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Non traumatic pseudoaneurysm of the cystic artery as a cause of haemobilia☆

Pseudoaneurisma no traumático de la arteria cística como causa de hemobilia

The term haemobilia refers to any gastrointestinal bleeding originating from the biliary Tree.[1] Its aetiology is quite varied and determines the necessary treatment. Currently, most cases are secondary to accidental or iatrogenic trauma.[2] Vascular disorders, such as arterial pseudoaneurysms, are an uncommon cause. This article will look at 2 cases of haemobilia secondary to pseudoaneurysms of the cystic artery, both associated with symptoms of acute cholecystitis.

The first case is a 74-year-old male patient with right upper quadrant pain associated with fever, haematemesis and melaena. His lab results showed elevated amylase, lipase and cholestasis markers. An upper gastrointestinal (GI) endoscopy was performed, which revealed bleeding from the duodenal papilla. A CT angiogram showed a 2-cm pseudoaneurysmal lesion adjacent to the infundibulum of the gallbladder, with a large gallstone lying in that region (Fig. 1) as well as signs of acute cholecystitis. The patient had a vascular anatomical variant: common hepatic artery with origin in the superior mesenteric artery, dividing into 3 branches before the hepatic hilum. An arteriogram was then performed which revealed the pseudoaneurysmal lesion, without managing to identify the artery supplying the pseudoaneurysm, making embolisation impossible. Finally, the patient underwent laparoscopic surgery, which revealed acute cholecystitis with active gallbladder bleeding, requiring conversion to open cholecystectomy. He had a large 3×2 cm gallstone lying against the wall of the gallbladder within the infundibulum, eroding the aforementioned pseudoaneurysm.

The second case is a 74-year-old female patient with right upper quadrant pain, fever and melaena, who developed anaemia during her hospital stay. She had elevated cytolysis and cholestasis markers. An abdominal ultrasound revealed acute cholecystitis with echogenic material within the gallbladder lumen and dilatation of the intrahepatic bile duct. The upper GI endoscopy showed blood clots in the 2nd part of the duodenum extruding from the papilla.

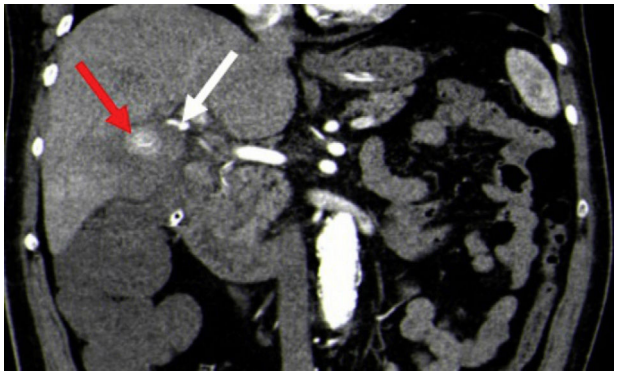

Figure 1 White arrow: pseudoaneurysm. Red arrow: gallstone.

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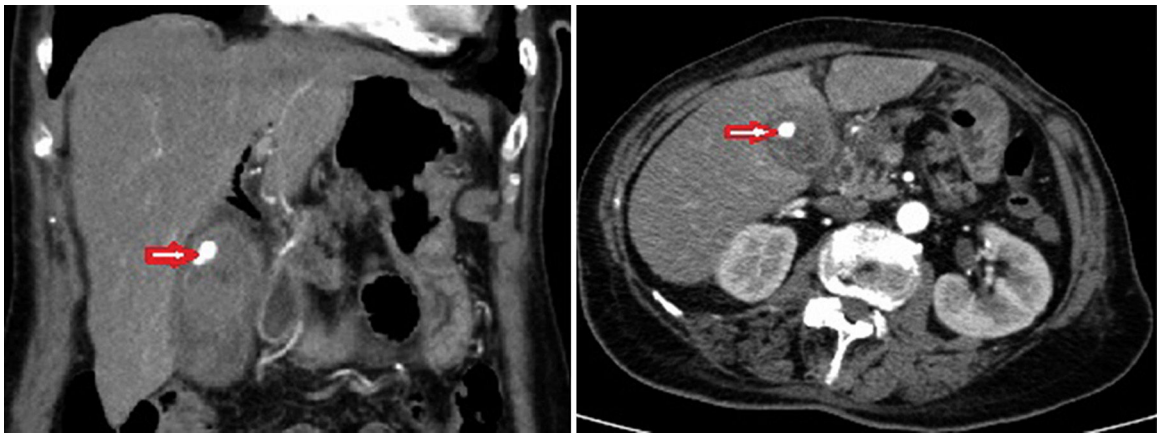

Figure 2 The arrow is indicating the arterial pseudoaneurysm.

