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Massive upper gastrointestinal bleeding due to right hepatic artery pseudoaneurysm secondary to cholecystitis

Case Report

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Abstract: A literature trawl reveals a substantial number of reports on true visceral aneurysms, including the hepatic artery, but only a handful of cases of visceral pseudoaneurysms. The ones in relation to the biliary tree are associated with previous gall bladder surgery and can result in significant gastrointestinal bleeding. There are more than 10 reported cases of cystic artery pseudoaneurysms but a thorough search revealed only two cases in English (1,2) and perhaps one in Japanese literature of right hepatic artery pseudoaneurysm secondary to cholecystitis presenting as massive upper gastrointestinal bleed. We present a probable fourth case in a 52 year old woman with classical clinical/biochemical picture, typical radiological appearance and who underwent successful interventional radiological treatment of this condition.

Keywords: *Pseudo aneurysm • Hepatic artery • Haemobilia • Cholecystitis*

1. Introduction

A literature trawl reveals a substantial number of reports on true visceral aneurysms, including the hepatic artery, but only a handful of cases of visceral pseudo aneurysms. The ones in relation to the biliary tree are associated with previous gall bladder surgery and can result in significant gastrointestinal bleeding. There are more than 10 reported cases of cystic artery pseudo aneurysms but a thorough search revealed only two cases in English [1,2] and perhaps one in Japanese literature of right hepatic artery pseudo aneurysm secondary to cholecystitis presenting as massive upper gastrointestinal bleed.

We present a probable fourth case in a 52- year old woman with classical clinical/biochemical picture, typical radiological appearance and who underwent successful interventional radiological treatment of this condition.

2. Case description

A 52- year old woman was admitted to the Surgical Admissions Unit with one day history of haematochezia, malaena and later developed haemetemesis. She was on the elective waiting list for cholecystectomy for ultrasound proven calculous cholecystitis. She had past medical history of rheumatoid arthritis, right knee replacement and appendicectomy. Her medication included Methotrexate and Non Steroidal Anti-inflammatory Drugs (NSAIDs). She was 20-a-day smoker.

On examination her pulse was 110 per minute and blood pressure 110/80 mmHg.

Clinically she was not jaundiced. There was right upper quadrant abdominal tenderness with no masses felt. Rectal examination showed old blood on fingerstall.

Routine blood results showed Hb of 10.2 g/dL (12.6-18.0), white cell count of 6.9 x10^9/L (4.0-12.0), normal renal function and deranged liver function tests showing

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obstructive picture [Bilirubin: 79 umol/L (0-17), Alkaline Phosphatase: 1449U/L (85-260)]. She was initially managed conservatively with intravenous fluids, proton pump inhibitors and later a blood transfusion due to drop in haemoglobin. In line with local practice, an urgent in-patient gastroscopy and a colonoscopy was organized. Gastrosocpy was normal and uncomplicated sigmoid diverticulosis only was seen on colonoscopy. An ultrasound scan showed thickened oedematous gall bladder and non dilated biliary tree. No other useful information was gained due to abdominal tenderness.

In view of clinical and haematological evidence of ongoing gastrointestinal blood loss, a Computerized Tomography Angiogram (CTA) (Figure 1) was arranged. This showed a 3.8 centimetre pseudo aneurysm arising from the right hepatic artery with surrounding haematoma and cholecystitis with a 3.5 cm stone impacted in the Hartmann's pouch. A subsequent mesenteric angiogram showed bleeding point from right hepatic artery pseudo aneurysm (Figure 2), which was successfully treated with metal coil embolization (Figure 3). She required further blood transfusion as her Hb dropped to 5.1 g/dL.

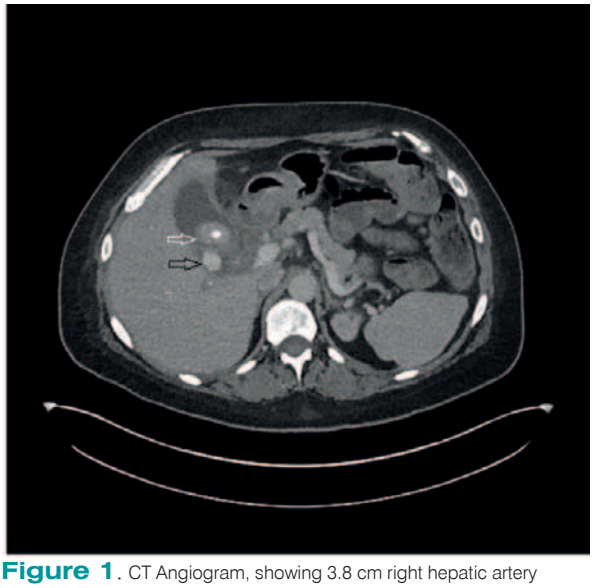

Figure 1. CT Angiogram, showing 3.8 cm right hepatic artery pseudo aneurysm (*black arrow*), surrounding haematoma and cholecystitis with impacted gall stone (*white arrow*).

