CASE REPORT

Pseudoaneurysm of anomalous cystic artery due to calculous cholecystitis

Varsha Kulkarni,[1] Hemant Deshmukh,[2] Rahul Gupta[3]

[1]Department of General Surgery, Seth GSMC & KEMH, Mumbai, Maharashtra, India
[2]Department of Radiology, Seth GSMC & KEMH, Mumbai, Maharashtra, India
[3]Department of General Surgery, PGIMER, Chandigarh, India

Correspondence to
Dr Rahul Gupta,
rahul.g.85@gmail.com

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SUMMARY

Pseudoaneurysm of the cystic artery is a rare cause of haemobilia resulting from either an inflammatory process in the abdomen or abdominal trauma. We report a case of a patient with chronic calculous cholecystitis associated with a pseudoaneurysm arising from an anomalous cystic artery who presented with haemobilia. The patient was managed successfully with multimodality treatment that included angioembolisation of the pseudoaneurysm and stenting of the common bile duct to relieve jaundice followed by elective open cholecystectomy.

BACKGROUND

Gastrointestinal bleeding from the biliary tree (haemobilia) is an uncommon event. Previously, the most common cause of this disease was traumatic hepatobiliary injury, however, iatrogenic trauma caused by percutaneous and endoscopic hepatobiliary procedures is now the cause in two-third of haemobilia cases.[1] Other causes are gallstones, inflammatory diseases, vascular malformations and neoplastic disease.[1] These aneurysms can be either congenital or acquired. Pseudoaneurysms due to cholecystitis are very rare. We report a rare case of haemobilia due to a pseudoaneurysm of an anomalous cystic artery secondary to chronic calculus cholecystitis with Mirrizi's syndrome.

CASE PRESENTATION

A 55-year-old man presented to the emergency department of our tertiary referral hospital with a 4-month history of repeated episodes of jaundice, right upper abdominal pain and malena. One year earlier the patient had been diagnosed with gall stones. On clinical examination the patient was pale and icteric, with no significant abdominal signs. Per rectal examination revealed malena.

INVESTIGATIONS

Blood investigations showed anaemia with direct hyperbilirubinaemia. Abdominal ultrasound revealed gallstones with mild central intrahepatic biliary radicle dilation.

The contrast-enhanced CT of the abdomen showed a pseudoaneurysm arising from the medial aspect of the gallbladder wall with a mass suspected to be a stone in the gallbladder (figure 1).

MR cholangiography revealed a pseudoaneurysm of the cystic artery (figure 2A,B) with blood clots, a stone within the gallbladder and type I Mirrizi's syndrome (figure 2C). The cystic artery bearing the pseudoaneurysm was found to originate directly from the common hepatic artery on selective hepatic catheterisation (figure 3A). Early branching of the left hepatic artery from the common hepatic artery was also seen (figure 3A).

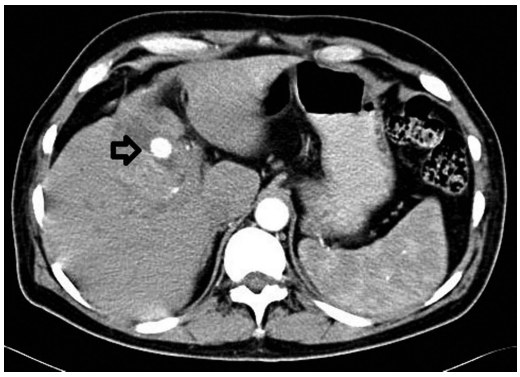

Figure 1 CT angiography showing cystic artery pseudoaneurysm arising from the gallbladder wall.

DIFFERENTIAL DIAGNOSIS

- Cholelithiasis with choledocholithiasis
- Mirrizi's syndrome
- Hepatic artery pseudoaneurysm

TREATMENT

Super selective catheterisation with microcoil embolisation of the anomalous cystic artery was performed resulting in occlusion of the pseudoaneurysm (figure 3B).

Endoscopic retrograde cholangiography (ERC) was performed for direct hyperbilirubinaemia. On ERC, there was passage of blood clots from the ampulla of Vater with extrinsic compression on the mid-portion of the common bile duct (CBD) without any filling defects. CBD stenting was performed to relieve jaundice.

