



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## A rare cause of upper gastrointestinal haemorrhage: Ruptured cystic artery pseudoaneurysm with concurrent cholecystojejunal fistula☆☆

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## ARTICLE INFO

## Article history:

Received 2 May 2013

Received in revised form 6 November 2013

Accepted 7 November 2013

Available online 20 November 2013

## Keywords:

Cystic Artery

Pseudoaneurysm

Cholecystojejunal fistula

Cholecystoenteric fistula

## ABSTRACT

**INTRODUCTION** Cystic artery pseudoaneurysms and cholecystoenteric fistulae represent two rare complications of gallstone disease.

**PRESENTATION OF CASE** An 86 year old male presented to the emergency department with obstructive jaundice, RUQ pain and subsequent upper gastrointestinal bleeding. Upper GI endoscopy revealed bleeding from the medial wall of the second part of the duodenum and a contrast-enhanced computed tomography scan revealed a cystic artery pseudoaneurysm, concurrent cholecystojejunal fistula and gallstone ileus. This patient was successfully managed surgically with open subtotal cholecystectomy, pseudoaneurysm resection and fistula repair.

**DISCUSSION** To date there are very few cases describing haemobilia resulting from a bleeding cystic artery pseudoaneurysm. This report is the first to describe upper gastrointestinal bleeding as a consequence of two synchronous rare pathologies: a ruptured cystic artery pseudoaneurysm causing haemobilia and bleeding through a concurrent cholecystojejunal fistula.

**CONCLUSION** Through this case, we stress the importance of accurate and early diagnosis through ultrasonography, endoscopy, and contrast-enhanced CT imaging and emphasise that haemobilia should be included in the differential diagnosis of anyone presenting with upper gastrointestinal bleeding. We have demonstrated the success of surgical management alone in the treatment of such a case, but accept that consideration of combined therapeutic approach with angiography be given in the first instance, when available and clinically indicated.

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## 1. Introduction

Visceral pseudoaneurysms and cholecystoenteric fistulae represent two rare complications of gallstone disease. We present a unique case of a patient presenting with abdominal pain, obstructive jaundice and upper gastrointestinal haemorrhage resulting from a ruptured cystic artery pseudoaneurysm and concomitant cholecystojejunal fistula.

## 2. Case presentation

An 86-year-old male presented to the Emergency Department with a two-day history of colicky right upper quadrant (RUQ) pain

associated with bilious vomiting. Relevant background included hypertension, diabetes mellitus and hypercholesterolaemia. On examination, the patient was icteric with an associated pyrexia of 38 °C, hypotension and tachycardia. Abdominal examination revealed mild tenderness in the RUQ.

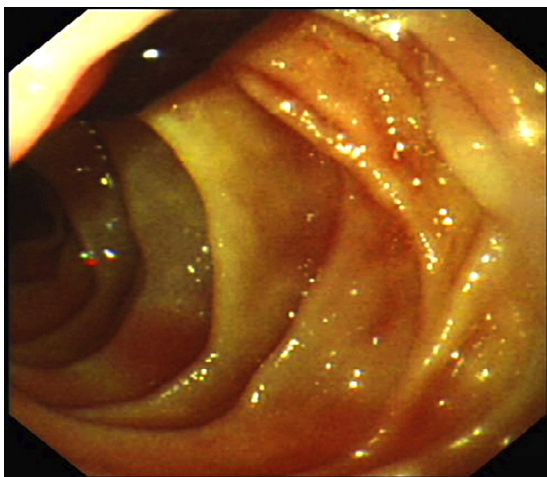
Laboratory investigation revealed raised inflammatory markers with a white blood cell count of  $24.0 \times 10^9 \text{ L}^{-1}$  and C-reactive protein of 95 mg/L (Normal range: <7.5 mg/L). Liver function tests (LFTs) were also deranged with an alkaline phosphatase of 450  $\mu\text{L}$  (46–111  $\mu\text{L}$ ), Alanine Transaminase of 142  $\mu\text{L}$  (<45  $\mu\text{L}$ ) and Bilirubin of 60  $\mu\text{mol/L}$  (<20  $\mu\text{mol/L}$ ). An ultrasound examination revealed a 5.7 cm calculus within the gallbladder surrounded by heterogeneous material; a diagnosis of cholangitis was made. The patient received fluid resuscitation with intravenous crystalloid and was commenced on broad-spectrum antibiotics.

Subsequently, the patient passed a significant volume of malaena and fresh blood per rectum (PR), associated with cardiovascular instability. Emergency oesophagoduodenoscopy (OGD) revealed fresh blood in the second part of the duodenum and a tubular clot adherent to the medial wall (Fig. 1). This was assumed to be originating from a duodenal ulcer and apparent haemostasis was achieved with injection of 15 ml of 1:10,000 adrenaline. However, the patient continued to pass malaena and PR blood following the procedure. As such, a contrast-enhanced computed

☆☆ This paper has not been presented previously as an abstract in any congress or symposium.

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**Fig. 1.** Second part of the duodenum viewed at OGD with a tubular clot adherent to medial wall, likely to be originating from the ampulla of Vater.

