

Case report

Pseudoaneurysm of the cystic artery associated with upper gastrointestinal bleeding

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Abstract: Pseudoaneurysm of the cystic artery is a cause of hemobilia, and is extremely rare, with only eight cases having been reported in the world literature. We report a case of pseudoaneurysm of the cystic artery in a 72-year-old Japanese man. The patient experienced epigastric pain and melena, and was found to have jaundice and liver dysfunction. Repeated gastroendoscopy did not reveal the cause of the alimentary tract bleeding; however, color-Doppler ultrasonography detected an aneurysm of the cystic artery in the gallbladder. Selective hepatic arteriography demonstrated that the posterior branch of the cystic artery was markedly dilated and that an aneurysm had formed in the midst of the artery. We diagnosed hemobilia due to the pseudoaneurysm of the cystic artery, and associated gastrointestinal bleeding. Cholecystectomy was performed immediately. Pathologically, the gallbladder showed acute calculous cholecystitis. This case emphasizes the importance of including hemobilia in the differential diagnosis whenever gastrointestinal bleeding is associated with signs of biliary disorder; color-Doppler imaging is a favorable modality for the diagnosis of pseudoaneurysm of the cystic artery.

Key words: pseudoaneurysm, cystic artery, color-Doppler, hepatic arteriography

Introduction

Hematemesis or melena is a frequent clinical symptom of upper gastrointestinal bleeding. Its severity ranges widely and patients may exhibit such serious conditions as hypovolemic shock. Since more than 90% of upper

gastrointestinal bleeding results from diseases of the alimentary tract such as peptic ulcers and varices,¹ most cases can be easily diagnosed by emergent endoscopy. In patients with bleeding from regions other than the alimentary tract, it is difficult to confirm the precise source of bleeding by endoscopy alone. Hemobilia, hemorrhage into the biliary tract, is a rare cause of upper gastrointestinal bleeding,¹ and is secondary to several other causes, such as hepatic trauma (accidental or iatrogenic), aneurysm of the hepatic artery, tumors of the biliary tract, gallstone disease, and inflammation of the liver.^{2,3} Pseudoaneurysm of the cystic artery is one of the causes of hemobilia, and is extremely rare, with only eight cases having been reported in the world literature.⁴⁻¹¹ The condition may be associated with inflammation of the gallbladder,^{6,10} however, its cause remains obscure. Here we present a rare case of hemorrhage from the gallbladder caused by pseudoaneurysm of the cystic artery.

Case report

A 72-year-old Japanese man experienced melena and consulted his local hospital on January 26, 1995. He had had intermittent epigastric pain for a few weeks, and the pain had worsened after meals. At the hospital, jaundice and liver dysfunction were observed and he was therefore referred for further evaluation and admitted to our hospital, on January 30. He had a history of acute appendicitis at age 20 and intestinal tuberculosis at 32, and had no history of abdominal trauma.

The patient's height was 152 cm and weight was 42 kg. Body temperature on admission was 36.8°C and blood pressure was 138/60 mmHg. On physical examination the bulbar conjunctiva showed slight icterus and the palpebral conjunctiva showed anemia. Inspection of the abdomen was unremarkable, except for an operation scar in the lower part; there was no tenderness over

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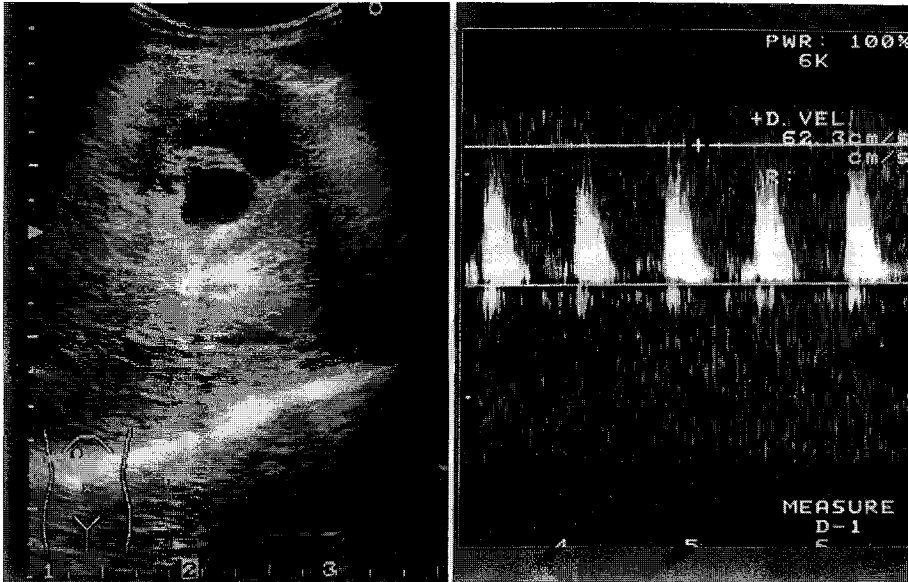


