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## CASE REPORT

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# Migration of Glue Cast into Biliary System Following Embolisation of a Mycotic Pseudoaneurysm of the Hepatic artery

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### ABSTRACT

*Pseudoaneurysm of the hepatic artery is at high risk of rupture, and thus requires early aggressive management. Patients usually present with haemobilia, upper gastrointestinal bleeding, and jaundice. Pseudoaneurysms are commonly caused by trauma, iatrogenesis, or vasculitis. Transcatheter embolisation is the mainstay treatment for intrahepatic pseudoaneurysm. We report a rare complication of migration of glue cast into the bile duct following embolisation of a hepatic pseudoaneurysm with glue.*

*Key Words: Aneurysm, false; Embolization, therapeutic; Hepatic artery*

## 中文摘要

### 肝臟真菌性假動脈瘤栓塞術後膠水移入膽道系統

楊子慧、梁安祥、陳崇文

肝臟假動脈瘤破裂的風險很高，因此要及早進行積極治療。患者通常出現血友病，上消化道出血和黃疸。假動脈瘤通常由創傷、醫源性或血管炎引起。經導管栓塞術是治療肝內假動脈瘤的主要手段。本文報告一例罕見肝假動脈瘤栓塞術後膠水移入膽道系統的併發症。

### INTRODUCTION

Pseudoaneurysm of the hepatic artery is at high risk of rupture, and thus requires early aggressive management. Patients usually present with haemobilia, upper gastrointestinal bleeding, and jaundice. Pseudoaneurysms are commonly caused by trauma, iatrogenesis, or vasculitis. Transcatheter embolisation is the mainstay treatment for intrahepatic pseudoaneurysm. We report a rare complication of migration of glue cast into the bile duct following embolisation of a hepatic pseudoaneurysm with glue.

### CASE REPORT

In March 2013, a 56-year-old woman with a history of recurrent pyogenic cholangitis presented with high fever, hypotension, and symptoms of anaemia. She had undergone cholecystectomy and choledochojejunostomy 10 years earlier. On admission, her liver function was deranged with total bilirubin of 36 mg/dl, alkaline phosphatase of 143 U/l, and alanine aminotransferase of 35 U/l. Complete blood count revealed elevated white cell count of  $22 \times 10^9/l$ , haemoglobin of 4.8 g/dl, platelet count of  $109 \times 10^9/l$ . Clotting profile was mildly

