

Case Report

Cystic artery pseudoaneurysm: an uncommon complication of a common disease

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Received: 30 September 2022

Accepted: 14 October 2022

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ABSTRACT

Cystic artery pseudoaneurysm, though rare, is a notable cause of upper GI bleed. Here, we report an unusual presentation of upper gastrointestinal haemorrhage due to a cystic artery pseudoaneurysm secondary to acute cholecystitis in a 54-year-old male. Initial imaging revealed cholelithiasis with evidence of acute cholecystitis. Further investigations with Doppler ultrasound and CT suggested evidence of a cystic artery pseudoaneurysm with hyperdense contents in the gall bladder suggesting blood products. Taking into consideration the risk of hemorrhage during the procedure, the patient was taken up for open cholecystectomy. This case emphasizes the need for a high level of awareness of pseudo-aneurysmal disease and inclusion of cystic artery pseudoaneurysm as one of the differential diagnosis for a patient presenting with recurrent upper quadrant pain, jaundice and melena or hemobilia.

Keywords: Cystic artery pseudoaneurysm, Melena, Hemobilia

INTRODUCTION

Cystic artery pseudoaneurysms (CAP) are rare clinical entities and are associated with quite a notable level of morbidity.¹ Very few reports are available in the literature making it a rare entity which needs reiteration. CAP can occur as a complication secondary to acute cholecystitis, post-cholecystectomy, trauma or acute pancreatitis.²⁻⁵ Even though pathophysiology of CAP in cholecystitis is unclear, it is hypothesized to occur most likely due to inflammatory damage of the adventitia in cholecystitis. Patients with CAP generally present with varying combinations of upper abdominal pain, jaundice, hemobilia and melena.⁶ The most common imaging modality to diagnose CAP is an arterial phase contrast-enhanced CT scan (CECT), which is readily available in most tertiary care treatment centres. Colour Doppler ultrasound and MRI imaging are other options available.¹ A strong suspicion and a prompt diagnostic analysis will positively affect the outcome of the condition with either interventional radiological or surgical management.^{7,8} We

present a 54-year-old male diagnosed to have cystic artery pseudoaneurysm secondary to acute cholecystitis managed by cholecystectomy.

CASE REPORT

A 54 years old male was referred to our Gastroenterology out-patient department presenting with recurrent right upper quadrant pain, recurrent jaundice and melena. Initial imaging at the outside facility revealed cholelithiasis with evidence of cholecystitis and choledocholithiasis.

Patient was referred to our tertiary center for further evaluation of melena.

On arrival to our hospital, patient was hemodynamically stable. Investigations performed showed Hb of 8 g/dl (Normal-13.2-16.6 g/dl). His liver function tests showed a bilirubin of 1.8.0 mg/dl (0.8-1.0 mg/dl) and alkaline phosphatase of 350 IU (80-240 IU). This revealed

reducing trends in serum bilirubin and alkaline phosphatase, compared to previous levels. Other lab parameters showed the total leucocyte count 18.4×10^3 cells/cc ($4-11 \times 10^3$ cells/cc), normal platelet count and normal prothrombin time.

Ultrasonogram with color Doppler imaging showed cholelithiasis, evidence of cholecystitis and an anechoic structure showing color flow within the gall bladder. Doppler evaluation showed pulsatile flow within the structure suggesting a pseudoaneurysm (Figure 1).

