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CASE REPORT

Massive hemobilia following plastic stent removal in common bile duct cancer associated with primary sclerosing cholangitis (with video)

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Abstract

Hemobilia is defined as bleeding into the biliary tract. Herein, we report a very rare case of massive hemobilia following plastic stent (PS) removal in common bile duct (CBD) cancer. A 72-year-old man with primary sclerosing cholangitis had undergone repeated insertion of a PS into the CBD. Biliary tract biopsy was performed based on suspicion of combined CBD cancer. Biopsy revealed poorly differentiated adenocarcinoma of the CBD. One month after the biliary tract biopsy, he was admitted for acute cholangitis, and endoscopic retrograde cholangiography was performed for the exchange of the PS. When one of the two biliary PSs was removed, spurting bleeding from the major papilla began abruptly. The massive bleeding caused the patient to be in a pre-shock state. A retrieval balloon catheter was compressed against the papilla for hemostasis. Although he was treated conservatively, the patient developed a bloody discharge. Upper gastrointestinal endoscopy revealed that the pulsatile bleeding beside the PSs started immediately after the removal of the coagula. Emergent contrast-enhanced computed tomography showed right hepatic artery aneurysm across the CBD. Therefore, transarterial embolization was performed. The patient's post-therapeutic course was uneventful. He received chemotherapy, but died about a half year after hemobilia occurred.

Keywords Hemobilia · Plastic stent · Biliary tract biopsy · Primary sclerosing cholangitis · Common bile duct cancer

Introduction

Hemobilia, which is defined as bleeding into the biliary tract, was first named by Sandblom in a paper reporting post-traumatic hemorrhage into the biliary tract in 1948 [1]. The cause of hemobilia is predominantly iatrogenic, resulting from percutaneous liver intervention, surgery, and endoscopic retrograde cholangiopancreatography (ERCP). Here, we report a case of massive hemobilia following plastic stent (PS) removal in a patient with common bile duct (CBD) cancer associated with primary sclerosing cholangitis (PSC).

Case report

A 72-year-old man, who was diagnosed as having PSC 1 year previously (Fig. 1a), had undergone repeated insertion of a PS into the CBD for the treatment of obstructive jaundice. While two biliary PSs (GEPD-5-12, Cook Ireland, Ltd., Limerick, Ireland) were being exchanged, biliary tract biopsy using biopsy forceps (E634043, Olympus Medical Systems, Tokyo, Japan) was performed based on the suspicion of CBD cancer due to the progression of CBD stenosis (Fig. 1b, c). The result of the biopsy indicated a poorly differentiated adenocarcinoma of the CBD (Fig. 1d).

