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Case Report

Interventional Therapy for the Treatment of Severe Hemobilia After Percutaneous Transhepatic Cholangial Drainage: A Case Series

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From May 2003 to May 2010, a total of 9 patients with severe hemobilia after percutaneous transhepatic cholangial drainage (PTCD) were diagnosed using superselective angiography and cholangiography, and then were treated with interventional procedures. Two patients with hepatic arterio-biliary fistula underwent proximal and distal arterial embolization of the responsible vessel. Six patients with pseudoaneurysm had pseudoaneurysm occlusion with proximal and distal embolization. Another patient with biliary-portal vein fistula received a biliary fully covered stent placement. The effects in these patients were evaluated using superselective angiography immediately after the intervention and at 3- and 6-month follow-up. In all patients, hemobilia was stopped right after the treatment and no sign of recurrence was noted at 3- and 6-month follow-up after the interventional therapy. Our findings demonstrate that interventional therapy is a simple, minimally invasive, and safe approach for treating severe hemobilia in patients receiving PTCD.

Key Words: Interventional therapy – Hemobilia – Percutaneous transhepatic cholangial drainage – Transcatheter arterial embolization

Percutaneous transhepatic cholangial drainage (PTCD) has been proven to be one of the most important treatments for benign and malignant obstructive jaundice. However, severe hemobilia can occur after PTCD, consequently causing life-threatening hemorrhagic shock, which has a high mortality rate (nearly 50%).[1] In a retrospective study performed by Rivera-Sanfeliz *et al*, the incidence of

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severe hemobilia was 2.31% in 346 patients receiving PTCD.[2] Damages or abnormal connections of intrahepatic vessels resulting from PTCD, namely, arterio-biliary fistula, biliary venous fistula, or pseudoaneurysm are the major causes of this hemobilia.[3] Additionally, coagulation disorder in patients with obstructive jaundice is also a recognized cause.[4]

For clinicians, the difficulties in diagnosis and treatment of severe hemobilia following PTCD lie in detecting the specific site of bleeding and choosing a correct surgical method. In recent years, superselective angiography has been developed as an effective method for the diagnosis of hemobilia, which is able to detect significant hemobilia in over 90% of patients.[5] Surgery, the predominant solution for the treatment of this bleeding, can effectively treat the primary lesions and completely remove the blood clots from the biliary in order to avoid obstruction or stone formation. Most patients with severe hemobilia after PTCD, however, have poor general health and unstable vital signs, greatly impairing their tolerance to anesthesia and surgery for the treatment of the hemobilia.[5,6] Thus, surgery may lead to exacerbation of the disease or even death of the patient during the hemostatic treatment. Therefore, an alternative solution to effectively and safely stop severe hemobilia following PTCD must be available to clinicians.

Interventional therapy has been tested as an effective approach for treating bleeding associated with severe hemobilia.[7,8] Nevertheless, few reports about the efficacy and safety of this therapy exist in the literature. In our hospital, from May 2003 to May 2010, 9 patients with severe hemobilia following PTCD received interventional therapy. The clinical results in these patients are presented in this report.

Materials and Methods

Patient population

Nine patients were included. Five were male (55.6%) and 4 were female (44.4%). The median age was 46 years (range, 22–79 years). All patients had obstructive jaundice and received PTCD. The etiology of obstructive jaundice was as follows: cholangiocarcinoma (n = 3), pancreatic carcinoma (n = 2), metastatic gastric carcinoma (n = 2), and choledocholithiasis (n = 2). Common clinical presentations of hemobilia after PTCD included persistent or recurrent abdominal pain, upper gastrointestinal bleeding, intermittent massive hemorrhagic bile from the drainage tube, no alleviation of jaundice and possibly even aggravation, hemorrhagic shock, fever, and melena (Table 1). Additional signs of severe biliary tract infection included recurrent high fever (>103 F°) and chills, observed in 1 patient. No patients were able to tolerate surgery owing to poor general conditions.

