

## CASE REPORT

## Hemobilia by idiopathic aneurysm of cystic artery, fistulized in the biliary ways – clinical case

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### Abstract

Aneurysm of the cystic artery is not common, and it is a rare cause of hemobilia. Most of reported cases are pseudoaneurysms resulting from either an inflammatory process in the abdomen or abdominal trauma. We report a healthy individual who developed hemobilia associated with cystic artery aneurysm. Visceral artery aneurysms are rare and can rupture with potentially grave outcome due to excessive bleeding. The patient was managed with cholecystectomy and concomitant aneurysm repair.

**Keywords:** cystic artery aneurysm, biliary fistula, hemobilia.

### Introduction

The existence of hemobilia raises the suspicion of fistula between a vascular structure and a biliary channel. Sandblom defined for the first time the term of hemobilia in 1948, as the pathological state characterized by the presence of blood in the biliary tract. This disease manifests by the classical triad including: biliary colic, jaundice and upper digestive bleeding [1].

Hemobilia is a rare case of acute upper digestive bleeding and is frequently associated to a biliary or liver trauma (recent or in medical history), transcuteaneous surgical procedures, laparoscopic cholecystomy, presence of a tumor of the biliary tree or the liver, presence of a liver artery or cystic artery aneurysm, presence of a cholecystitis or liver abscess, etc. [2].

The clinical and paraclinical diagnosis of hemobilia is often difficult to make and requires a cholangiographic investigation that may highlight changes of the biliary tree. The echography and computed tomography (CT) may highlight possible segmentary dilations of the biliary ways, with the highlight of blood flow inside, and if the blood flow is low, in the biliary ways there may be highlighted echogenic material (thrombi) [3, 4].

Idiopathic hemobilia resulted from the rupture of an aneurism in the cystic and liver artery appears as a result of certain laparoscopic procedures in the medical history [5–7].

We present the case of a patient with idiopathic aneurism of cystic artery, fistulized in the biliary ways

that raised some problems of positive and differential diagnosis.

### Case presentation

The patient LV, aged 83 years, retired, was admitted to hospital between November 20–29, 2013, within the Department of Pulmonology of the Emergency County Hospital of Arad, Romania, with repeated hemoptysis in low quantities, asthenia, dry cough.

From the disease history, there was observed that the patient had been diagnosed with chronic obstructive bronchopneumopathy a few years before, sequels after an ischemic stroke and high blood pressure (HBP), for which he received an intermittent treatment. Hemoptysis appeared about three days before, they were reduced quantitatively and they were caused by recurring cough.

The general clinical examination highlighted an altered general state, with maintained consciousness, suffering face expression, pale skin and poorly represented conjunctive-adipose tissue. Regarding the respiratory system, the patient presented emphysematous thorax, with low respiratory movement, with sibilant rales, associated with dyspnea and tachypnea. The blood pressure was high at admission (170/100 mmHg) and maintained above normal values during the entire period of hospitalization, even under hypotensive treatment. The abdomen examination highlighted spontaneous epigastralgia and, at palpation, pains in the cystic point, moderate hepatomegaly (the anterior margin of the liver was 3 cm below the rib cage).

The patient was admitted in the Department of Pulmonology for paraclinical investigations, diagnosis setting and specialized treatment.

Starting from the suspicion of pulmonary tuberculosis or bronchopulmonary cancer, the patient was subjected to some imagistic and laboratory investigations. Thus, the thorax-mediastinum-lung X-ray and the lung CT did not highlight any specific lesions of lung tuberculosis or lung proliferative processes. The bacteriological examination on culture environments and the microscopic examination of sputum in special stainings did not identify the presence of Koch bacilli. Based on the clinical and paraclinical data, there was established the diagnosis of chronic obstructive bronchopneumopathy with clinically manifest lung failure, recurring hemoptysis of unknown origin, type 2 HBP, followed by an appropriated treatment. The clinical symptoms improved slowly with the applied treatment.

After a few days from admission, the patient's general state started to alter, with symptoms of abdominal pains in the epigastrium and the right hypochondrium, associated with appetite loss, nausea, digestive discomfort and melenas stools. Under these circumstances, the patient was transferred to the Clinic of Surgery within the Emergency County Hospital of Arad for continuing the investigations and treatment. At admission in the Clinic of Surgery, the patient was conscious, compliant, slightly dyspneic, pale, hemodynamically balanced, with a present bowel transit, afebrile, present diuresis. The clinical examination highlighted the altering of general state, with scleral and skin jaundice, palpation pains in the epigastrium and right hypochondrium, painful hepatomegaly, first-degree splenomegaly. Upon admission within Department of Surgery, the patient develops two episodes of superior digestive hemorrhage, outlined by abundant hematemesis and abundant melenas, with clinical and biological signs of severe anemia. After both episodes of upper gastrointestinal hemorrhage, emergency gastroscopy was performed without the bleeding site being observed.

