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## LETTER TO THE EDITOR

### Hemobilia due to pseudoaneurysm of the cystic artery

### Hémobilie due à un pseudo-anévrisme de l'artère cystique

#### Introduction

Gastrointestinal bleeding from the biliary tree (hemobilia) is an uncommon event. Iatrogenic or traumatic liver injury are the most frequent causes of the disorder, accounting for over 50% of cases, followed by gallstones, acalculous inflammation, vascular abnormalities and neoplastic disease [1]. Rare cases of spontaneous intracholecystic bleeding have been published in patients with vascular and coagulation disorders [2]. We report a rare case of hemobilia due to a pseudoaneurysm of the cystic artery.

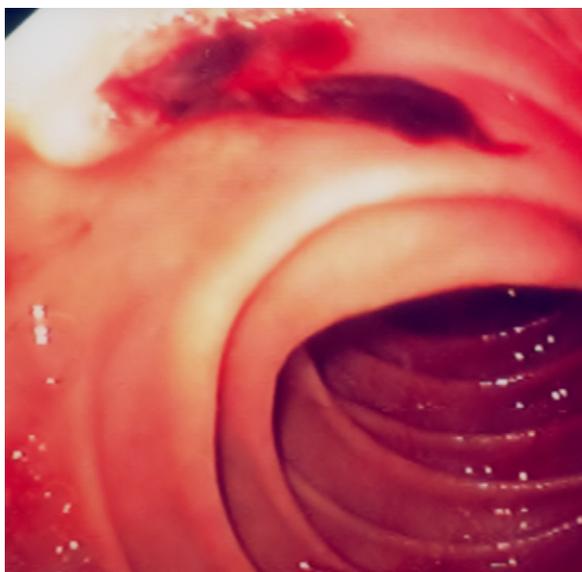
#### Case report

An 84-year-old man presented with hematemesis which had been preceded by severe epigastric pain and nausea, several hours before. Both had improved when the patient vomited blood. He had no fever or prior hemorrhagic events, no recent anorexia, weight loss or altered bowel habits. He was a light drinker (<25 g of ethanol per week) and a former smoker (80 unit per pack per year, stopped 30 years ago). Past medical history included obesity (body mass index 31 kg/m<sup>2</sup>), type 2 diabetes mellitus, open surgery for benign prostate hypertrophy 4 years before, and a cardiac pacemaker for 6 years. Medication included oral antidiabetics (metformin 500 mg tid, gliclazide 80 mg id) and acetylsalicylic acid 100 mg id. He was afebrile, blood pressure (180/75 mmHg) and heart rate (68 bpm) were stable. The abdomen was tender at the epigastrium and a painless enlargement of the spleen which was palpable about 8 cm from the costal grid on nipple line was found on clinical examination. Fresh blood was expelled from a nasogastric tube. Laboratory tests showed hemoglobin 8.7 g/dL with normal leucocytes and platelet counts, as well as coagulation times. Liver chemistry revealed total bilirubin 2.8 mg/dL, direct bilirubin 1.9 mg/dL, alanine aminotransferase 160 IU/L (N: 13–40 IU/L), aspartate aminotransferase 232 IU/L (N:

10–42 IU/L),  $\gamma$ -glutamyltransferase 194 IU/L (N: <50 IU/L) and alkaline phosphatase 199 IU/L (125–240 IU/L). Amylase-mia was normal and serum C-reactive protein was 1.4 mg/dL (N: <0.5 mg/dL). Ultrasound (US) confirmed a 17.5 cm homogeneously enlarged spleen and showed a stone-containing gallbladder with an irregularly thickened wall, suggesting cholecystitis or neoplasia (Fig. 1). Emergent upper gastrointestinal endoscopy revealed an adherent clot over the superior wall of the 2nd portion of the duodenum. Local injection of 4 cm<sup>3</sup> of epinephrine (1:10000) was performed with no active bleeding during or after the injection. The papilla of Vater was not clearly seen. Rebleeding occurred 4 hours later. A second endoscopy revealed fresh blood and clots emerging from the papilla of Vater (Fig. 2). Computed tomography (CT) showed marked thickening of the gallbladder wall, which contained a single calcified stone and high density material suggesting recent intraluminal bleeding. The arterial phase of dynamic CT showed a vaguely nodular 14 mm lesion at the infundibulum, with an enhancement pattern parallel to the arteries resulting in extravasation of iodine in the gallbladder lumen. These results suggested a vascular pseudoaneurysm with active intracholecystic bleeding and possible cholecystitis (Fig. 3). Even though the patient was still stable, a surgical solution was chosen due to possible concomitant cholecystitis. The gallbladder was found to be adherent to the omentum on laparotomy. During retrograde dissection of the gallbladder from its liver bed, an aneurysmatic dilatation of the cystic artery was identified and ligated, immediately stopping active bleeding. Resection of the gallbladder was completed with no further incident. Gross examination of the resected specimen showed a larger piece (5 × 3 cm) with a 1 cm thickened, yellowish wall with hemorrhagic areas and a smaller (2 × 1.5 cm) congestive piece containing blood clots. Microscopic examination showed mucosal ulcerations and mixed inflammatory and hemorrhagic infiltrates coexisting with fibrous thickening of the wall, muscular propria hypertrophy, non-obliterative endarteritis and nervous fillets' hyperplasia. Blood clots were also observed inside the wall of the smaller fragment. An acute process superimposed on chronic inflammation of the gallbladder was the final diagnosis. The postoperative course was uneventful jaundice improved progressively and 18 months after surgery, the patient is alive and well.