Procedures

Superselective hepatic arteriography was performed in all patients to detect bleeding lesions as well as the rate of bleeding. The portal vein was also monitored with hepatic arteriography at delayed phase and cholangiography. Out of 9 patients, 2 were diagnosed with hepatic arterio-biliary fistula, 6 were diagnosed with pseudoaneurysm, and 1 was diagnosed with biliary-portal vein fistula. For the 2 patients with arterio-biliary fistula and the 5 patients with pseudoaneurysm, gelatin sponge (Gelfoam, Pfizer, New York, NY) and detachable platinum coils (GDC, Boston Scientific, Natick, MA) were used to embolize the proximal and distal ends, respectively, of the targeted bleeding vessel through superselective catheterization. Another patient with pseudoaneurysm at the end of the right hepatic artery initially received gelatin sponge and platinum coil embolization of the right hepatic artery. However, recurrent hemobilia was detected 10 days later. Angiography showed that another small branch adjacent to the pseudoaneurysm was responsible for the bleeding. Due to the small diameter of this vessel, embolization with Glubran 2 acrylic glue (GEM, Viareggio, Italy) was applied instead of gelatin sponge or coil. For the patient with biliary-portal vein fistula, a biliary fully covered stent (WallFlex RX, Boston Scientific, Natick, MA) was placed to close the fistula. Repeat angiographies were performed in all patients 1 day after intervention and at 3- and 6-month follow-up to determine whether absolute hemostasis was obtained.

Results

The outcomes of all patients after interventional therapy are summarized in Table 1. The angiographies of pre-intervention and post-intervention in the representative patients with hepatic arterio-biliary fistula, pseudoaneurysm, and biliary-portal vein fistula are shown in Fig. 1. Three- and 6-month follow-up examinations showed that absolute hemostasis was achieved in all patients, and no severe complications were observed during the follow-up period.

Table 1 The outcomes of interventional therapy for the treatment of severe hemobilia after PTCD in all patients

Patient	Histopathology	Cause of hemobilia	Materials for embolization	Whether absolute hemostasis was achieved at 3- and 6-month follow-up	Complications related to embolization[a]
1	CCC	AF	Gelatin sponge + coil	Yes	Fever, elevation of liver aminotransferases
2	CDL	PA	Gelatin sponge + coil	Yes	Transient abdominal pain
3	CCC	AF	Gelatin sponge + coil	Yes	Elevation of liver aminotransferases, transient abdominal pain
4	CDL	PA	Gelatin sponge + coil	Yes	N/A
5	PAN	PA	Gelatin sponge + coil + Glubran 2 acrylic glue[b]	Yes	Elevation of liver aminotransferases, transient abdominal pain, fever
6	PAN	PA	Gelatin sponge + coil	Yes	Elevation of liver aminotransferases, transient abdominal pain, fever
7	CCC	BVF	Biliary stent	Yes	Elevation of liver aminotransferases
8	MGC	PA	Gelatin sponge + coil	Yes	Transient abdominal pain, fever
9	MGC	PA	Gelatin sponge + coil	Yes	Fever, elevation of liver aminotransferases

AF, arterio-biliary fistula; BVF, biliary-portal vein fistula; CCC, cholangiocarcinoma; CDL, choledocholithiasis; MGC, metastatic gastric carcinoma; N/A, not applicable; PA, pseudoaneurysm; PAN, pancreatic carcinoma.