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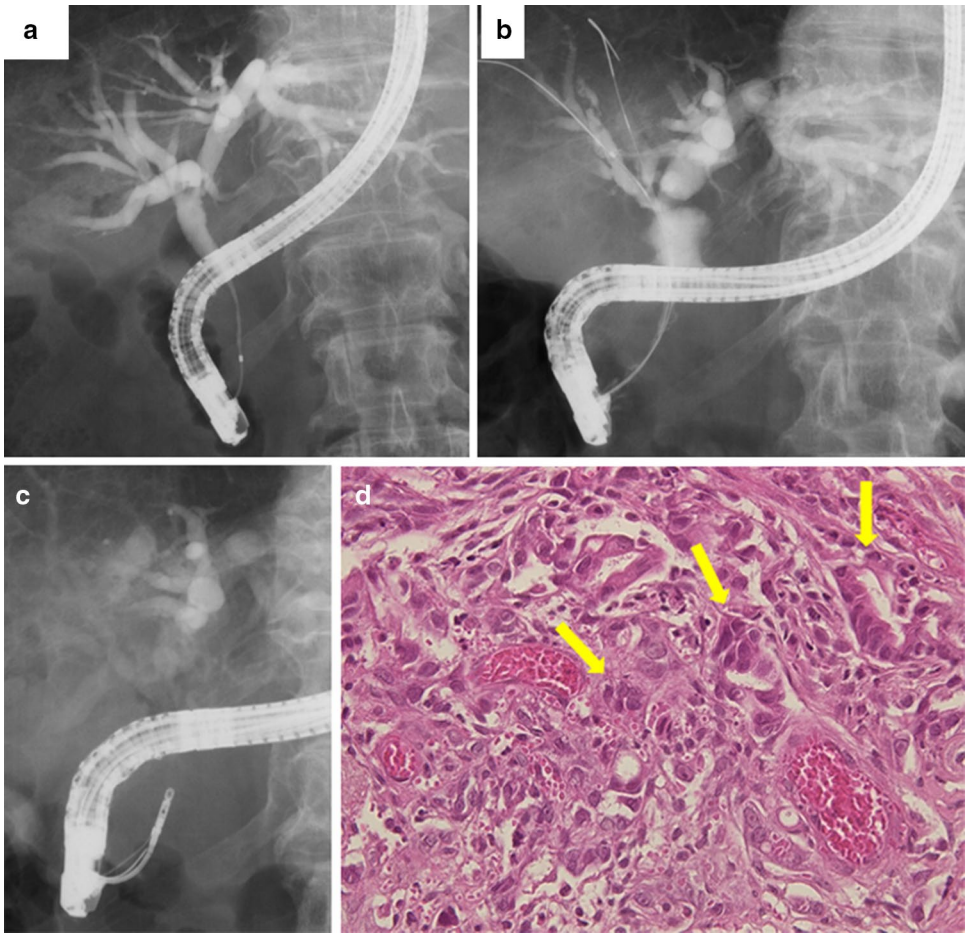

Fig. 1 **a** Endoscopic retrograde cholangiography (ERC) for the diagnosis of primary sclerosing cholangitis showed stricture of the lower common bile duct (CBD) and origin at the right hepatic duct. **b** ERC on the last exchange of the plastic stent (PS) showed progression of CBD stenosis. **c** Following ERC in Fig. 1b, biliary tract biopsy was performed using biopsy forceps at the stricture of CBD. **d** Microscopic image of the biliary tract biopsy showing that small cells with clear nucleolus formed a small glandular cavity structure with palisade arrangement (arrow). Poorly differentiated adenocarcinoma of CBD was diagnosed

One month after the biliary tract biopsy, the patient was admitted owing to acute cholangitis. On the day following admission, ERCP was performed for the exchange of the PS. When one of the two biliary PSs was removed using a snare (Fig. 2a), pulsatile bleeding from the major papilla began abruptly (Fig. 2b, Video 1). As a result of the massive bleeding, the patient went into a pre-shock state owing to decreased blood pressure and tachycardia. During the bleeding, cannulation of the major papilla was performed and a guidewire was inserted into the biliary tract. A retrieval balloon catheter (B-V232P-B, Olympus Medical Systems, Tokyo, Japan) was compressed against the major papilla for hemostasis (Fig. 2c). After cessation of bleeding from the major papilla, one PS (Flexima Biliary Stent with Delivery System, 7FrX7cm, BOSTON SCIENTIFIC CORP., Marlborough, MA, USA) was reinserted into the biliary tract (Fig. 2d).

Despite conservative treatment, the patient developed bloody discharge on day 11 after ERCP. Upper gastrointestinal endoscopy showed that the PSs were covered by fresh coagulated blood, and the pulsatile bleeding beside the PSs started immediately after the removal of the coagula. It was judged that endoscopic hemostasis would be difficult. Emergent contrast-enhanced computed tomography (CT) showed a right hepatic artery aneurysm across the CBD (Fig. 3a). Therefore, transarterial embolization (TAE) with coils was performed on day 13 post-ERCP (Fig. 3b, c). A self-expandable metal stent (SEMS) (Bonastent® full cover stent, 8 mm × 7 cm, Sewoon Medical Co., Ltd., Cheonan-si, Chungcheongnam-Do, Korea) was then inserted into the CBD on day 15 post-ERCP (Fig. 4a, b). The patient's post-therapeutic course was uneventful, and he was discharged on day 44 post-ERCP.