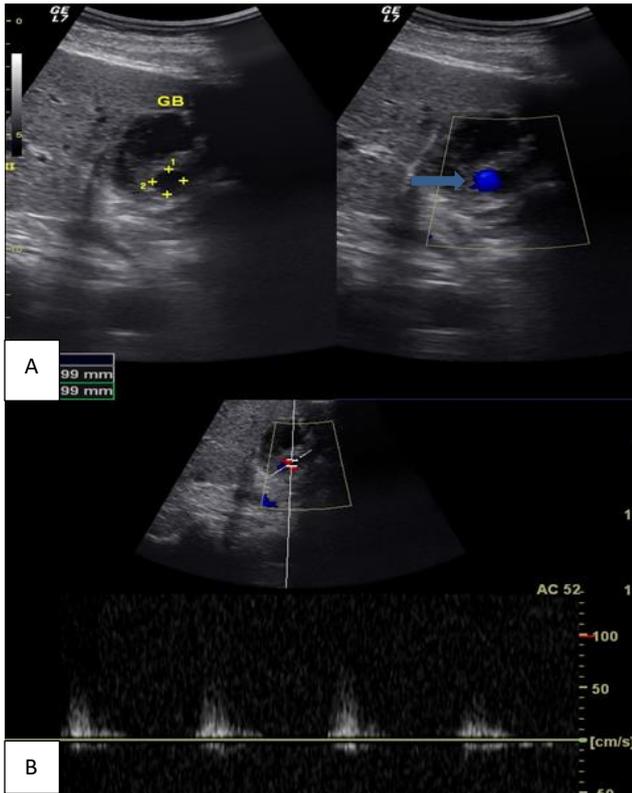


Figure 1 (A and B): USG shows features of cholecystitis with color flow and arterial spectral waveform in the anechoic structure in the gall bladder suggesting a pseudoaneurysm.

Dual phase contrast enhanced computed tomography (CT) (Figure 2) was performed, which revealed an enhanced pooling of contrast on arterial phase consistent with a cystic artery pseudoaneurysm within the gall bladder with adjacent hyperdense contents suggesting blood products. The blood products within the GB suggests a contained rupture of the pseudoaneurysm which can occur intermittently in inflammatory conditions. There was no intra or extrahepatic bile duct dilatation either on sonography or on CT performed in our center.

The imaging findings suggestive of cholelithiasis, acute cholecystitis with cystic artery pseudoaneurysm were discussed in our multidisciplinary departmental meeting with the surgeons and interventional radiologists. As the

patient was hemodynamically stable and the aneurysm was found to be located within the gall bladder, we did not feel the need of endovascular embolisation.

A laparoscopic cholecystectomy was initially planned for the patient, but deferred since our surgeons felt there could be a significant risk of bleeding during the procedure in view of the pseudoaneurysm. It was also expected that the arterial walls would be damaged secondary to repeated inflammation.

An open cholecystectomy was then performed after taking proximal control of the hepatic artery and ligating the cystic artery. A thrombosed pseudoaneurysm with moderate hemorrhagic contents in the gall bladder was identified during the procedure. The patient made an uneventful recovery post-procedure. There was no recurrence of symptoms on follow up.

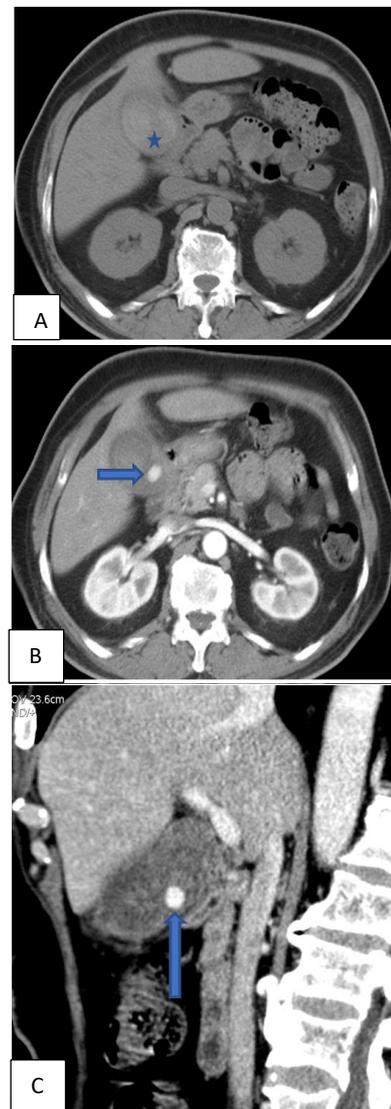


Figure 2 (A-C): Non-contrast CT shows dense contents (asterisk) within the gall bladder. Arterial phase contrast CT shows contrast pooling consistent with a pseudoaneurysm (arrows) within gallbladder.