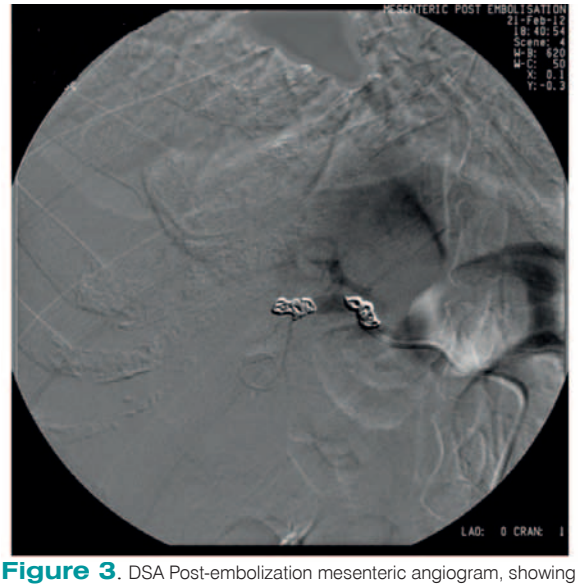

Figure 3. DSA Post-embolization mesenteric angiogram, showing metal coils occluding the right hepatic artery.

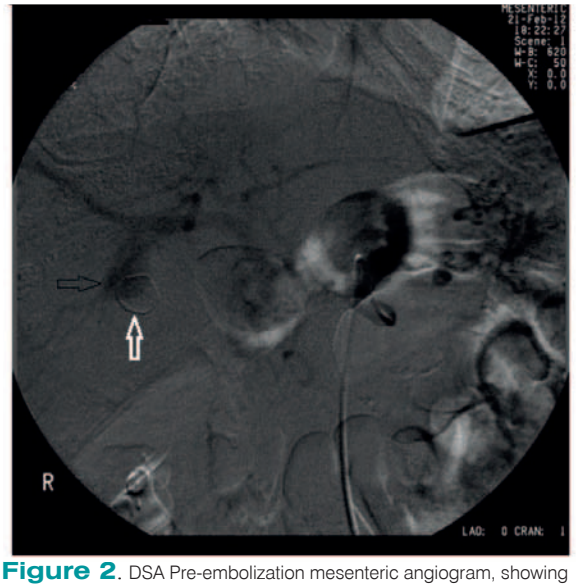

Figure 2. DSA Pre-embolization mesenteric angiogram, showing gall stone (*white arrow*) projected over the aneurysm (*black arrow*).

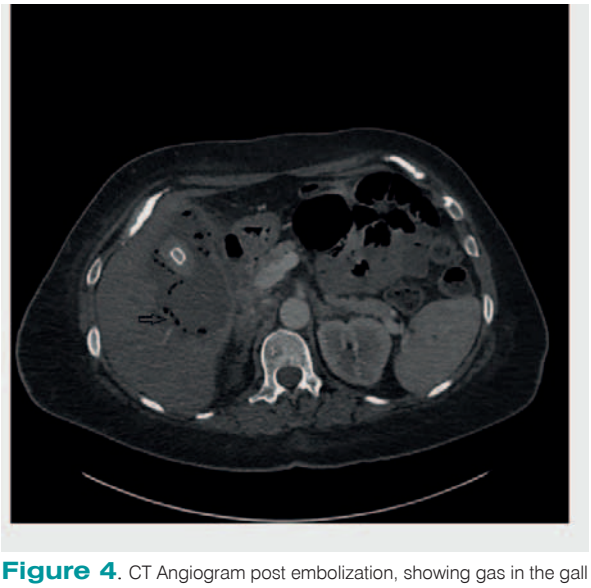

Figure 4. CT Angiogram post embolization, showing gas in the gall bladder wall as well as periphery of the pseudo aneurysm (black arrow). Note the aneurysm remains unenhanced suggesting successful embolization.