The patient subsequently showed no sign of bleeding and he received chemotherapy (gemcitabine monotherapy) in an outpatient clinic. However, he was moved to the palliative care unit because of a worsening performance status and a severe decrease in platelet count. He ultimately died about a half year after ERCP.

Discussion

Insertion of PS is a very popular procedure that is usually performed as the first choice for biliary drainage. However, we observed a life-threatening case of hemobilia that

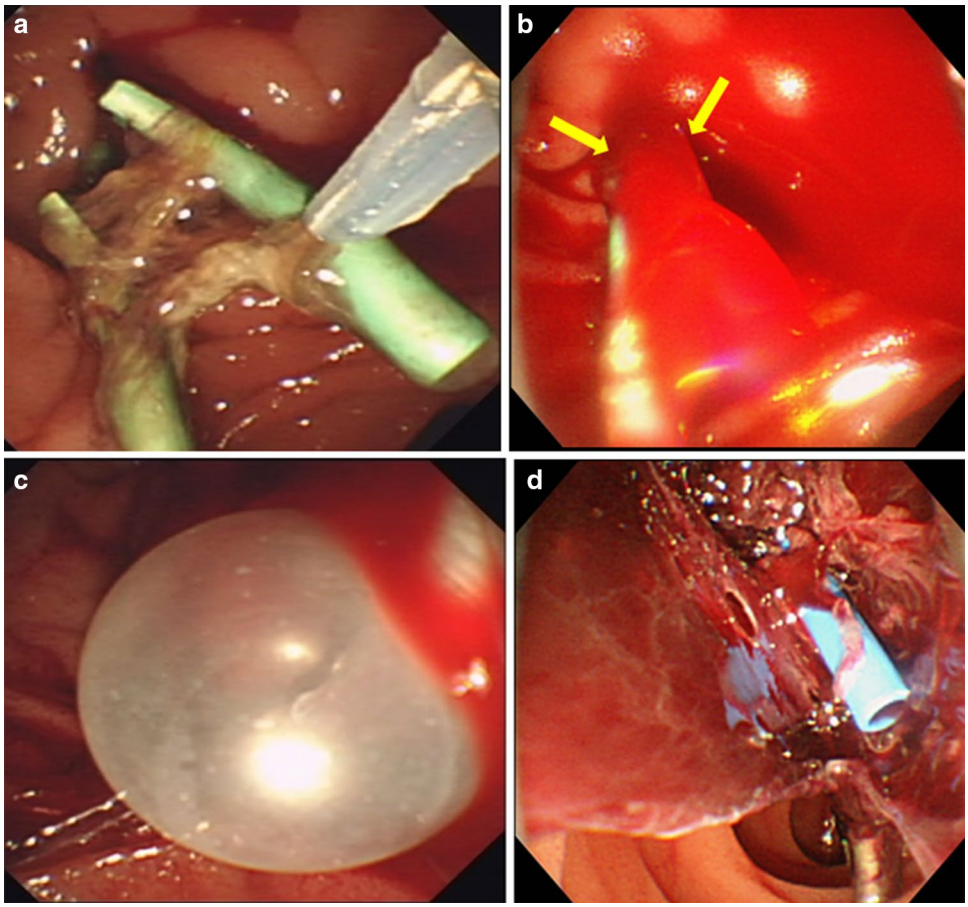

Fig. 2 **a** Endoscopic retrograde cholangiography was performed on the day following admission. One of the two biliary plastic stents (PS) was removed using a snare. **b** Pulsatile bleeding (arrow) from the major papilla began abruptly. **c** Retrieval balloon catheter (B-V232P-B, Olympus Medical Systems, Tokyo, Japan) was compressed against the major papilla for hemostasis. **d** After cessation of bleeding from the major papilla, a PS (Flexima Biliary Stent with Delivery System, 7 Fr×7 cm, BOSTON SCIENTIFIC CORP., Marlborough, MA, USA) was reinserted into the biliary tract

occurred with removal of PS in a patient with CBD cancer associated with PSC. We performed a literature search to identify cases of hemobilia associated with PSs, and found only 13 cases of hemobilia associated with PSs, which demonstrates that it is very rare (Table 1) [2–12]. We analyzed these cases in detail and have related it to our case.