Fig. 1. Ultrasonography of the gallbladder. Abdominal ultrasonography shows distended gallbladder with an echolucent cystic lesion attached to the thickened wall. Doppler imaging detected pulsatile waves in this cystic lesion

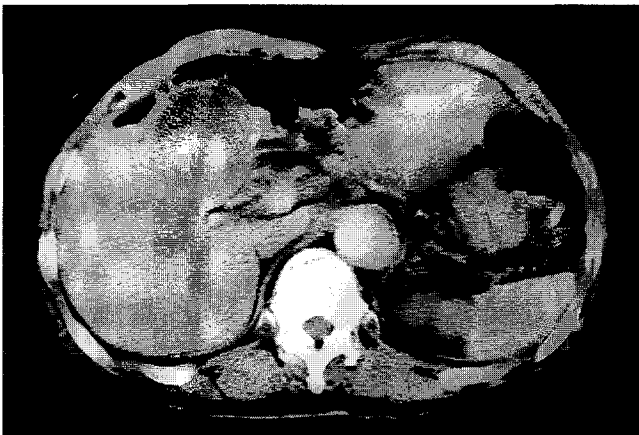


Fig. 2. Enhanced computed tomographic (CT) scan of the gallbladder. CT scan demonstrates a low-density lesion in the gallbladder, which is strongly enhanced by infusion of contrast medium

the abdomen. Physical examination was otherwise unremarkable.

Hematological values were: white blood cell count $6500/\text{mm}^3$, red blood cell $230 \times 10^4/\text{mm}^3$, hemoglobin 7.3 g/dl, and platelets $48.8 \times 10^4/\text{mm}^3$. Coagulation test results were within normal limits. Blood chemistry showed the following abnormal values: total bilirubin 2.5 mg/dl (normal 0.2–1 mg/dl), direct bilirubin 2.2 mg/dl, aspartate aminotransferase 105 IU (normal 8–38 IU), alanine aminotransferase 233 IU (normal 5–33 IU), alkaline phosphatase 1980 IU (normal 88–288 IU), leucin aminopeptidase 262 IU (normal 30–64 IU), and γ -glutamyl transpeptidase 116 IU (normal 3–40 IU).

Gastroendoscopy was performed immediately on admission, revealing only for a small amount of “coffee

ground” material in the stomach. No blood was detected in the duodenum. The patient received a transfusion of two units of packed red blood cells, and his condition remained relatively stable during the next few days. Gastroscopy was repeated, but no abnormalities were detected in the upper gastrointestinal tract. Abdominal ultrasonography showed a distended gallbladder with a few gallstones and an echolucent cystic lesion, 2.2 × 3.0 cm in diameter, attached to the thickened wall. Pulsatile waves were detected in the cystic lesion by color-Doppler imaging (Fig. 1). The common bile duct was not extended and no stone was observed in it by ultrasound imaging. Computerized tomographic (CT) evaluation of the abdomen demonstrated a well defined cystic lesion, approximately 3 cm in diameter, in the lumen of the gallbladder, which was strongly enhanced by the infusion of contrast medium (Fig. 2). From these findings, we suspected hemorrhage into the gallbladder due to an aneurysm of the cystic artery, which had caused melena. Side-viewing duodenoscopy was performed, and bleeding from the papilla of Vater was confirmed on February 8, 1995. Subsequently, hepatic arteriography was carried out, revealing marked dilatation of the posterior branch of the cystic artery forming an aneurysm in the midst of the artery. Pulsatile jet flow of the contrast medium was followed into the aneurysm of the cystic artery by digital subtraction arteriography (DSA) (Fig. 3a). Pooling of contrast medium was recognized in the venous phase of DSA (Fig. 3b). We diagnosed pseudoaneurysm of the cystic artery as a cause of gastrointestinal bleeding, and cholecystectomy was performed on February 9.

Pathology evaluation showed a grayish-red gallbladder that was distended and contained turbid fluid, a giant hematoma, old blood clots, and two gallstones.