Six days later the patient underwent elective open subtotal cholecystectomy with under-running of the cystic artery pseudoaneurysm. Intraoperatively, the gallbladder was thickened, distended and intrahepatic, and contained pus; it also contained a single large stone. There were dense adhesions in Calot's triangle. An ovoid vascular lesion was found projecting into the lumen of the gallbladder from the posterosuperior wall of its body (figure 4). The gallbladder wall around it was left behind as it was densely adhered to the liver.

OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative recovery with regression of symptoms including jaundice

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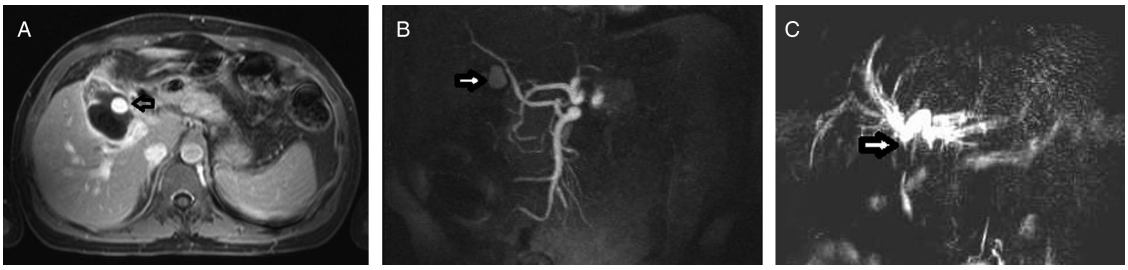
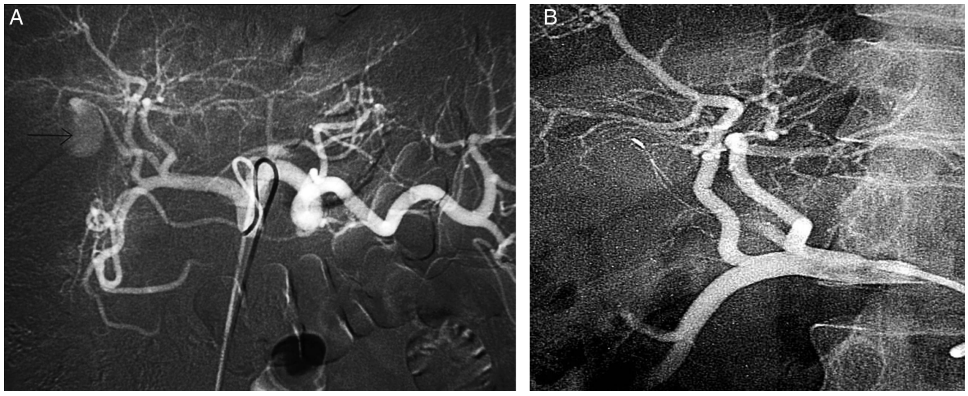


Figure 2 MR angiography of the abdomen (A) cystic artery pseudoaneurysm on axial images, (B) cystic artery pseudoaneurysm on sagittal images, (C) MR cholangiopancreatography showing mid-common bile duct narrowing with intrahepatic biliary radical dilatations.

Figure 3 Digital subtraction angiography (A) before embolisation showing anomalous cystic artery with pseudoaneurysm at its tip, (B) after embolisation.



and malena. Histopathology revealed acute on chronic cholecystitis. On follow-up after 2 years he was operated for an inguinal hernia and since then he has been asymptomatic.

DISCUSSION

The association of upper gastrointestinal bleeding with signs of biliary disorders should suggest haemobilia. Haemobilia is usually associated with Quincke's triad of biliary colic (occurring in 70% of patients), jaundice (exhibited in 60% of cases) and gastrointestinal bleeding, which is present in all patients. The complete classic triad is found in about 22% of patients[1] with acute bleeding first causing biliary colic followed by haematemesis or malena, which in turn leads to pain relief.