**Figure 1** Abdominal US: irregularly thickened gallbladder wall and a stone in the lumen.



**Figure 2** Upper gastrointestinal endoscopy: fresh blood and clots emerging from the papilla of Vater.

## Discussion

We report a case of hemobilia caused by a pseudoaneurysm of the cystic artery. The exact mechanism of the development of a pseudoaneurysm is unknown, and probably varies according to local pathological phenomena. As in previous cases, we can assume that chronic inflammation led to the development of the pseudoaneurysm, which was eroded by the overlapping acute ulceration of the mucosa [3]. Acute cholecystitis alone could also have caused both an ulceration of the serosa and partial erosion of the elastic and muscular components of the vascular wall, leading to the development and rupture of the pseudoaneurysm. In either case, the erosive process may have been accelerated by the presence of the stone [4]. The possibility that inflammation will lead to early thrombosis of the pseudoaneurysm may explain the rarity of the entity [3,4].



**Figure 3** a: plain computed tomography: calcified gallbladder calculus (large arrow) and spontaneous high density of the gallbladder content (arrowheads) suggesting recent bleeding; b-c: maximum intensity projection (MIP) reconstruction from the arterial (b) and portal venous (c) phases of the dynamic study showed the calcified gallbladder calculus (large arrow) and a dense nodule (thin arrow) at the infundibulum, with enhancement paralleling the adjacent arteries, corresponding to the pseudoaneurysm.

The clinical presentation of hemobilia is usually associated with Quincke's triad of biliary colic (occurring in 70% of patients), jaundice (exhibited in 60% of the cases) and some gastrointestinal bleeding which is present in all patients [1]. The complete classic triad is found in only 32 to 40% of patients [1] with acute bleeding first causing biliary colic followed by hematemesis or melenae [2], which in turn leads to pain relief. There are 16 case-reports in the literature of pseudoaneurysm of the cystic artery as a cause of hemobilia. Indeed, if our case is considered in relation to the series

by Akatsu et al., the classic signs appeared more frequently than those reported for hemobilia in general because abdominal pain, gastrointestinal bleeding and jaundice occurred in 94% ( $n = 16$ ), 94% ( $n = 16$ ) and 65% ( $n = 11$ ) of patients, respectively, and Quincke's triad was present in 59% ( $n = 10$ ). These differences may be because bleeding occurs in a confined space such as the gallbladder causing organ distension, which could explain the almost systematic relief of severe biliary pain when the blood is expelled. In hemobilia, the severity of jaundice reflects the degree of obstruction of the bile duct with clots. When the gallbladder is the site of bleeding this follows the blood's emergence from the organ, which may explain the similar rate of jaundice in this particular case and in hemobilia in general.

The association of upper gastrointestinal bleeding and biliary disorders should suggest hemobilia because the longer treatment is delayed, the higher the rates of morbidity and mortality [3,4]. Diagnostic tests reflect the history and clinical presentation of hemobilia [2]. Upper endoscopy shows clots exiting through the papilla of Vater in 12 to 30% of cases of hemobilia [5]. US, CT and magnetic resonance imaging (MRI) can identify inflammation of the gallbladder, stones, neoplasms and vascular abnormalities, while dynamic CT or MRI can identify the source of active bleeding [2,4,5]. Moreover pre-operative identification of a vascular pseudoaneurysm inside or near the gallbladder can be detected by Doppler US [3], and aneurysms over 1 cm, as in our case, can be found by contrast-enhanced CT scanning [4,6].

Despite these advances, angiography is still considered by many to be the technique of choice in the management of hemobilia of any cause and for suspected pseudoaneurysms in particular [6], due to its high diagnostic accuracy and therapeutic potential [1,3]. Nevertheless, angiography has certain diagnostic limitations for the variable flow rate and intermittent bleeding, and also when there are hepatic artery abnormalities or has been previous manipulation [1]. Moreover, although the reported success rate of angiographic control of hemobilia by transarterial embolization is 80 to 100% [5], this entails some serious risks such as hepatobiliary necrosis, bleeding, abscess formation and gallbladder fibrosis [1]. As a result, some authors argue that the treatment of choice for hemobilia should be surgery whenever cholecystitis, gallstones or resectable neoplasms are present or if embolization fails [1,2,5]. Generally, hemobilia should be managed by a combined approach, regardless of the cause: embolization of the bleeding vessel to stabilize the patient followed by cholecystectomy at a later, safer time [3,4,6], which should be better even though two invasive procedures are involved. However, only two of the 17 cases of pseudoaneurysm of the cystic artery described in the literature were treated in this two-step fashion [4] hence it may not be the most adequate strategy for this particular situation.

In conclusion, careful, non-invasive imaging examinations should be performed whenever hemobilia is suspected in particular if the bleeding is from the gallbladder. Detailed identification of the bleeding vessel allows carefully planned and directed surgery, avoiding an additional invasive angiographic procedure.

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## Papillomatose étendue des voies biliaires : une indication rare de transplantation hépatique

### Diffuse biliary papillomatosis: A rare indication for liver transplantation

#### Introduction

La papillomatose des voies biliaires est une maladie rare, décrite comme pouvant être localisée, l'affection est caractérisée par une tendance à la diffusion au niveau de l'ensemble des voies biliaires. Elle présente un potentiel évolutif malin, qui fait considérer cette maladie comme une affection préneoplasique. Nous rapportons une observation qui illustre les difficultés diagnostiques et thérapeutiques habituellement rencontrées dans cette maladie.