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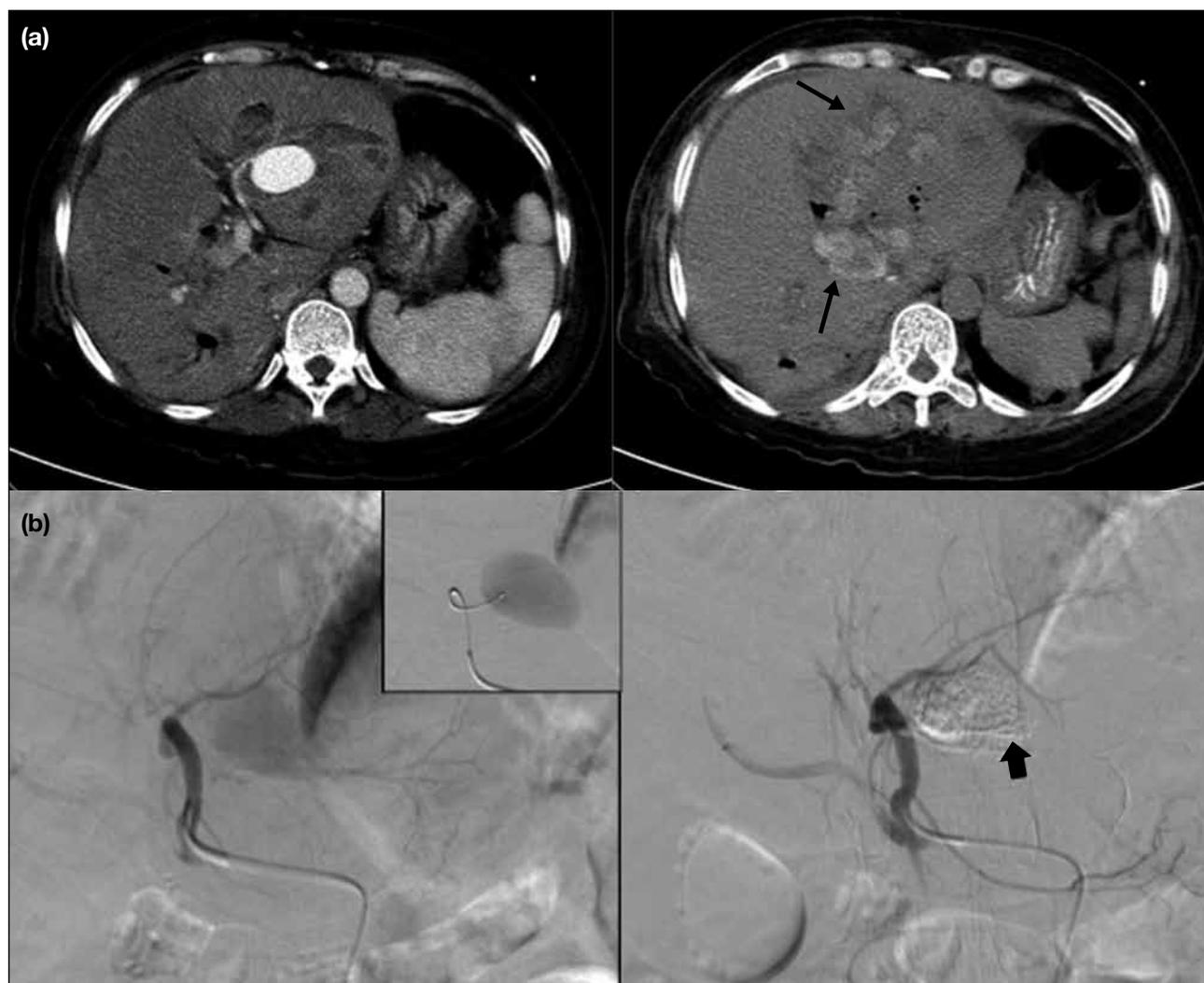
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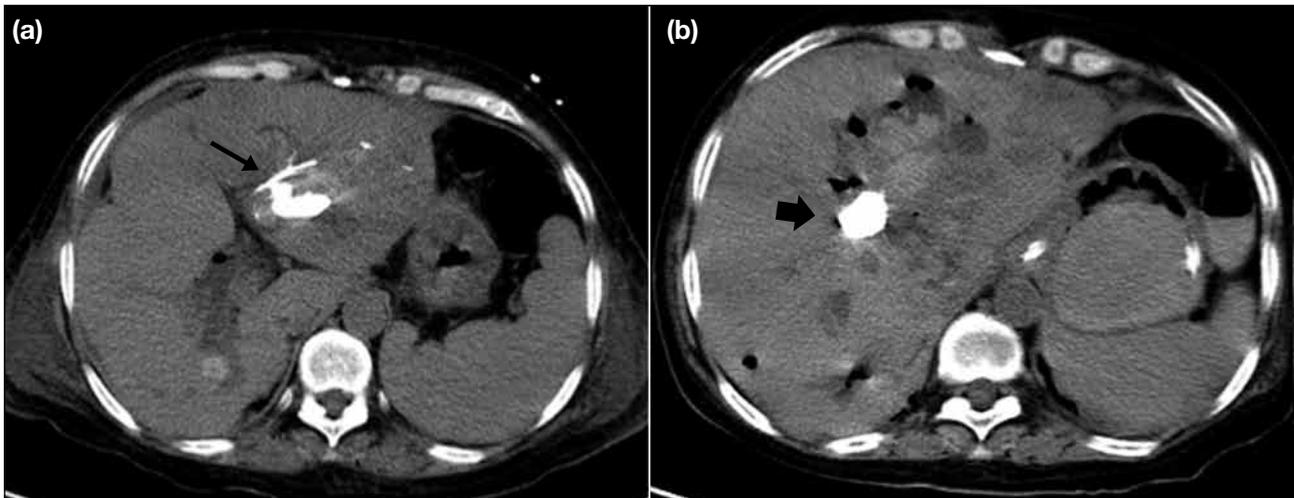
deranged with an international normalised ratio of 1.5. The patient was admitted to an intensive care unit in view of her unstable haemodynamic condition and early pattern of disseminated intravascular coagulation. Urgent oesophagogastroduodenoscopy revealed blood coming from the ampulla of Vater, with no bleeding source identified in the upper gastrointestinal tract. Urgent computed tomography (CT) showed multiple stones in bilateral dilated intrahepatic ducts, cholangitic abscesses in the left hepatic lobe, and a 3-cm pseudoaneurysm arising from segment II of the hepatic artery associated with haemobilia. Angiography confirmed a pseudoaneurysm arising from segment II hepatic artery. Superselective transcatheter embolisation with n-butyl cyanoacrylate diluted in

ethiodised oil (25% mixture) was performed. The glue cast filled the branches of the left hepatic artery connecting to the pseudoaneurysm, as well as the neck and the sac of the pseudoaneurysm. Post-procedural angiography demonstrated complete obliteration of the pseudoaneurysm, with preservation of most of the left hepatic artery branches (Figure 1).

