24 mg/dl. Because of suspicious ischemic bowel disease or hallow organ perforation, abdominal CT scan was immediately arranged again which found multiple hepatic aneurysms [Figure 2] in the territories of replaced left hepatic artery, replaced right hepatic artery from superior mesenteric artery origin and middle hepatic artery from gastroduodenal artery origin. Ruptured pseudoaneurysm in active bleeding status with hemoperitoneum and hematoma formation over hepatic hilum region which compromised left portal vein was diagnosed.

The patient’s condition deteriorated to HR: 146/min, BP: 68/45 right after the CT scan despite aggressive fluid, and blood products resuscitation with inotropic agent support. Due to unstable hemodynamics, radiologic intervention could not be arranged, and trauma/acute care surgeon was consulted. Emergent exploratory laparotomy was arranged under the diagnosis of traumatic pseudoaneurysm rupture after blunt liver injury.

During the operation, hugely engorged round ligament was firstly noted while massive blood was suctioned after entering the abdomen. Pringle maneuver was applied, but active bleeders were even exaggerated along the round ligament to the hilar plate despite multiple attempts of suture ligation. Considering the arterial communication in the liver parenchyma from replaced left hepatic artery, we immediately looped and ligated it, and the bleeders from hepatic hilum improved significantly [Figure 3]. Since the hemodynamic status had been getting stable, the patient was transferred to the intensive care unit after closing the abdomen.

Diagnostic angiography was arranged in the coming morning to determine the extent of residual lesions which were found in the preoperative CT scan. The aortogram showed the ligated replaced left hepatic artery and multiple attenuated lesions along the territory of middle hepatic artery with one impending ruptured pseudoaneurysm. During the examination, angioembolization of the middle hepatic artery with 8 coils for complete control

![Figure 1](https://via.placeholder.com/150)

**Figure 1:** Contrast-enhanced abdomen computed tomography on the first Emergency Department visit showed no lesions, and the portal vein (white arrow) and hepatic arteries (black arrow) were normal

![Figure 2](https://via.placeholder.com/150)

**Figure 2:** (a) Multiple hepatic aneurysms in the territories of replaced left hepatic artery (arrowhead), middle hepatic artery from gastroduodenal artery origin (black arrow), and (b) replaced right hepatic artery from superior mesenteric artery origin (white arrow)
of the aneurysms was performed, and the diagnosis of pseudoaneurysm rupture was confirmed as well according to the image characteristics of contrast CT with dynamic study and angiography.

The alanine aminotransferase (ALT) and aspartate aminotransferase (AST) levels elevated to ALT: 3293, AST: 2437 after surgery and angioembolization, but returned to normal on postoperative day 10. Autoimmune diseases including vasculitis were surveyed without clinical diagnosis in the patient. Her hospital course was smooth and she was discharged home on postoperative day 12. The follow-up CT scan in 3 months [Figure 4] showed normal perfusion of the left lobe of liver and the 1-year follow-up showed stable status.

DISCUSSION

According to the structure of the wall, it is possible to classify hepatic artery aneurysms (HAA) as true aneurysms or pseudoaneurysms\(^1\) and approximately 50% of HAA are pseudoaneurysms. The definitive diagnosis between true and false hepatic artery aneurysms can be made through CT angiography according to the shape and contrast attenuation. In comparison to pseudoaneurysms with different shapes and variable attenuations, the true ones are usually fusiform shape and filled contrast.\(^2\) It can be congenital or secondary to an acquired condition such as atherosclerosis or infection. It also can be seen after surgical injury, chronic damage, pancreatitis, cholecystitis, or most commonly after a penetrating trauma. The presentation usually occurs in a delayed but symptomatic fashion, averaging 5.7 months after injury. The previous report of Abbas \textit{et al}.\(^3\) found only 18 cases of HAA during a two decades chart review in Mayo Clinic, and the operative mortality rate of ruptured hepatic artery aneurysm is estimated 33%–70%,\(^4\) whereas the complication rate is high.