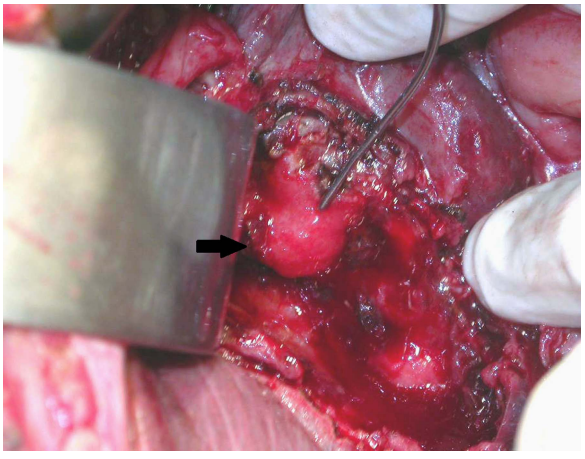


Figure 4 Intraoperative photograph of pseudoaneurysm during open cholecystectomy.

A cystic artery pseudoaneurysm due to an inflammatory process is a rare cause of haemobilia; an extensive search identified only 58 published cases (table 1).

The exact cause of association between pseudoaneurysms and inflammation is not clear but it has been hypothesised that the inflammatory process leads to ulceration of the gallbladder wall and partial erosion of the elastic and muscular components of the vascular wall, leading to the development and rupture of the pseudoaneurysm.[2] Despite cholecystitis being common, the rarity of cystic artery pseudoaneurysms can be explained by early thrombosis of the cystic artery due to inflammation.[3]

Cystic artery pseudoaneurysms can rupture in the biliary tree[4] or rarely into the peritoneum.[5] A high index of suspicion is required for diagnosis. Abdominal colour Doppler ultrasound can be an effective initial diagnostic tool but lacks high sensitivity.[4] Endoscopy is helpful in diagnosing haemobilia in a patient with upper gastrointestinal bleeding. MRI and CT scan can identify the underlying pathology such as stones, cholecystitis, neoplasms and vascular abnormalities. However, selective hepatic angiography is the technique of choice in the management of haemobilia of any cause and for suspected pseudoaneurysms in particular, due to its high diagnostic accuracy and therapeutic potential.[1 6] It is useful especially in elderly, haemodynamically unstable patients unfit for surgery, in stopping the active bleeding. Nevertheless, angiography has certain diagnostic limitations for the variable flow rate and intermittent bleeding, and also when there are hepatic artery abnormalities or when there has been previous manipulation. Despite there being reports of gallbladder infarction after hepatic artery embolisation with occlusion of the cystic artery, there has been no report of gallbladder gangrene, to date, after embolisation of the cystic artery pseudoaneurysm, probably due to collateral blood supply from the epicholedochal artery.[7]

Table 1 Summary of cases of cystic artery pseudoaneurysm secondary to inflammation reported in literature