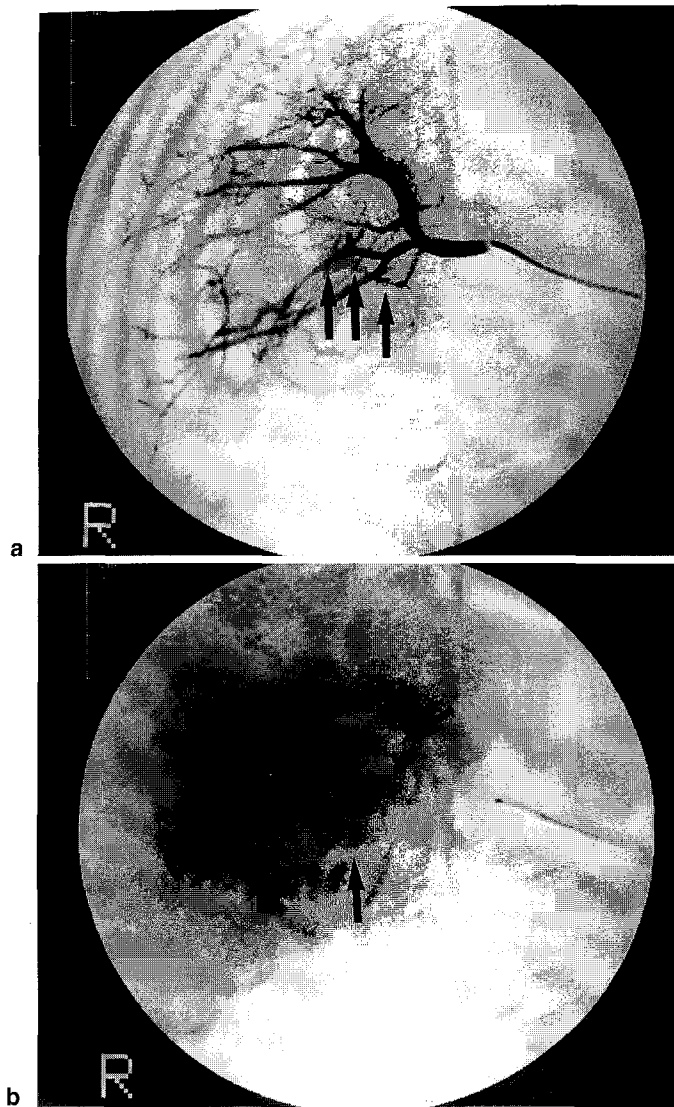


Fig. 3a,b. Hepatic arteriography. **a** The posterior branch of the cystic artery is markedly dilated and has formed an aneurysm, and pulsatile jet flow can be followed in it (*arrows*). **b** Pooling of contrast medium is recognized in the venous phase of digital subtraction arteriography (*arrow*)

The mucosa of the gallbladder was shed in some areas and preserved in others. The wall of the gallbladder was diffusely thickened. The hematoma, 3 cm in diameter and 2 cm in height, was attached to the lumen of the gallbladder (Fig. 4a). Histologically, edema and extravasated bleeding were marked, and inflammatory cells had infiltrated the wall of the gallbladder (Fig. 4b). No other unusual findings, such as arterio-venous malformation or anomalies of artery, were detected pathologically or clinically. Pseudoaneurysm of the cystic artery with acute calculous cholecystitis was definitively diagnosed.

Postoperatively, the patient recovered and was discharged. Six months later he was alive and well.

Discussion

The term "hemobilia" was first applied by Sandblom¹², in 1948, to the syndrome of gastrointestinal bleeding caused by hemorrhage into the biliary tract. The clinical features of hemobilia had already been reported, by Quincke¹³ in 1871, as biliary colic, jaundice, and gastrointestinal bleeding. Nevertheless, in many cases there have been delays in making a clinical diagnosis, as in the present case.^{4,9,10,14} Emergent endoscopy for melena did not detect the source of gastrointestinal bleeding, but the patient already had the classical triad of colic, jaundice, and gastrointestinal bleeding on admission. Retrospectively, it was possible to readily diagnose hemobilia only according to the clinical symptoms, without any diagnostic procedure. This emphasizes the importance of considering hemobilia whenever gastrointestinal bleeding is associated with signs of biliary disorder.^{3,9,14}