HAA is a potential deadly condition due to its tendency to induce massive hemorrhages with minimal symptoms and signs, but the early diagnosis of this condition is difficult since the clinical manifestations are not specific and nonsuspicious.\(^5\) For those HAA in bleeding, the most common complaint is right upper quadrant or epigastric pain while other presentations include fever, gastrointestinal bleeding, hemoperitoneum, hematemesis, abdominal mass, jaundice, and hemobilia.\(^6\) However, patients presenting with rupture into the peritoneal cavity are rare. The key to diagnosis is to be aware of the possibility of an HAA in patients who present with gastrointestinal bleeding, abdominal pain, shock in 2 weeks or more after abdominal trauma, or after any interventional procedure of the liver or biliary tract.\(^7\)

Despite several imaging modalities have been available to aid in the diagnosis of HAA and pseudoaneurysm, such as ultrasound with the addition of color Doppler,\(^7,8\) there is no strong evidence in using bedside ultrasound in the ED to detect such lesions since the sensitivity rate was low, and it is insufficient to detect all ruptured visceral aneurysms.

In the setting of ED, CT with contrast (venous phase) helps, but the situation of impending ruptured may not be early detected as what happened in our case unless CT angiography was performed. The presence of a hematoma around the hepatoduodenal ligament on contrast/noncontrast CT imaging studies might raise suspicion of vascular injury and should lead to tests.
to exclude this possibility.\textsuperscript{[9]} For those with suspicions, angiography is the definitive diagnostic procedure. In the meantime, CT angiography can not only provides accurate information regarding the anatomy of visceral aneurysms rapidly but also delineates the extent of extravasation and hemoperitoneum as well as demonstrates any associated intraperitoneal or retroperitoneal pathology.

For hemodynamically stable cases of ruptured extrahepatic artery aneurysm after resuscitation, endovascular embolization can be considered firstly. Otherwise, open surgical treatment is still preferred to control the bleeding points. Owing to the perfusion provided by the gastroduodenal and right gastric branches, lesion of the common hepatic artery can usually be ligated or resected without reconstruction while reconstruction is suggested in distal lesions to avoid ischemic damage to the liver and bile ducts due to several reports of central liver necrosis following ligation of proximal hepatic lesions.\textsuperscript{[10]} However, Lam \textit{et al.}\textsuperscript{[10]} proposed that the establishment of arterial collaterals should be expected because the buffer response of the portal venous system can protect the liver from ischemic injury after arterial ligation till the compensatory arterial supply develops. Besides, the use of intraoperative ultrasound facilitates the decision-making in devising the operative strategy for this condition even if the lesion is distal to the gastroduodenal artery.

When the ruptured aneurysm is located intrahepatically, the primary treatment modality of endovascular embolization with highly selective techniques is imperative to minimize the extent of liver ischemia, but it may still result in liver infarction and subsequent infection after the arterial embolization which often requiring resection of the involved liver parenchyma.\textsuperscript{[11]} As what the Mayo Clinic team recently reported,\textsuperscript{[12]} the HAA repair in the setting of rupture carries a high mortality rate, and open repair is associated with notable morbidity rates of >20%. Endovascular therapy may be effective and offer innovative ways to treat HAA and to avoid the morbidity associated with open repair. No matter what method we have, the treatment for HAA has to be tailored for each patient.

In our case, the ruptured HAAs were presented both intra- and extra-hepatically complicated with unstable hemodynamic status despite the effort of massive transfusion and inotropes use. Thus, we immediately proceed to the operating room for arterial ligation followed by angioembolization by the radiologist after stabilizing the hemorrhage. Through the multidisciplinary team approach, the patient was saved without any complications except short-term liver enzyme elevation due to ischemic injury. The follow-up contrast CT images in arterial phase showed blood flow is fair, which implies the theory of ligation distal to proper hepatic artery is feasible.

\section*{CONCLUSION}

Due to low incidence, high morbidity and mortality associated with ruptured HAA, a multidisciplinary team approach between emergency physician, acute care surgeon, and intervention radiologist is important in instant diagnosis and treatment. The unsolved epigastric or right upper abdominal pain, as well as perihilar edematous change in CT image, shall be warning. The open and endovascular methods of surgery must be organized and discussed timely. The safety of ligation distal to proper hepatic artery is still controversial but may be feasible in some circumstances under intraoperative monitor with Doppler sonography.

\section*{Declaration of patient consent}

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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\section*{Conflicts of interest}

There are no conflicts of interest.

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