The cases consisted of 5 men and 8 women, aged 25–82 years. The underlying diseases for hemobilia associated with PSs were malignant tumor (5 cases), choledocholithiasis (2 cases), extrahepatic portal venous obstruction (EHPVO, 3 cases), benign biliary tract stricture (2 cases), and bile duct leakage after living-donor liver transplantation (LDLT, 1 case). The tips of the PSs were either of a pigtail type (5 cases) or of a straight type (3 cases). The caliber of PSs was 7–11.5 Fr and the number of indwelling PSs was 1–3. The indwelling site of the PSs was right hepatic duct (3 cases), left hepatic duct (2 cases), and CBD (2 cases). The onset of hemobilia from the time of PS insertion was 2 weeks to 1 year. The causes of hemobilia included hepatic artery aneurysm (6 cases), arterio-biliary fistula (2 cases), and bilio-portal fistula (2 cases). The modality to detect the source of hemobilia was contrast-enhanced CT (4 cases), angiography (2 cases), laparotomy (2 cases), and ERC (2 cases). The interventions for hemostasis were transarterial embolization (6 cases), SEMS insertion (2 cases), ligature suture of the vessel (2 cases), a balloon-tipped catheter to press (2 cases), PS insertion (1 case), and no intervention (1 case).

The underlying disease for our case was CBD cancer associated with PSC, which is in line with that malignant diseases most frequently underlie hemobilia associated with PS. PSs used in our case were two 5 Fr straight-type stents and placed in CBD. The previous reports showed that hemobilia induced by a PS is not related to the shape of tips, the number, or size of PSs. The indwelling sites were varied, however, the bleeding sites were almost all near the top of PSs. The onset of hemobilia from the time of PS insertion was not fixed. Hepatic artery aneurysm was the most frequent cause of hemobilia, including our case. One rare cause of hemobilia was fistula with portal vein, which was underlain mainly EHPVO. Therefore, it is also necessary to pay attention to portal vein obstruction or portal vein hypertension. The modality to detect the cause of hemobilia was mostly contrast-enhanced CT as for our case. However, we should reflect on the fact that we could not perform contrast-enhanced CT right after the first hemobilia for our case because of deterioration with the patient's condition due to rebleeding. If