Serial number	Author	Year	Journal	Aetiology	Presentation	Management
1	Glaysher *et al*	2014	*International Journal of Surgery Case Reports*	Calculous cholecystitis with cholecystoenteric fistula	Jaundice+pain+malena	Open subtotal cholecystectomy
2	Suzuki *et al*	2013	*Clinical Journal of Gastroenterology*	Acute calculous cholecystitis	Epigastric pain+jaundice	Open cholecystectomy
3	Nana *et al*	2013	*International Journal of Surgery Case Reports*	Acute cholecystitis Acute cholecystitis	Quinke triad Haemobilia+pain	TAE+lap cholecystectomy TAE
4	Fung *et al*	2013	*Scottish Medical Journal*	Acute cholecystitis	Haemobilia	–
5	Dewachter *et al*	2012	*Journal Belge de Radiologie - Belgisch Tijdschrift voor Radiologi*	Acute calculous cholecystitis	Epigastric pain+nausea	Lap cholecystectomy+resection of pseudoaneurysm
6	Chong *et al*	2012	*Canadian Association of Radiologists Journal*	–	Fever+epigastric pain+vomiting	–
7	Komatsu *et al*	2011	*Journal of Medical Cases*	Acute calculous cholecystitis	Epigastic pain+haematemesis +malena	TAE
8	Anand *et al*	2011	*Annals of Gastroenterology*	Idiopathic	RUQ pain+haematemesis +malena	Cholecystectomy
9	Nkwam *et al*	2010	*Journal of Surgical Case Reports*	Acute calculous cholecystitis	RHC pain	TAE
10	Desai *et al*	2010	*Radiology Case*	Chronic calculous cholecystitis	Epigastic pain+malena	TAE
11	Ahmed *et al*	2010	*The British Journal of Radiology*	Xanthogranulomatous cholecystitis	RHC pain+lump	TAE+open cholecystectomy
12	Leung *et al*	2010	*Hong Kong Medical Journal*	Acute cholecystitis	Haematochezia	TAE
13	Hague *et al*	2010	*CardioVascular and Interventional Radiology*	Acute calculous cholecystitis Acute calculous cholecystitis –	RHC pain RHC pain Epigastric pain+malena	TAE TAE TAE
14	Siddique *et al*	2011	*BMJ Case Reports*	Acute xanthogranulomatous cholecystitis	Epigastric pain+fever	TAE+elective open cholecystectomy
15	Mullen *et al*	2009	*CardioVascular and Interventional Radiology*	Acute calculous cholecystitis Acute calculous cholecystitis	RHC pain+malena RHC pain	TAE TAE
16	Sousa *et al*	2009	*Gastroentérologie Clinique et Biologique*	Acute calculous cholecystitis	Epigastric pain+haematemesis	Cholecystectomy
17	Radouane *et al*	2008	*Journal of Radiology*	Calculous cholecystitis	Not known	
18	Machida *et al*	2008	*Radiation Medicine*	Acute calculus cholecystitis	RUQ pain	Cholecystectomy
19	Al' Aref *et al*	2008	*Hepatobiliary & Pancreatic Diseases International*	Idiopathic	RUQ pain+malena	TAE
20	Shimada *et al*	2008	*Digestive Surgery*	Xanthogranulomatous cholecystitis	Jaundice	TAE—extended right hepatectomy
21	Ghoz *et al*	2007	*Hepatobiliary & Pancreatic Diseases International*	Acute on chronic cholecystitis	Abdominal pain+haematemesis	TAE+cholecystectomy
22	Akatsu *et al*	2007	*Surgery Today*	Acute cholecystitis	RUQ pain+jaundice	Cholecystectomy
23	Saluja *et al*	2007	*BMC Gastroenterology*	Acute calculus cholecystitis	Haematemesis+Malena	TAE+partial cholecystectomy
24	Chun-Jung Lin *et al*	2007	*Chinese Journal of Radiology*	Acalculous cholecystitis	Joundice+abdominal pain	TAE+cholecystectomy
25	Pérez-Castrillón *et al*	2006	*Endoscopy*	Acute cholecystitis	Abdominal pain+anaemia	TAE
26	Lee	2006	*Clinical Radiology*	Chronic cholecystitis	Lower GI bleed	Not known
27	Sibulesky *et al*	2006	*American Journal of Surgery*	Calculous cholecystitis	RHC pain+malena	Open cholecystectomy
28	Joyce *et al*	2006	*Irish Journal of Medical Sciences*	Acute on chronic cholecystitis	Haemobilia	Cholecystectomy
29	Oueslati *et al*	2005	*Journal de Chirurgie*	Idiopathic	Haemobilia	Cholecystectomy
30	Morioka *et al*	2004	*Journal of Gastroenterology & Hepatology*	Chronic calculus cholecystitis	Haemobilia	Cholecystectomy
31	Gutierrez *et al*	2004	*American Journal of Surgery*	Acute calculus cholecystitis	RHC pain+malena	TAE failed f/b open cholecystectomy
32	Hiroshi *et al*	2002	*Journal of Biliary Tract Pancreas*		Dyspnoea+malena	TAE
33	Maeda *et al*	2002	*Journal of Hepato-Biliary-Pancreatic Surgery*	Calculous cholecystitis	Epigastric pain+jaundice	TAE+elective open cholecystectomy
34	AA Palejwala *et al*	2000	*CME Journal Gastroenterology, Hepatology and Nutrition*	Acute cholecystits	RUQ pain+haematemesis +jaundice	Cholecystectomy