An endoscopic retrograde cholangiopancreatography (ERCP) was performed to remove blood clots from the main bile duct. A CT angiogram revealed acute cholecystitis and a pseudoaneurysmal lesion of the cystic artery, with no signs of active bleeding, although there was hyperdense material within the gallbladder, suggesting intraluminal bleeding (Fig. 2). Finally, a laparoscopic cholecystectomy was performed, which confirmed the presence of acute cholecystitis with a large blood clot and two 2-cm gallstones in the gallbladder.

Both patients had a favourable outcome with no further gastrointestinal bleeding.

Vascular disorders are an uncommon cause of haemobilia. Aneurysms or pseudoaneurysms occur mainly in the hepatic artery or one of its branches.[3] Cases of haemobilia caused by vascular disorders of the cystic artery are very rare and generally occur following iatrogenic trauma, such as a laparoscopic cholecystectomy.[4] Non-traumatic pseudoaneurysms of the cystic artery are rare and are generally due to inflammatory processes of the gallbladder, which, due to their proximity, cause vascular lesions which may produce a fistula between the artery and the gallbladder. The inflammatory process damages the vascular adventitia and causes thrombosis of the *vasa vasorum*, weakening the vessel wall. Another mechanism involved is erosion caused directly by large gallstones, which may lie against the cystic artery.[5,6]

The diagnostic methods to be used in these cases will depend on the patient's haemodynamic status. Provided that the patient remains stable, the first exam to be performed should be an upper GI endoscopy, which will allow doctors to rule out other sources of gastrointestinal bleeding and check for signs of bleeding from the papilla. Nevertheless, this does not allow treatment at the source of the bleeding and may give negative results if bleeding is intermittent.[6]

A CT angiogram is very useful because, in addition to revealing lesions that are the source of bleeding, it also identifies contrast extravasation if extensive enough.[7]

The most important examination method in cases of haemobilia with proven (generally by CR angiogram) or suspected active bleeding is angiography, especially since it allows for selective embolisation of the bleeding vessel.[3] More and more authors consider embolisation to be the *gold standard* for the initial treatment of haemobilia with proven active bleeding, especially in high-risk surgical patients.[6,7] Nevertheless, the intermittent nature of bleeding or the presence of atherosclerotic plaque, tortuous vessels or anatomical variants (as in our first case study) may make embolisation attempts impossible.[2] Unlike cases of haemobilia involving branches of the hepatic artery, where the embolisation success rate is reasonably high (reaching up to 100%),[8] the success rate in pseudoaneurysms of the cystic artery is lower (40%).[7] The gallbladder must generally be removed following embolisation of the cystic artery due to the risk of gallbladder ischaemia, although some cases not requiring cholecystectomy but with good outcomes have been reported.[7]

Despite the possibility of angiographic treatment, most authors consider laparoscopic cholecystectomy with ligation of the cystic artery to be the treatment of choice. The patient's situation or technical complications may require use of or conversion to an open surgical procedure.

To conclude, the treatment of choice is cholecystectomy with ligation of the cystic artery, preferably using a laparoscopic approach, although angiographic treatment is an ideal alternative for haemodynamically stable, high-risk surgical patients.

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Autoimmune severe acute fulminant hepatic failure (FHF) during pregnancy☆

Insuficiencia hepática aguda grave (IHAG) fulminante de origen autoinmune en gestante

Acute liver failure (ALF) is an uncommon syndrome due to severe deterioration of liver cell function that may require an emergency liver transplantation (ETx). Clinical signs include a decrease in prothrombin index (PI) (<40%) and hepatic encephalopathy. Severe ALF can be fulminant when it appears within <2 weeks of clinical onset or subfulminant if it appears within 2–8 weeks.[1] In Spain, approximately one third of ALF cases are due to hepatitis B virus (HBV) infection, although around 5% are due to autoimmune hepatitis (AIH). This figure, however, is underestimated since cases caused by non-classical phenotype AIH are classified as cryptogenic ALF.