The simple X-ray of the abdomen did not highlight the presence of pneumoperitoneum or other hydroaeric levels, only a marked aerocolia (Figure 1). CT of the

abdomen and pelvis, with i.v. administered contrast substance, highlighted a large volume liver, with a longitudinal diameter of the right lobe of 20 cm, with no karyokinetic processes, with no dilations of intrahepatic biliary ways. The cholecyst was more distended with multiple dense, heterogeneous images, with the inflowing of the contrast substance into the cholecyst (Figure 2). The pancreas had normal sizes, with signs of involution, with no pancreatic focal masses, with a discrete peri-pancreatic edematous infiltration, especially at tail level, a suggestive aspect for acute pancreatitis. The spleen was slightly larger, with the long axis of 14.5 cm, with no focal lesions. CT exam after the second upper digestive hemorrhage raises suspicion of cystic artery aneurysm (Figure 3).

The paraclinical examinations showed a severe anemia, the hemoglobin (Hb) value reaching 8.2 mg/dL, with 2.69 million red blood cells/mm<sup>3</sup> and a hematocrit of 23%; leukocytosis (11.3 thousand leukocytes/mm<sup>3</sup>) with neutrophilia (88.2%), hyperglycemia (158 mg/dL), increase of transaminases (AST – aspartate aminotransferase 346 IU/mL; ALT – alanine aminotransferase 174 IU/mL); high bilirubin (total bilirubin 4.5 mg/dL; direct bilirubin 3.9 mg/dL), amylasemia (627 IU/L).

Clinical association of pain, jaundice, hematemesis, raises clinical suspicion of hemobilia.

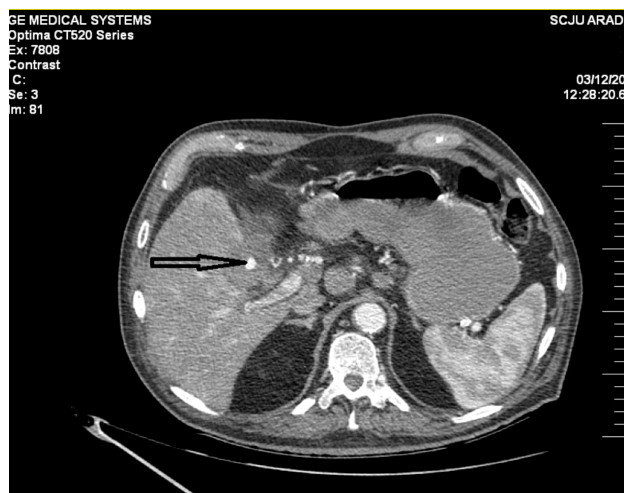
After several perfusions and transfusions with isogroup blood, isoRh and after a thorough preoperative preparation, there was performed a surgery for establishing the cause of digestive bleeding. During surgery, there was highlighted a cystic artery aneurism, fistulized in the biliary ways (Figure 4) with massive hemobilia, with distended cholecyst, with areas of parietal incipient necrosis and blood clots inside. In this context, there was performed a complete cholecystomy with exploratory choledotomy, biliary drainage and ligature of the cystic artery aneurism. Subsequently, during surgery, there was also performed a control cholangiography that did not highlight any changes of the remaining biliary tree (Figure 5) or any choleduct obstructions, the contrast substance being distributed homogeneously in the intrahepatic biliary ways and duodenum.



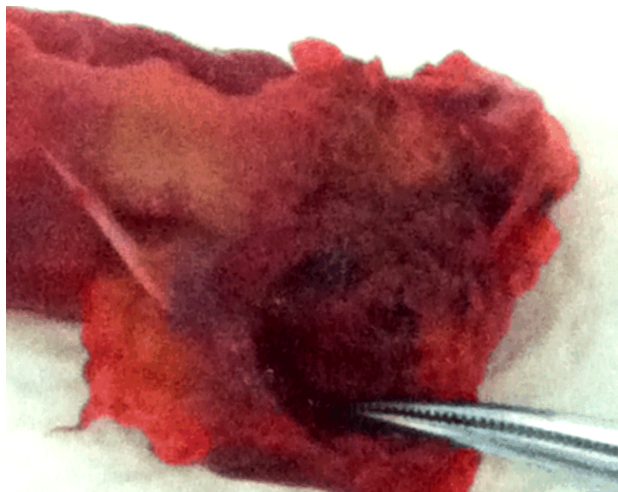
**Figure 1** – Abdominal X-ray image where cannot be highlighted any pathological changes (pneumoperitoneum or hydroaeric levels).