A follow up CTA 3 days later showed non-enhancement with contrast of the previously noted pseudo aneurysm suggesting that embolization was successful. However, it showed evidence of gas in the gall bladder wall as well as in the periphery of the thrombosed pseudoaneurysm likely to be due to global post embolization ischaemic changes (Figure 4). At this time patient had a temperature of 38.4° C and a white cell count of 11.4 x10^9/L. There were no further drops in haemoglobin.

In view of its complexity, the case was discussed with and transferred to the regional tertiary care hepatobiliary unit. We later understood that the patient had percutaneous drainage of gall bladder with release of blood stained fluid and pus, which resulted in symptom relief within a short time period. She was discharged home symptom free with drain in-situ. She later underwent an elective uneventful laparoscopic cholecystectomy followed by one-day hospital stay, by the hepatobiliary specialist under private arrangement. On a recent telephone interview 7 months postoperatively she remains symptom free.

3. Discussion

By definition, pseudo aneurysm or false aneurysm is a haematoma formed as a result of slow arterial extravasation. While communicating with the arterial lumen, it is confined external to the arterial wall by surrounding tissues.

Hepatobiliary pseudo aneurysm can result in bleeding into the biliary tree. The first case of bleeding in the biliary tree was described by Francis Glisson in 1654 following a case of stab wound to the liver, whereas Sandblom [3] actually coined the term “haemobilia” in 1948. In modern times the main causes of pseudo aneurysm formation are iatrogenic (medical/surgical/radiological intervention) and, even rarely, localised inflammation. In relation to the biliary tree, the classical presentation is with right upper quadrant pain, upper gastrointestinal bleeding and jaundice. This symptom complex, called Quincke’s Triad was first described circa 1871 [4], is seen in 22% of patients with haemobilia and was present in our patient. The accompanying jaundice could be due to blood clot in the bile duct as bile and blood do not mix. This was evident in our patient whose bilirubin level significantly dropped following successful haemostasis with embolization.

As with any rare disorder, diagnosis requires a high index of suspicion and exclusion of common disease processes.

The initial imaging investigations could include ultrasound scan which not only confirms calculous cholecystitis but also potentially identifies the pseudoaneurysm as a cystic lesion in the porta hepatis area. Doppler mode shows a typical anechoic lesion with pulsatile wave pattern through it. Although appearance on contrast enhanced CT (CECT) scan is diagnostic but role of Contrast Enhanced Magnetic Resonance Angiography (CEMRA) has been described (2); however, latter investigation does not have a high resolution. With the advent of CTA and CEMRA the role of Digital Subtraction Angiography (DSA) is mainly therapeutic. Gastroscopy and colonoscopy have contributing role in the early part of undiagnosed gastrointestinal bleeding.

The management of exsanguinating and cholecystitis associated pseudaneurysms is two fold; first, urgent control of bleeding and second to deal with the source of inflammation i.e. gall bladder. In a stable patient, the former can be achieved by superselective DSA and embolization preferably with stainless steel micro coils which do not get absorbed and produce permanent vascular occlusion [1]. The risks include biliary necrosis, abscess formation and further bleeding. However, if encountered during emergency laparotomy, the options are either exclusion or excision of the aneurysm. In case of successful embolization, the gall bladder treatment can be deferred in unwell patients. However, in the event of ischaemic complications an emergency cholecystectomy may become necessary. Successful endovascular treatment with coronary stent-graft has been described in a case report of iatrogenic right hepatic artery pseudo aneurysm [5]; however, the authors admitted that, although this approach can avoid ischaemic complications of embolization, the long term durability of stent is unknown. Percutaneous cholecystostomy is a viable option for immediate symptom relief in case of superimposed sepsis and was successful in our patient.

4. Conclusion

Hepatic artery pseudo aneurysms in relation to the cholecystitis are extremely rare and can be an exceptional cause of life threatening gastrointestinal haemorrhage. Diagnosis and treatment can be successfully achieved by radiological means in suitable patients. Hepatobiliary ischaemia is a recognized complication of the pseudo aneurysm per se as well as its treatment with angiographic embolization. Follow up imaging is recommended to assess the result of embolization and its potential complication. Timely tertiary care referral is also recommended as these cases may require complex hepato-biliary resection/reconstruction [1].

Authors decleration

KH was responsible for writing the paper, collecting the medical details and the images for publication and contributed to the design of the paper. KH was responsible for the conception of the paper. KH is guarantor of the paper and all authors approved the final manuscript. GW reported the CT scan and performed angiography and embolization.

Competing interest

None declared

Consent

Patient's consent has been obtained and a consent form will be produced upon request.

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