Case Report

Cystic Artery Pseudoaneurysm: A Rare Cause to Obscure Upper Gastrointestinal Bleeding

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ABSTRACT

Obscure gastrointestinal bleeding is a surgical enigma of disastrous proportions. Patient's haemodynamic status often dictates the path of management ranging from endoscopy, embolization and/or surgery. Minority of the cases has failed to identify the exact source of bleeding during endoscopic and imaging techniques. Emergency surgery is warranted in hypovolaemic shock which has failed to respond to fluid and blood resuscitation. We present a 72-year-old male with an obscure upper gastrointestinal bleeding due to ruptured cystic artery pseudoaneurysm and illustrate the rarity of the presentation with successful management.

Keywords: obscure gastrointestinal bleeding, pseudoaneurysm, cystic artery

INTRODUCTION

Upper gastrointestinal bleeding (UGIB) is a common surgical admission to the emergency department. Initial management warrants fluid and blood resuscitation, proton pump inhibitor with subsequent definite intervention by an endoscopy. Rebleeding episode or failure to impede it endoscopically requires angiography and embolization in certain centre especially in developed countries. However, endoscopic and imaging techniques are unsuccessful to locate the source of bleeding in approximately 5% of patients[1]. Hence, it is calamitously labelled as obscure UGIB.

The management ranges from computed tomography of the abdomen, a three-vessel angiogram, red blood cell nuclear scan, small bowel enteroscopy, and ultimately surgical intervention. We present an interesting case of an obscure massive upper gastrointestinal bleeding due to a ruptured cystic artery pseudoaneurysm. This case illustrates the rarity of the presentation and successful management.

CASE PRESENTATION

A 72-year-old man presented to emergency department with a history of syncope and melena. He denied any history of haematemesis, abdominal pain, jaundice and fever to suggest of cholangitis or obstructive jaundice. He has intermittent episodes of biliary colic especially after taking fat-laden food which resolved spontaneously but no hospitalization, accident, endoscopic procedure and surgery before. He was clinically in hypovolemic shock and was significantly pale. Haematological investigation showed a haemoglobin level of 7 g/dL with no evidence of coagulopathy. His total white cell was 12×10^9/L. Following resuscitation, emergency oesophagogastroduodenoscopy (OGDS) was done, however no source of bleeding was identified. Following another subsequent episode of UGIB, a second OGDS was performed in which showing bleeding and pus passing from ampulla of Vater. An endoscopic retrograde cholangiopancreatography (ERCP) was performed and revealed gallstones

with filling defect in the common bile duct (Figures 1a and 1b). Subsequently, computed tomography angiogram (CTA) was preceded but yet failed to identify any source of bleeding.

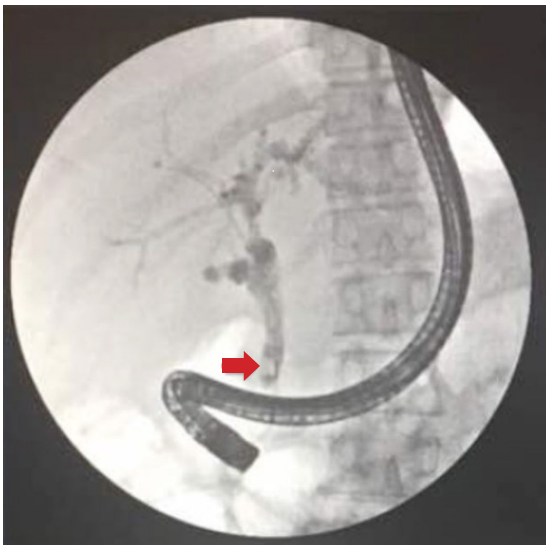

Figure 1a ERCP revealed a filling defect in the common bile duct suggestive of choledocholithiasis (red arrow)

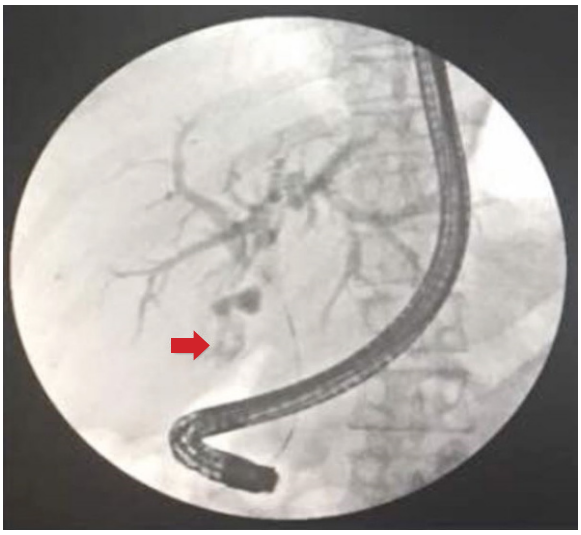

Figure 1b Another view of ERCP showed evidence of gallstones (red arrow)

In view of deteriorating haemoglobin level and further signs of worsening shock, he was subjected to an emergency laparotomy. Intra-operatively, the findings were intense inflammatory changes at Calot's triangle with pseudoaneurysm of the cystic artery. Cholecystectomy with ligation of cystic artery proximal to the pseudoaneurysm was undertaken. Post-operatively, the patient recovered well without any further blood loss and he was discharged successfully. Histopathological examination of the gallbladder was consistent with chronic inflammatory changes (Figures 2a and 2b).