Our case study is a 29-year-old female patient, with no significant medical history, who was 20 weeks pregnant. She was referred to the hospital after suffering from jaundice and choluria for 2 days. She had no risk factors for hepatic diseases and said she had not travelled recently or consumed any drugs/hepatotoxic substances. She had yellow discolouration of the skin and sclera (jaundice) but was alert and oriented and had no asterixis. At admission, her lab test results were: AST 3205 IU/ml, ALT 2664 IU/ml, direct bilirubin 12.9 mg%, indirect bilirubin 3.1 mg%, leukocytes 13,820/mm^3, INR 1.91 and PI 43% (Fig. 1). The Doppler ultrasound was normal. The Obstetrics department confirmed it was a normal pregnancy. Hepatotropic virus results (HAV, HBV, HCV, HDV, HEV, CMV, EBV and herpes simplex) were negative, while autoantibody screen results (ANA-1/320, SMA-1/320 and AMA-1/320 and IgG–IgA–IgM–IgE 1870–235–79–124 mg/dl) were positive. Other serological test results (copper, ceruloplasmin and α-1 antitrypsin) were normal. No percutaneous/transjugular liver biopsy (biopsy) was performed due to abnormal clotting and foetal radiation. In accordance with consensus criteria,[2–5] a diagnosis of autoimmune hepatitis with overlap syndrome (ANA+, AMA+) was considered.[6] On day 5, a course of methylprednisolone was started (50 mg/24 h-IV/10 days followed by a tapering dose regimen). On day 6, the patient deteriorated: malleolar and pretibial oedema, grade II encephalopathy (*flapping* +), blood pressure 24.2 mg%, INR 1.8, PI 40%, factor V 72% and urine sediment normal. Her condition was classified as fulminant ALF with MELD score of 24[1] (Fig. 1). On day 8, encephalopathy was grade III and the possibility of an ETx was contemplated since 3 of the *King's College* criteria (non-A, non-B hepatitis; TB > 18 mg%; and duration of jaundice before onset of encephalopathy >7 days) were met. After 24 h, the patient improved (PI 54%, INR 1.5, TB 18.6 mg%, AST/ALT 307/665 IU/ml), ruling out an ETx. On day 26, once PI had returned to normal, a percutaneous biopsy was performed (Fig. 1): grade 2, stage 2 AIH, ''portal inflammation with necrosis of the limiting plate and bridging periportal fibrosis, with destruction of bile ducts and disappearance of interlobular bile ducts'' (Fig. 2). On day 43, the patient was discharged with normal obstetric

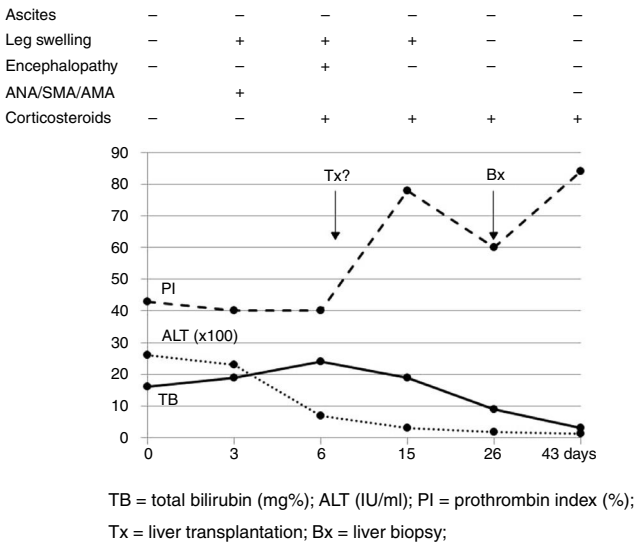

Figure 1 Progress during hospital stay. PI, prothrombin index (%); TB, total bilirubin (mg%); Bx, liver biopsy; ALT (IU/ml); Tx, liver transplantation.

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