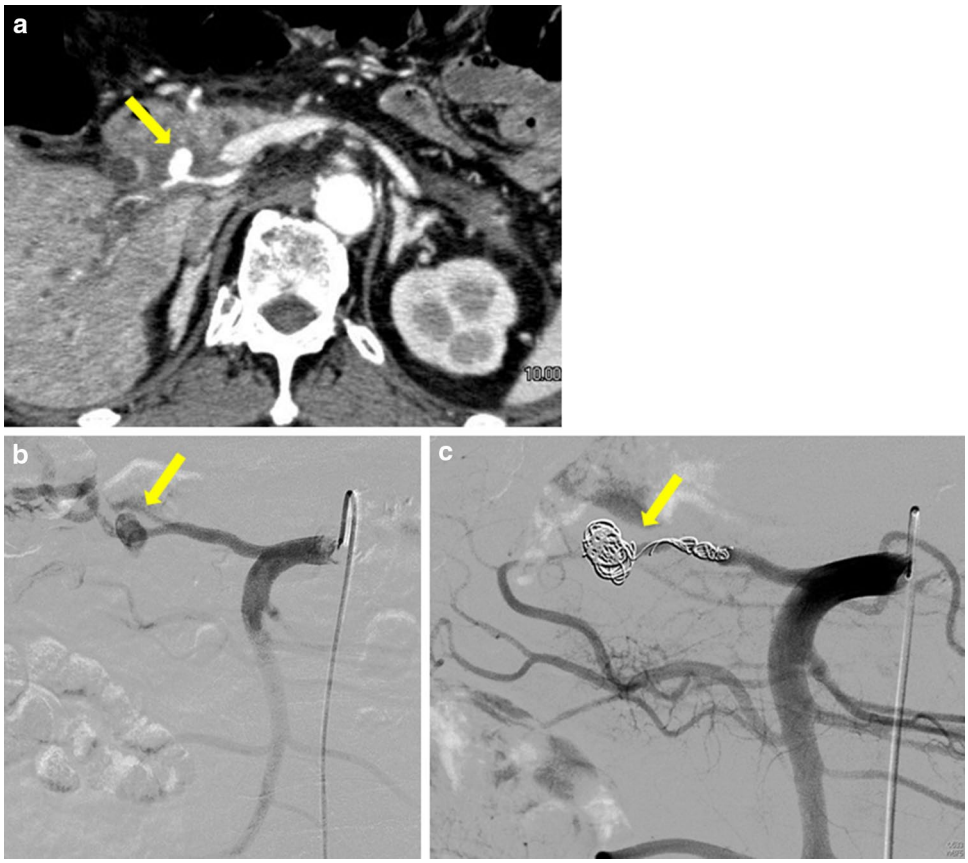

Fig. 3 **a** Emergent-enhanced computed tomography showing right hepatic artery aneurysm across the common bile duct (arrow). **b** Superior mesenteric arteriography showed an aneurysm of the replaced right hepatic artery (arrow). **c** Transarterial embolization was performed with coils, and the blood flow to the aneurysm was blocked (arrow)

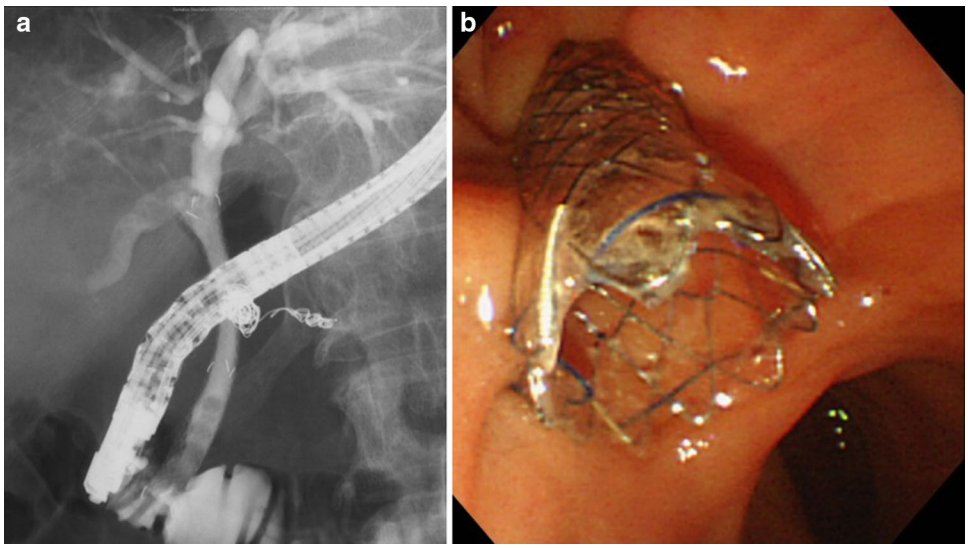

Fig. 4 **a, b** Self-expandable metal stent (Bonastent® full cover stent, 8 mm × 7 cm, Sewoon Medical Co., Ltd., Cheonan-si, Chungcheongnam-Do, Korea) was inserted into the common bile duct across the major papilla

immediate contrast-enhanced CT after the first hemobilia is performed, prompt diagnosis and appropriate treatment for the bleeding from the hepatic artery aneurysm would be accomplished. Finally, the most used treatment was TAE with coil. We also treated hepatic artery aneurysm with TAE with coil. On the other hand, it was reported that SEMS insertion or injection sclerotherapy with cyanoacrylate glue was performed against bleeding from the bilio-portal fistula.