**Figure 2** – Abdominal CT highlighting a large volume cholecyst, with contrast substance debris in the lumen.



**Figure 3** – CT exam after the second upper digestive hemorrhage raises suspicion of cystic artery aneurysm.



**Figure 4** – Macroscopic aspect of the surgical exeresis piece where there is highlighted the aneurism dilation of the cystic artery and communication with the biliary way.



**Figure 5** – Cholangiography during surgery highlighting the possible malformations of the biliary ways.

After surgery, the patient's postoperative evolution was slowly favorable.

Unfortunately, after five days since surgery, the patient developed nosocomial bronchopneumopathy, with an unfavorable progression, which led to his death.

## Discussion

Hemobilia was observed and described from 1654 by Francis Glisson, who observed the presence of blood in the biliary ways in a young man with severe liver lesions after a sword fight, but the term "hemobilia" was introduced in 1948 by Sandblom, who described this condition in detail [8–10].

Hemobilia commonly manifests with upper digestive bleeding, associated with epigastralgia or pains in the right hypochondrium, altering of general state through a digestive tract bleeding, upper digestive bleeding and jaundice [1, 11, 12]. The most frequent causes of hemobilia are traumas of the liver, followed by iatrogenic traumas during the diagnosis and treatment procedures of hepatobiliary pathologies [12]. The non-traumatic causes include acute or chronic cholecystitis, pancreatitis or hepatobiliary neoplasms [13, 14].

The cystic artery aneurism is a rarely found pathology; as a result of our research in the published studies up to now, we identified only 27 similar cases, most of them being pseudoaneurysms caused by traumatic lesions or surgical procedures of the liver or biliary ways. The causes of a cystic artery aneurism complicated with hemobilia include abdominal traumas and intra-abdominal inflammatory processes, such as acute cholecystitis or pancreatitis [8, 15]. The cystic artery aneurism fistulized in the biliary ways commonly manifests by hemobilia, with hematemesis and/or massive melena [16–18].

The etiology of cystic artery aneurism in the case of our patient could not be established, as the patient did not suffer any abdominal traumas or abdominal invasive surgical procedures. It is possible that there might have existed a congenital malformation of the cystic artery or of the biliary tree that may have favored the fistulization of the cystic artery in the biliary ways.

Due to the scarcity of this pathology, there is no agreement regarding the clinical or surgical management of cystic artery aneurisms. While cholecystomy is considered as the basic treatment in most cases of cholecystitis, for the treatment of arterial aneurisms, arterial embolization seems to be the most indicated ones. In certain situations, there was used arterial embolization combined with cholecystomy. The angiographic treatment is not possible in all situations. Most of the reported cases were associated with cholecystitis, the election treatment in these cases being cholecystectomy. Morioka *et al.* [19] reported a patient whom could not undergo embolization, due to the reduced diameter of the aneurysm, only cholecystectomy being possible in the end.

We consider that, due to the fact that the hemobilities caused by fistulas of the idiopathic aneurysms of the hepatobiliary vessels are quite rare, the clinical and paraclinical diagnosis presenting a high degree of difficulty and special management. In our case, the initial symptoms (hemoptysis cough) raised the problem of a karyokinetic process of the tracheal bronchi tree. Then, the onset of melena stool and abdominal pains directed the diagnosis to an upper digestive bleeding of unknown cause. The onset of jaundice with phenomena of hepatocytolysis and

pancreatic dysfunction raised the problem of an acute pancreatitis, especially because the CT examination identified a moderate peripancreatic pathology. The highlighting of a distended cholecyst and the presence of the contrast substance in the cholecyst raised the problem of a hepatobiliary pathology. The certainty diagnosis was established during surgery, when there was observed the presence of blood clots in the cholecyst and biliary ways, and the presence of the cystic artery aneurism fistulized in the biliary way.

We, together with other authors, consider that hemobilia is a diagnosis challenging due to its rarity and varied symptoms [20–22].

## ✉ Conclusions

The cystic artery aneurism is a rare pathology that may rupture, thus causing massive internal bleeding, with tragic consequences. Medical imaging may highlight, more or less, the changes caused by this pathology. In our case, the surgical procedure allowed the establishment of diagnosis and the applied therapeutic method (cholecystectomy, exploratory choledocotomy, cystic artery aneurism drainage and ligation) was considered the election treatment, due to the cholecystitis caused by the obstruction of the cystic canal, by the presence of blood clots.

## Conflict of interests

The authors declare that they have no conflict of interests.

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