Pseudoaneurysm of the cystic artery is a rare cause of hemobilia,⁸ and, to the best of our knowledge, only eight cases have been reported previously in the literature.⁴⁻¹¹ Barba et al.¹⁰ reported that pseudoaneurysm of the cystic artery was associated with severe acute cholecystitis and cholelithiasis. The rarity of pseudoaneurysm of the cystic artery may be because the disease was considered to be related to early thrombosis of the cystic artery as a reaction of the inflammatory process.^{6,10} Pseudoaneurysm of the cystic artery has also recently been reported as a complication of laparoscopic cholecystectomy.¹¹ Since, in our patient, acute calculous cholecystitis was present synchronously with pseudoaneurysm of the cystic artery, it is possible that acute inflammation of the gallbladder may have caused the pseudoaneurysm. However, we found no evidence that elucidated the genesis of the pseudoaneurysm in this patient. Further research will be needed to resolve the etiology of the condition.

The last decade has seen major advances in scanning technology. Ultrasonography has been recognized as a safe, inexpensive, and highly effective diagnostic procedure, and color-Doppler ultrasound may offer much information for diagnosis and prove to be useful in equivocal cases.¹⁰ In our present patient, pseudoaneurysm of the cystic artery was shown as a cyst in the gallbladder without color-Doppler, although persistent pulsatile flow was detected in the lesion by color-Doppler imaging. This finding on Doppler ultrasound led us to strongly suspect aneurysm of the cystic artery as the cause of gastrointestinal bleeding. Barba et al.¹⁰ also reported the advantage of color-Doppler imaging in the diagnosis of pseudoaneurysm of the cystic artery. Color-Doppler ultrasonography was beneficial in the diagnosis of this rare disease, and it may have further possibilities in the analysis of other abdominal diseases.

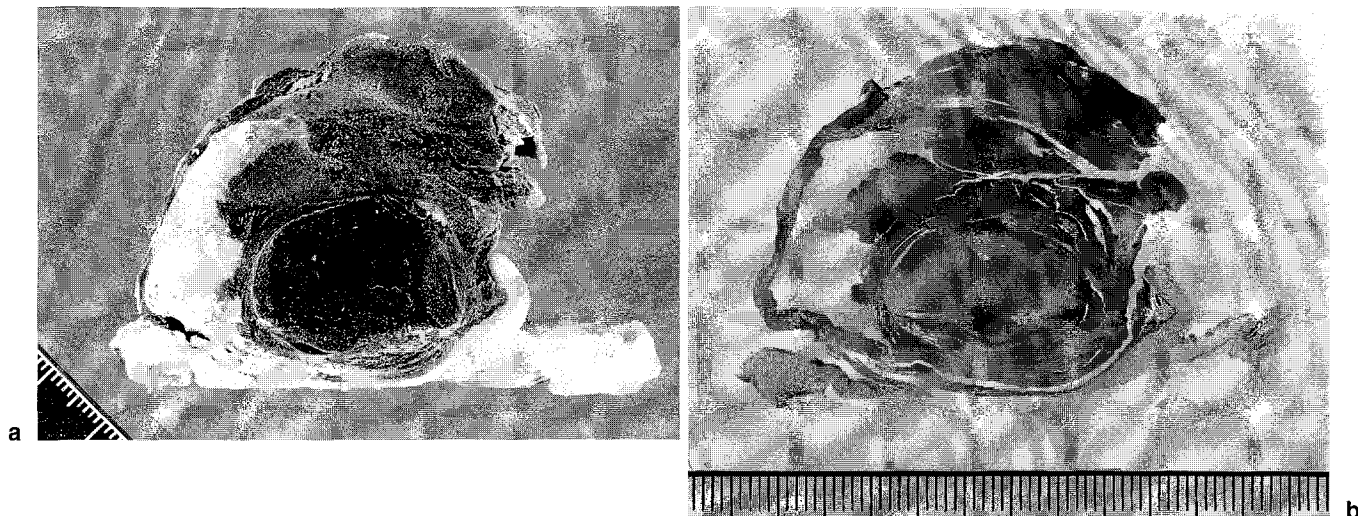


Fig. 4a,b. Pseudoaneurysm of the cystic artery. **a** The wall of the gallbladder is diffusely thickened. The hematoma, 3 cm in diameter and 2 cm in height, was attached to the lumen of the gallbladder. **b** Histologically, edema and extravasated bleeding were marked, and inflammatory cells had infiltrated the wall of the gallbladder. H&E, $\times 1.2$