DISCUSSION

Cystic artery pseudoaneurysms, though uncommon are associated with a significantly high morbidity. It most frequently occurs following an acute cholecystitis or post-cholecystectomy but also occurs rarely following trauma or acute pancreatitis.⁹⁻¹¹ The pathophysiology of CAP in cholecystitis is not clear, though it is likely related to inflammatory damage and weakening of the adventitia with subsequent pseudoaneurysm formation.^{2,4} It has been postulated by other authors that sometimes inflammatory reaction predisposes to early thrombosis of the pseudoaneurysm, preventing its haemorrhagic rupture and associated sequelae.⁷ In addition, cholecystectomy surgeries may contribute further to the development of CAP due to vascular erosion from rough manipulation of the structures, during clip application, or as a result of thermal injury, although the incidence of vascular injury in cholecystectomy is quite low (0.2-0.5%).^{5,6} Our patient developed the pseudoaneurysm secondary to cholecystitis.

Cystic artery pseudoaneurysms usually present with varying combinations of upper abdominal pain, jaundice and melena. Infrequently, all three symptoms occur together and are known as Quinke's triad.¹² The reported incidence of Quinke's triad in patients with haemobilia is 22-35%.³ Other unusual presentation includes pain with localized rupture leading to a subhepatic hematoma.

The first investigation offered for a patient suspected to have a pseudoaneurysm is ultrasonography with color Doppler. It usually demonstrates an anechoic lesion with colour flow, although the drawback of poor localisation exists with ultrasound. A CECT typically demonstrates a hyperdense lesion without contrast, which opacifies in the arterial phase. It also aids in confirming the diagnosis in patients with an equivocal Doppler examination before proceeding for an invasive procedure and may suggest the blood vessel involved.¹³ An additional advantage is the availability of CT imaging in most treatment centres.¹ Digital subtraction hepatic angiography (DSA) was considered the gold standard in delineating the pseudoaneurysms, however, multidetector CT angiography is the modality of choice in the present day for depicting the pseudoaneurysm and also clearly demonstrating the arterial anatomy. Hence we recommend DSA only when an interventional procedure like embolisation is planned. Colour Doppler ultrasound and MR imaging can be reserved for patients with contraindications to the use of intravenous contrast agents.¹

Once the pseudoaneurysm is confirmed, embolization before a cholecystectomy may be useful to ensure the safety of the patient.^{13,14} Transcatheter embolization for stabilization of the patient, followed by an elective cholecystectomy, is a successful management plan, although no guidelines are established in the literature.^{10,15} Following endovascular intervention, open

or laparoscopic cholecystectomy and cystic artery ligation can be performed as an elective procedure. Our patient was hemodynamically stable and the pseudoaneurysm was found to be located within the gall bladder, hence we did not feel the need of endovascular embolisation before a definitive surgery. An Open Cholecystectomy was performed after taking proximal control of the hepatic artery and ligating the cystic artery.

Close monitoring and follow up of these patients is required to ensure treatment success. We suggest an individualised institutional based protocol for treating these patients based on the patient presentation and location of the pseudoaneurysm.

CONCLUSION

Although cystic artery pseudoaneurysm is a rare phenomenon, it occurs as a complication of a very common surgical procedure (i.e., cholecystectomy) or as a sequelae of one of the most common gallbladder conditions (i.e., cholecystitis). CAP is associated with notable levels of morbidity causing significant discomfort to the patient and can be life threatening sometimes. It is therefore of paramount importance for the surgeons to be aware of its presentation, diagnosis, and management.

Funding: No funding sources

Conflict of interest: None declared

Ethical approval: Not required

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Cite this article as: Jaganathan S, Rangarajan M, Jyothirmayi V. Cystic artery pseudoaneurysm: an uncommon complication of a common disease. *Int Surg J* 2022;9:1894-7.