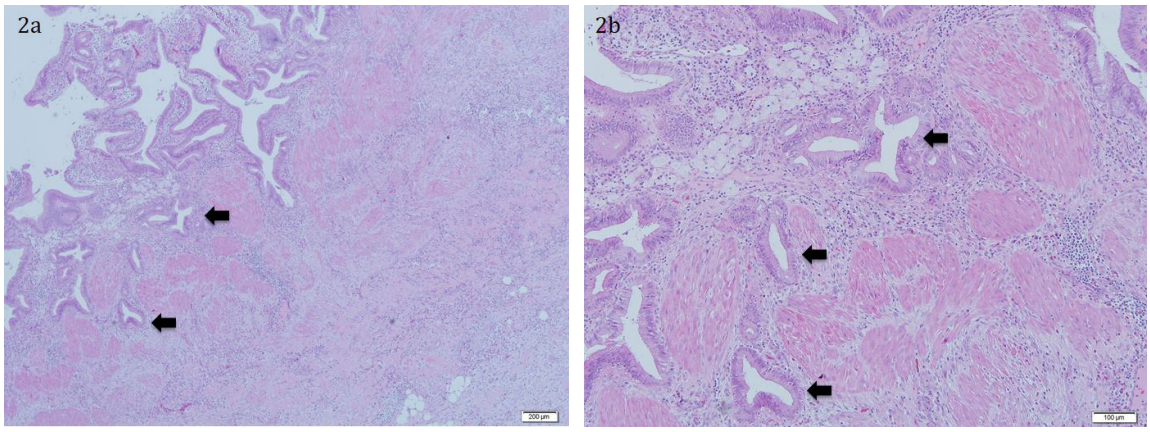

Figure 2 Histopathological examination of the gallbladder showed features of chronic cholecystitis as evidenced by presence of lymphocytic infiltration with Rokitansky-Aschoff sinuses (black arrows) at ×4 magnification (2a) and at ×10 magnification (2b)

DISCUSSION

Obscure UGIB has a variety of aetiologies. One of them is due to cystic artery pseudoaneurysm. The number of reported cases are scarce with only 24 cases being reported up to date[2]. Acute cholecystitis is one of the causative factors of a cystic artery pseudoeurysm to occur. It can lead to gangrenous necrosis which has a very high mortality rate[3]. The numbers of cystic artery pseudoaneurysm that occur are low in contrast to the high frequency of cholecystitis cases occurring worldwide. This possibly is a reflection of the pathological process that occurs with inflammation whereby the vessel is occluded earlier in the course of the disease[4].

Anatomically, cystic artery is a branch of right hepatic artery. A normal hepatic artery anatomy occurs in 89% of the population[5]. There are various common variants; ranging from a completely replaced hepatic arterial system with a gastroduodenal artery coming from the celiac axis and even from the superior mesenteric artery[6]. These anatomical variants represent the need for angiography upon development of an obscure intestinal bleed. In our case however, the angiography had failed to yield a positive result.

Patients typically present with Quinke's triad (upper quadrant pain, obstructive jaundice and gastrointestinal bleeding) to suggest of acute cholecystitis. However in our case, the patient directly presented with symptoms of UGIB. Even with the help of endoscopy, yet no obvious cause of the bleeding could be determined. An ERCP showed presence of gallstones that could have eroded chronically into the cystic artery. The eroded vessel had led to a bleeding in the biliary tree and subsequently causing UGIB. Francis Glison first described this presentation of UGIB in 1993[7]. A few decades have passed and these cases are still exceptional.

The presentation was further unusual as the patient denied any symptoms suggestive of cholecystitis that could possibly be related to the ruptured cystic artery pseudoaneurysm. The passage of pus and blood as seen during the ERCP contributed to a diagnostic dilemma. This event could possibly lead to a misdiagnosis and delay of treatment. The surgical specimen however showed inflammatory changes that explained the probable causative factor for the pseudoaneurysm formation.

Endovascular intervention remains the gold standard in managing cystic artery pseudoaneurysm despite its possible complication profiles such as hepatobiliary necrosis, bleeding, abscess formation and contrast related complications such as nephropathy and allergic reaction[4, 8]. However in cases where the presentation are vague and the diagnosis is in

doubt; surgical exploration, cholecystectomy and ligation of the pseudoaneurysm still prove to be an effective and safe way to treat this condition.

CONCLUSION

Any episode of non-variceal UGIB requires standard management which is acceptable worldwide. Resuscitation using fluid and blood transfusion, initiation of proton pump inhibitor and OGDS are imperative. If the diagnosis is dubious, step-up modalities are required such as angiography, ERCP and lastly surgical exploration.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this case.

CONSENTS

Written informed consent was obtained from the patient to publish the case. A copy of written consent is available for review by the Chief Editor.

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