Taken together, PS insertion might result in hemobilia by inducing hepatic artery aneurysm or fistula between the biliary tract and artery or portal vein regardless of the size, tip shape, number, indwelling site, and indwelling time of PSs. Therefore, when gastrointestinal bleeding happens in patients with PSs

Table 1 Reports of hemobilia due to plastic stents

	References	Age	Sex	Primary disease	Shape of tips	Caliber (Fr)	Indwelling site	Time to hemobilia	Cause of hemobilia	Modality to detect the source of hemobilia	Intervention for hemostasis
1	Yamauchi [2]	78	F	Benign biliary stenosis	Pigtail	NS	Left hepatic duct (B3)	2 weeks	Hepatic artery aneurysm	CE-CT	TAE with coil
2	Yasuda [3]	78	F	Choledocholithiasis	Pigtail	7	CBD	1 year	Hepatic artery aneurysm	CE-CT	TAE
3	Chun [4]	47	M	Liver abscesses and leakage at the bile duct anastomosis after LDLT	Pigtail/straight	7/11.5	Right hepatic duct	NS	Hepatic artery aneurysm	CE-CT	TAE with coil
4	Tonozuka [5]	82	M	Lower bile duct cancer after CRT	Straight	7	CBD	6 months	Hepatic artery aneurysm	Angiography	TAE with coil after placement of a SEMS
5	Inoue [6]	68	F	Klatskin tumor	NS	8.5	Right hepatic duct	20 days	Hepatic artery aneurysm	CE-CT	TAE with coil
6	Tsuji [7]	73	M	Hepatoma	Straight	10	NS	2 months	NS	NS	Insertion of a plastic stent
7	Frost [8]	63	F	Choledocholithiasis	Pigtail	NS	Duct of right posterior segment	10 months	Fistula between the posterior branch of the right hepatic artery and the duct of right posterior segment	Laparotomy	Ligature suture of a posterior branch of the right hepatic artery
8	Park [9]	62	F	Klatskin tumor after CRT	Pigtail	10	Left hepatic duct	2 months	Hepatic artery aneurysm	Angiography	TAE
9	Wolters [10]	75	M	CBD stricture due to extrinsic compression by pancreatic pseudocysts	NS	10	NS	3 weeks	Erosion of a major branch of the right hepatic artery	Laparotomy	Ligation of the spurting vessel
10	Mutignani [11]	39	M	EHPVO	NS	10	NS	1 month	A bilio-portal fistula	ERC	Insertion of a SEMS
11		35	F	EHPVO	NS	11.5	NS	1 month	NS	NS	None
12		25	F	EHPVO	NS	10/10/11.5	NS	3 months	A bilio-portal fistula	ERC	Inflation of a balloon-tipped catheter and injection of cyanoacrylate glue
13	Conio [12]	59	F	Metastatic ovarian carcinoma	NS	8	NS	NS	NS	NS	Inflation of a balloon-tipped catheter
14	Our case	72	M	CBD cancer and PSC	Straight	5/5	CBD	1 month	Hepatic artery aneurysm	CE-CT	TAE with coil

F female, *M* male, *NS* not shown, *CE-CT* contrast-enhanced computed tomography, *CBD* common bile duct, *TAE* transarterial embolization, *LDLT* living-donor liver transplantation, *CRT* chemoradiotherapy, *SEMS* self-expandable metal stent, *EHPVO* extrahepatic portal venous obstruction, *ERC* endoscopic retrograde cholangiography, *PSC* primary sclerosing cholangitis

inserted, contrast-enhanced CT should be performed as soon as possible after bleeding to identify abnormality of the vascular system. Moreover, it is necessary to consider radiological interventions, including TAE to treat hemobilia due to PSs.

In conclusion, we report a very rare case of massive hemobilia occurring after PS removal in the biliary tract. It should be recognized that PSs may cause hemobilia, which is induced by hepatic artery aneurysm or fistula between the biliary tract and artery or portal vein.

Compliance with ethical standards

Conflict of interest Hiroshi Miyamoto, Kumiko Tanaka, Fumika Nakamura, Takahiro Ikeda, Shinji Kitamura, Tetsuo Kimura, Koichi Okamoto, Masahiro Sogabe, Naoki Muguruma, Yoshimi Bando, and Tetsuji Takayama declare that they have no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments

Informed consent Informed consent was obtained from all patients for being included in the study.

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