Continued

Table 1 Continued

Serial number	Author	Year	Journal	Aetiology	Presentation	Management
35	Delgadillo *et al*	1999	*Journal of Vascular & Interventional Radiology*	Acute pancreatitis	Haemobilia	TAE
36	dePerrot *et al*	1999	*The British Journal of Surgery*	Pancreatitis	Epigastric pain	Surgery
37	Kaman *et al*	1998	*American Journal of Gastroenterology*	Acute calculous cholecystitis	Haemobilia	Cholecystectomy with ligation of pseudoaneurysm
38	Kirchgatterer *et al*	1998	*Wiener Klinische Wochenschrift*	Chronic calculous cholecystitis	Haemobilia	Cholecystectomy
39	England *et al*	1998	*Clinical Radiology*	Acute calculous cholecystitis	Haemobilia	Open cholecystectomy f/b TAE
40	Miura *et al*	1998	*Nippon Shokakibyo Gakkai Zasshi*	–	Haemobilia	–
41	Ritz *et al*	1997	*Journal de Chirurgie*	Acute calculous cholecystitis	Haemobilia	Cholecystectomy
42	Matsuba *et al*	1996	*Japanese Journal of Gastroenterology Surgery*	Cholecystitis	RUQ pain	Cholecystectomy
43	Nakajima *et al*	1996	*Journal of Gastroenterology*	Acute calculous cholecystitis	Haemobilia	Cholecystectomy
44	Barba *et al*	1994	*Canadian Journal of Surgery*	Cholecystitis	Haemobilia	Cholecystectomy with ligation of pseudoaneurysm
45	Read *et al*	1991	*ANZ Journal of Surgery*	Cholecystitis	Haemobilia	Cholecystectomy
46	Strickland *et al*	1991	*CardioVascular and Interventional Radiology*	Calculous cholecystitis	Haemobilia	–
47	Smague *et al*	1990	*Journal* de *Chirurgie*	Acute cholecystits	Upper abdominal pain +haematemesis	Cholecystectomy
48	Read *et al*	1990	*ANZ Journal of Surgery*	Chronic calculous cholecystitis	Haemobilia	Cholecystectomy
49	Wu *et al*	1988	*Acta Chirurgica Scandinavica*	Acute cholecystitis	–	Surgery
50	Rhee JW *et al*	1987	*New York State Journal of Medicine*	Acute calculous cholecystitis	RUQ pain+fever	Cholecystectomy
51	Reddy *et al*	1983	*Southern Medical Journal*	Cholecystitis	Haemobilia	Cholecystectomy
52	Glazer *et al*	1980	Journal *of Radiology*	Postcholecystectomy	UGI bleed	–
53	Hakami *et al*	1976	*American Journal of Proctology*	? Cholecystitis	Haemobilia	Cholecystectomy
54	Devin *et al*	1971	*Journal* de *Chirurgie*	Acute pancreatitis	Epigastric pain, jaundice, malena	Partial cholecystectomy

?, suspected; f/b, followed by; GI, gastrointestinal; RHC, right hypochondrium; RUQ, right upper quadrant; TAE, transarterial embolisation; UGI, upper GI.

In our patient, the cystic and left hepatic arteries were arising from the common hepatic artery. These anomalies have been described in the literature but are very uncommon.[8 9]

Haemobilia is known to cause biliary obstruction due to blood clots leading to jaundice, as seen in our case, which can be relieved by endoscopic drainage with stenting or nasobiliary drainage.[10]

Maeda *et al*[5] were the first to report successful management of a cystic artery aneurysm by a combined approach: embolisation of the cystic artery to stabilise the patient, followed by cholecystectomy at a later, safer time. It seems a logical combination as the cause of inflammation, the stone, needs to be treated as well. Our case was also managed similarly with success.

Learning points

- In patients with gallstone disease, presence of malena should raise suspicion of a pseudoaneurysm.
- A cystic artery pseudoaneurysm can be successfully managed by angioembolisation and open total/subtotal cholecystectomy.
- Patients with a cystic artery pseudoaneurysm not fit for surgical intervention can be managed by embolisation alone.

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Competing interests None.

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