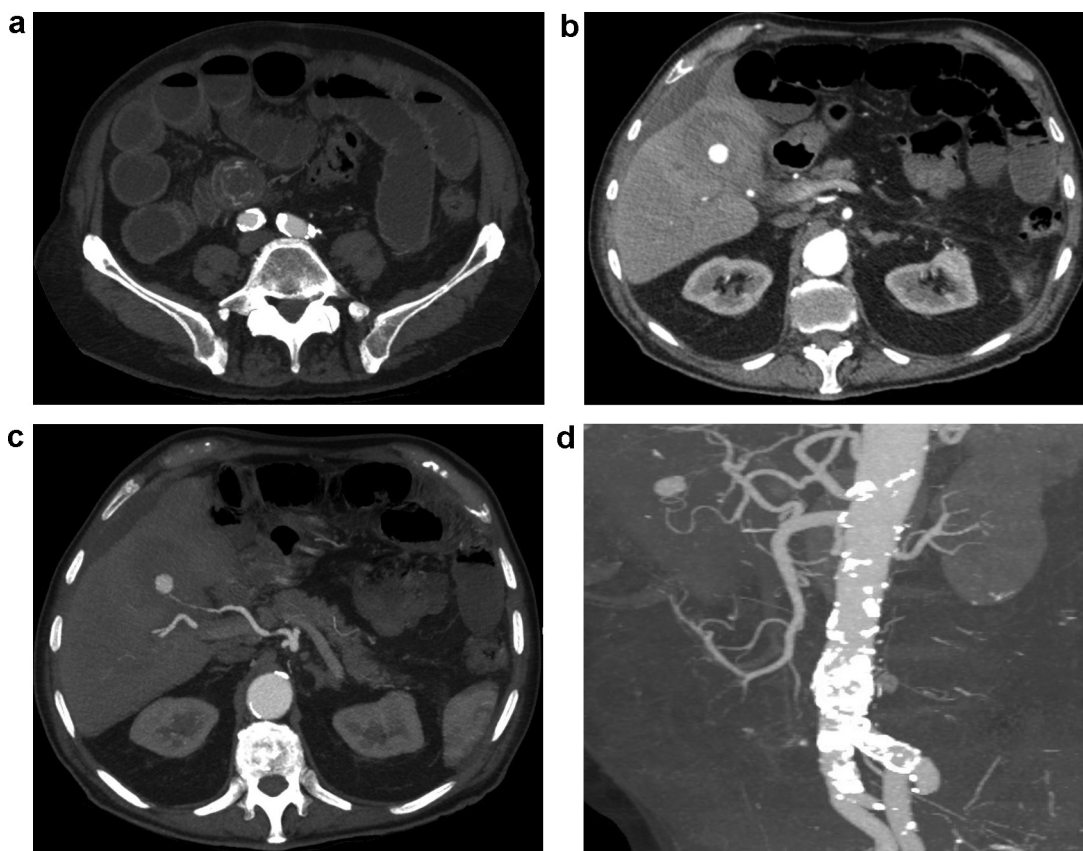
tomography (CT) scan was performed, demonstrating: (1) small bowel obstruction secondary to a gallstone impacted in the distal ileum (Fig. 2a) (2) a thick-walled and inflamed gallbladder with evidence of recent intra-cholecystic bleeding (Fig. 2b), and (3) a 12 mm pseudoaneurysm emanating from the cystic artery (Fig. 2c and d). In retrospect, the blood clot seen at OGD was likely to be haemobilia originating from the ampulla of Vater.

The patient underwent emergency laparoscopy and the gallbladder was found to be necrotic and surrounded with dense adhesions. Division of these exposed a cholecysto-jejunal fistula

containing clotted blood that was extending into the gallbladder. Evacuation of this clot revealed a 1 cm × 2 cm pseudoaneurysm originating from the cystic artery, which began to bleed during dissection and the procedure was subsequently converted to an open laparotomy via a Kocher incision. A subtotal cholecystectomy was performed with oversewing of Hartmanns pouch and the aneurysm was successfully resected from the gallbladder fossa. The diseased jejunal loop was mobilised and the defect from the fistula was oversewn. Despite detailed examination of the small bowel, the gallstone located on CT could not be identified within the terminal ileum. The laparotomy wound was therefore closed under the assumption that the stone had passed spontaneously. The patient was discharged following a fifteen-day inpatient stay and remained well at six-week outpatient follow-up.

### 3. Discussion

Cystic artery pseudoaneurysms remain a rare cause of upper gastrointestinal bleeding, with only twenty-two documented cases in the English literature.<sup>1,2</sup> They develop primarily as a consequence of adventitial damage and thrombosis of the vasa vasorum, resulting in damage to the muscular and elastic components of the media and intima with ensuing extravasation of arterial blood, progressive enlargement and eventual rupture, as governed by the Law of Laplace.<sup>2–4,8</sup> This can occur secondary to inflammatory conditions (e.g. cholecystitis, pancreatitis), malignancy, biliary tract manipulation or trauma. Formation may be further accelerated by patient factors, such as atherosclerosis, hypertension, bleeding disorders and vasculitis.<sup>1–7</sup> Cystic artery pseudoaneurysms tend to enlarge and erode into the gallbladder and adjacent biliary tree with



**Fig. 2.** Axial computed tomographic images with contrast demonstrating (a) small bowel obstruction secondary to a gallstone impacted in the distal ileum, (b) a thick-walled, inflamed gallbladder with evidence of recent intra-cholecystic bleeding, seen to be originating from (c) a 12 mm cystic artery pseudoaneurysm. This cystic artery pseudoaneurysm was further defined on CT angiography (d).

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