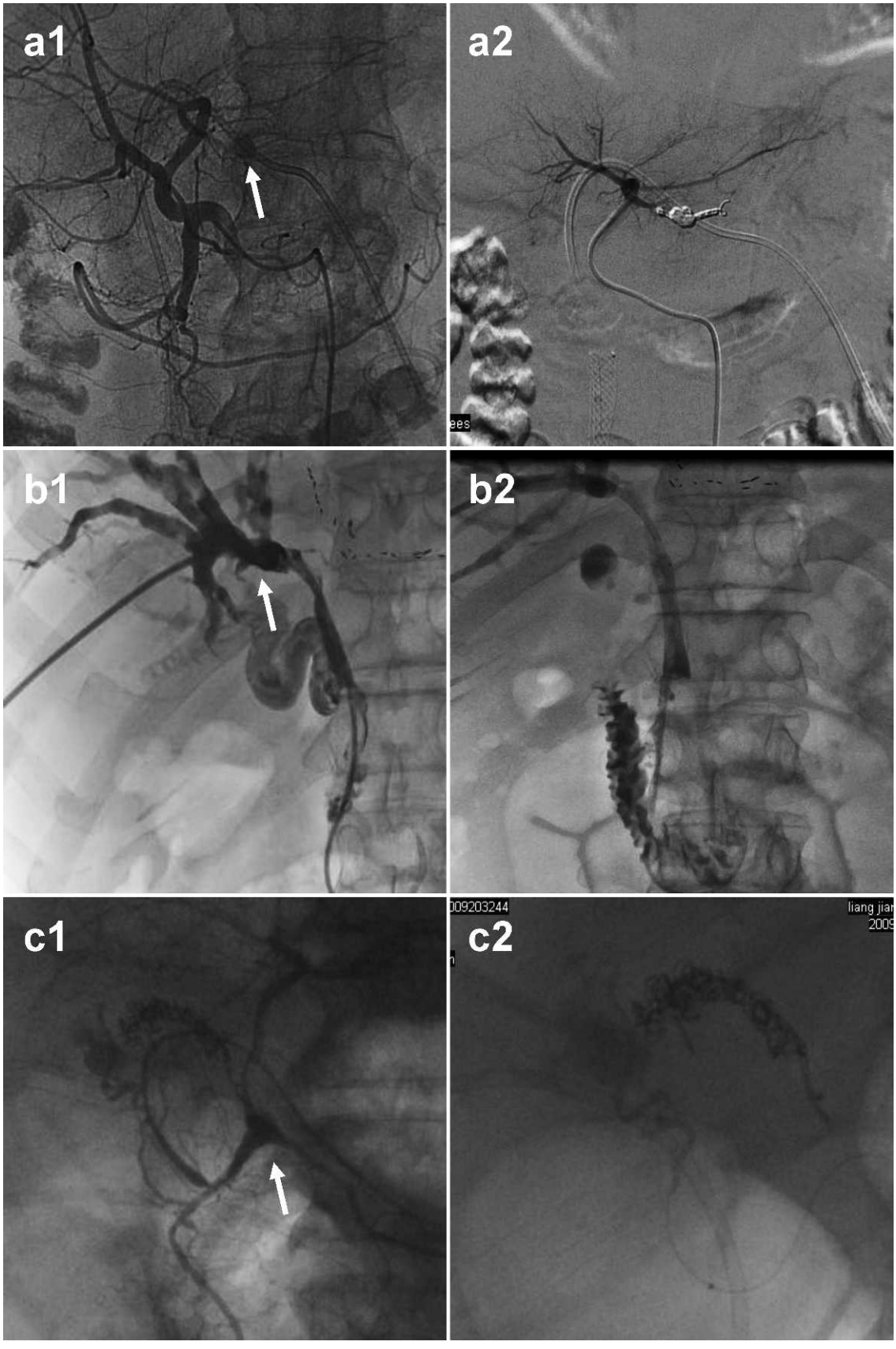
[a]Complications occurring were finally alleviated by symptomatic treatment.

[b]Glubran 2 acrylic glue was applied 10 days after the embolization with gelatin sponge and coil.

Discussion

Selective hepatic arteriography is considered the preferred method for diagnosis of severe hemobilia because of its high sensitivity, being capable of immediately and accurately detecting bleeding at such low rates as approximately 0.5 to 2 mL/min.[9] The diagnostic accuracy has been reported to be approximately 85% to 95%. Cholangiography, performed by introducing angiographic media through the biliary drainage tube, is mainly used to detect biliary-portal vein fistula but is another way to diagnose hemobilia. Therefore, for the patients with severe hemobilia after PTCD, in parallel to the immediate supportive treatments such as blood transfusion, hepatic arteriography and cholangiography should be conducted as early as possible to detect the primary hemorrhage site. Ultrasonography, computed tomography (CT), and duodenoscopy are potential alternatives in cases where the selective hepatic arteriography and cholangiography are not available, but the bleeding lesion is usually difficult to position accurately with these methods.[10]