Three days later, CT revealed a decreased amount of haemobilia and associated biliary dilatation. A glue cast was in situ at segment II of the liver (Figure 2a). Laparotomy and left hepatectomy were planned initially, but were not performed due to unstable intra-operative condition and marked peritoneal adhesions. The patient was treated conservatively with intravenous antibiotics.



**Figure 1.** (a) Computed tomography and (b) angiography showing a 3-cm pseudoaneurysm arising from segment II hepatic artery associated with extensive haemobilia (arrows) and its complete obliteration (thick arrow) after embolisation, with preservation of most left hepatic artery branches.



**Figure 2.** Computed tomography at (a) day 3 and (b) month 1 showing migration of the glue cast from segment II of the liver (arrow) into bilateral hepatic ductal confluence (thick arrow) resulting in biliary obstruction.

Endoscopic retrograde cholangiopancreatography was performed to remove stones in bilateral dilated intrahepatic ducts.

One month later, the patient complained of increasing right upper abdominal pain and jaundice. Repeat CT showed migration of the glue cast from the segment II region to the central part of the liver, corresponding to the bilateral hepatic ductal confluence. It was associated with progressively dilated left intrahepatic ducts (Figure 2b). No recurrence of pseudoaneurysm or cholangitic abscess was detected. Oesophagogastroduodenoscopy / retrograde cholangiopancreatography with a choledochoscopic approach confirmed the glue cast located at the opening of the left sectorial duct. The glue cast was removed endoscopically.

One month later, the patient underwent laser lithotripsy for residual biliary stones via the choledochoscopic route. She had no recurrence of symptoms after 3 years of follow-up.

## DISCUSSION

Hepatic artery pseudoaneurysm is at high (80%) risk of rupture if not treated.<sup>1</sup> The risk of rupture of a pseudoaneurysm is higher than that of a true aneurysm of comparable size due to poor support of the aneurysm wall. A pseudoaneurysm is formed when there is a breach in the vessel wall such that blood leaking through the wall is contained by the adventitia or surrounding perivascular soft tissue. Blood flows between the

vessel lumen and the aneurysm lumen through the hole in the vessel wall. Symptoms of pseudoaneurysm include haemobilia, upper gastrointestinal bleeding, and even haemoperitoneum. Pseudoaneurysm is commonly caused by trauma, iatrogenic insult including percutaneous and laparoscopic biliary procedures and vasculitis. In our patient, the pseudoaneurysm was presumed to be mycotic in origin secondary to recurrent pyogenic cholangitis. The diagnosis can be readily made by ultrasonography and CT. The role of angiography is mainly for delineation of vascular anatomy and subsequent treatment.

Transcatheter embolisation is the mainstay treatment for intrahepatic pseudoaneurysm as it is minimally invasive and highly effective. Other treatment options include direct imaging-guided percutaneous thrombin injection and endovascular-covered stent placement.<sup>2,3</sup> Embolisation should be performed superselectively to avoid non-target embolisation or liver necrosis. As the pseudoaneurysm is weak, any forceful injection during superselective catheterisation may lead to rupture and thus caution is needed. Permanent embolic agents should be used such as coils and tissue adhesive. The treatment goal is to occlude the parent vessel proximal and distal to the pseudoaneurysm so as to prevent blood supply to the front door and prevent collateral flow from the back door.

Packing the hepatic artery pseudoaneurysm sac with embolic agent is considered the treatment of choice.<sup>4,5</sup>

However, the weak pseudoaneurysm may be unable to hold the embolic agent. A ruptured pseudoaneurysm during coil packing necessitates emergency surgery. In addition, erosion of embolic agent into the biliary tree can be related to sac packing. Thus, we consider this technique to be unpredictable and inferior to proximal and distal coil packing of the hepatic artery.

Complications of embolisation include deranged liver function, liver and / or gallbladder necrosis due to devascularisation, abscess formation, and liver rupture. Embolic agent erosion into the biliary tree and migration is a rare complication.<sup>6</sup> To the best of our knowledge, this is the first such complication associated with the use of glue. Migration of the glue was possibly due to a small arterial-biliary fistula that caused haemobilia initially. With time, the fistula may enlarge with erosion of the glue into the biliary tree. The concurrent inflammation in the vicinity may have accentuated progression of the arterial-biliary fistula. The glue inside the biliary tree can cause biliary obstruction and stone formation and thus necessitates removal. To avoid such

complication, pseudoaneurysmal sac packing should be avoided, and attention should be paid to the more central hepatic artery pseudoaneurysms, which are close to the bile ducts.

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