CT is also a highly accurate method of investigating the liver, the bile duct, and the gallbladder.^{6,10,11} However, it was not so helpful in the diagnosis of pseudoaneurysm of the cystic artery in our patient. Only a cystic lesion was revealed in the gallbladder by non-enhanced plain CT, and the lesion was enhanced by bolus injection of contrast medium. Nevertheless, it was difficult to determine whether the cystic lesion was an aneurysm of the artery or a hemorrhage into the gallbladder, since CT did not detect the arterial pulse directly. Compared with color-Doppler imaging, CT appears to be less specific for detecting aneurysm of the cystic artery. Hepatic arteriography shows direct findings of the aneurysm and provides a definitive diagnosis when pseudoaneurysm is suspected.^{4,15} Using preoperative arteriography, we were able to establish with certainty the diagnosis of pseudoaneurysm of cystic artery, thereby simplifying the surgical procedure. However, arteriography, compared with color-Doppler imaging, is more invasive and requires more complex equipment and greater human resources in terms of time and personnel. Endoscopy is the ultimate maneuver for diagnosis and treatment in most cases of upper gastrointestinal bleeding. However, we could not confirm the precise source of bleeding by emergent endoscopy. If endoscopy had revealed bleeding through the papilla of Vater, hemobilia could have been diagnosed more readily in our patient. Endoscopy cannot always be a definitive procedure in patients with hematemesis or melena.^{15,16}

This rare case of pseudoaneurysm of the cystic artery shows us the importance of hemobilia as a cause of alimentary tract bleeding. The advantages of color-Doppler ultrasound, from the perspective of non-invasiveness, utility, and cost, suggest that this modality may be more suitable for diagnosis of aneurysm of the cystic artery than selective hepatic arteriography.

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References

1. Bogoch A. Bleeding from the alimentary tract. In: Haubrich WS, Schaffner F, Berk JE (eds) *Bockus gastroenterology*. Philadelphia: WB Saunders, 1995:61–67.
2. Sherlock S, Dooley J. *Diseases of the liver and biliary system*. Oxford: Blackwell Scientific, 1992.
3. Sandblom P, Mirkovitch V. Hemobilia: Some salient features and their causes. *Surg Clin North Am* 1977;57:397–408.
4. Reddy SC. Pseudoaneurysm of cystic artery with upper gastrointestinal hemorrhage. *South Med J* 1983;76:85–86.
5. Rhee JW, Bonnheim DC, Upson J. Cystic artery pseudoaneurysm. *NY State J Med* 1987;87:47.
6. Wu TC, Liu TJ, Ho YJ. Pseudoaneurysm of the cystic artery with upper gastrointestinal hemorrhage. *Acta Chir Scand* 1988;154:151–152.
7. Smague EA, Schulte F, Guse S. Rezidivierende Hämobilie aufgrund eines in der Gallenblase rupturierten Pseudoaneurysmas der Arteria cystica. *Chirurg* 1990;61:199–200.

8. Strickland SK, Khury MB, Kiproff PM, et al. Cystic artery pseudoaneurysm: A rare cause of hemobilia. *Cardiovasc Intervent Radiol* 1991;14:183–184.
9. Clements WDB, Wilson RH, Crothers JG, et al. Pseudoaneurysm of the cystic artery following cholecystectomy. *J Roy Coll Edinb* 1993;38:348–349.
10. Barba CA, Bret PM, Hinchey EJ, et al. Pseudoaneurysm of the cystic artery: A rare cause of hemobilia. *Can J Surg* 1994;37:64–66.
11. Bergey E, Einstein DM, Herts BR. Cystic artery pseudoaneurysm as a complication of laparoscopic cholecystectomy. *Abdom Imaging* 1995;20:75–77.
12. Sandblom P. Hemorrhage into the biliary tract following trauma—traumatic hemobilia. *Surgery* 1948;24:571–586.
13. Quincke H. Ein Fall von Aneurysma der Leberarterie. *Berl Klin Wochenschr* 1871;30:349–352.
14. Lewis RT. Nontraumatic hemobilia: Disparate episodes 7 years apart in the same patient. *Can J Surg* 1979;22:175–177.
15. Yoshida J, Donahue PE, Nyhus LM. Hemobilia: Review of recent experience with a worldwide problem. *Am J Gastroenterol* 1987;82:448–453.
16. Ryvicker MJ, Schatz SL, Deutsch AM, et al. Angiographic demonstration of bleeding into gallbladder. *Radiology* 1980;136:326.