In recent years, superselective hepatic artery embolization has been accepted as an effective interventional therapy for the treatment of severe hemobilia.[5] Its efficacy and safety have been evaluated by several studies,[8,9] indicating it can offer the same advantages as minimally invasive therapy such as a smaller incision, fewer complications, and a faster recovery; thus it is more appropriate for patients with poor general health and unstable vital signs, specifically patients with hemorrhagic shock. Embolizing both the distal and proximal parts of the bleeding artery simultaneously is currently recognized as the standard procedure by most researchers and clinicians because it effectively prevents recurrent hemorrhage caused by the countercurrent flow of blood through a branch to the bleeding artery as well as controls the initial hemorrhage.[5]



The materials used for embolization include gelatin sponge, absolute ethanol, n-butyl-2-cyanoacrylate (NBCA), detachable balloon, and coils. Gelatin sponge and coils, the most commonly used materials, usually are applied in combination because embolization with only gelatin sponge rarely achieves absolute hemostasis and easily induces recurrent hemorrhage.[11,12] Therefore, for the patients with hepatic arterio-biliary fistula and pseudoaneurysm in our study, gelatin sponge and platinum coil were combined for hepatic artery embolization, and absolute hemostasis was achieved in all patients after 3- and 6-month follow-up. However, it should be noted that Glubran 2 acrylic glue was also used in a patient with pseudoaneurysm in addition to the combination of gelatin sponge and platinum coil. Glubran 2 acrylic glue, as a liquid nonirritating embolization agent with low viscosity, is able to enter and embolize the small and circuitous bleeding artery branch in the cavity of pseudoaneurysm, effectively preventing compensatory blood supply and thus resulting in absolute hemostasis. In this patient, another small branch adjacent to the pseudoaneurysm was detected to be responsible for the recurrent hemobilia after the embolization with gelatin sponge and coil. Considering the thin diameter and circuitous nature of this branch, Glubran 2 acrylic glue was applied in order to achieve absolute hemostasis.

Only one patient was diagnosed as having biliary-portal vein fistula, which has a very low incidence rate in patients receiving PTCD. Surgery is the generally accepted treatment for this complication, though there are few reports on its efficacy. In our study, this patient received the embolization with a biliary fully covered stent, and absolute hemostasis was obtained after the embolization without any recurrent hemobilia at 3- and 6-month follow-up. According to the authors' experience, biliary-portal vein fistula often occurs between the choledochus and the main portal vein, thus allowing the placement of a biliary stent for embolizing orificium fistulae. Therefore, considering its efficacy in controlling hemorrhage, embolization with a biliary stent may be recognized as a new and effective method for the treatment of biliary-portal vein fistula. Nevertheless, further studies and more cases are needed to investigate and confirm the clinical efficacy of this approach.

In our study, the complications associated with embolization included transient abdominal pain, fever, and elevation of liver aminotransferases, which could be alleviated by symptomatic treatment. More serious complications, including necrosis in the embolization area, hepatic failure, ectopic embolization, gastrointestinal hemorrhage, and septicemia, have been reported by previous studies but were not observed in patients of our study.[13] In conclusion, our results indicate that interventional therapy is an effective and safe method for the treatment of severe hemobilia after PTCD because of the advantages including a smaller incision, fewer complications, and faster recovery. We conclude that it should be used as a preferable approach for the patients with poor general health conditions.

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Fig. 1 The angiographies of pre-intervention and post-intervention in the representative patients. (a1) A pseudoaneurysm with the size of 31 × 25 mm was detected at the end of the left hepatic artery (arrow). (a2) Embolization with gelatin sponge and coil at the distal and proximal parts of the artery with no recurrent hemobilia observed. (b1) A biliary-portal vein fistula was detected by cholangiography (arrow). (b2) A biliary fully covered stent was placed to embolize orificium fistulae, and absolute hemostasis was achieved. (a3) A small and circuitous branch was detected to be responsible for the recurrent hemorrhage of a pseudoaneurysm after embolization with gelatin sponge and platinum coil (as shown by arrow). (b3) Hemorrhage was controlled after the embolization with Glubran 2 acrylic glue.

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