CASE REPORT

Hemobilia due to hepatic artery pseudoaneurysm following biliary pigtail stent placement

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ABSTRACT

Hemobilia is a potentially fatal condition if not diagnosed and treated early. One of the rare causes of hemobilia is hepatic artery pseudoaneurysm. Herein, we present a case of hemobilia caused by hepatic artery pseudoaneurysm rupture. A patient with a benign stricture in the distal common bile duct who had undergone multiple endoscopic retrograde cholangiopancreatography (ERCP) procedures had a pigtail stent placed during the last ERCP. Ten days after the procedure, the patient presented to our emergency department with abdominal pain and was admitted with a diagnosis of acute pancreatitis. During follow-up, the patient underwent another ERCP due to suspected cholangitis, and severe hemobilia was observed following removal of the pigtail stent. Computed tomography (CT) angiography revealed a hepatic artery pseudoaneurysm, which was successfully treated with coil embolization. Patients with hemobilia may present with symptoms of upper gastrointestinal bleeding as well as conditions such as cholangitis and pancreatitis due to hepatic artery pseudoaneurysm rupture. Coil embolization is the first-line of interventional to stop bleeding. If this method is insufficient, surgical treatment may be considered.

KEYWORDS: hemobilia; hepatic artery pseudoaneurysm; biliary stent

INTRODUCTION

Hepatic artery pseudoaneurysm (HAP) accounts for 20% of nontraumatic abdominal visceral artery pseudoaneurysms [1]. HAPs are extrahepatic in 75% and intrahepatic in 25% of cases, and they most commonly originate from the right hepatic artery. Patients may present with hemobilia, anemia, and jaundice [2]. Furthermore, the most common causes of HAP are trauma and iatrogenic factors. Iatrogenic causes include liver transplantation, liver biopsy, laparoscopic cholecystectomy, and percutaneous transhepatic biliary drainage [3]. Herein, we present a case of HAP and associated hemobilia occurring as a rare complication of biliary pigtail stent placement.

CASE PRESENTATION

A 59-year-old man with no known chronic illnesses had initially undergone endoscopic retrograde cholangiopancreatography (ERCP) for distal common bile duct benign stricture 6 years ago. To address this issue, the patient underwent 12 ERCPs for dilation and stent replacement, and during the 8th ERCP, the plastic stent migrated into the common bile duct. However, this stent could not be removed in subsequent ERCP procedures. A second plastic stent was placed alongside the first stent, and the patient was followed-up with the second stent being replaced at each subsequent procedure. In the most recent ERCP procedure at another hospital, the migrated stent could not be removed again. The other stent was removed, and a pigtail stent was placed for the first time.

Ten days later, the patient presented to our emergency department with abdominal pain, and tests revealed a white blood cell (WBC) count of $18.2 \times 10^3/\mu L$ (N: 3.7–$10.1 \times 10^3/\mu L$), hemoglobin (Hgb) of 10.6 g/dL (N: 13–16g/dL), hematocrit (Hct) of 34.4% (N :39%–49%), aspartate aminotransferase (AST) of 54 U/L (N: <37U/L), alanine aminotransferase (ALT) of 31 IU/L (N: <41 IU/L), gamma-glutamyl transferase (GGT) of 230 U/L (N: <60U/L), total bilirubin (T.BIL) of 2.4 mg/dL (N: <1.2mg/dL), direct bilirubin (D.BIL) of 2.3 mg/dL (N: <0.3mg/dL), amylase of 359 U/L (N: <100U/L), lipase of 843 U/L (N: <60U/L), and C-reactive protein (CRP) of 27 mg/L (N: 0–5mg/L). The patient was admitted to our hospital with a diagnosis of acute pancreatitis. During follow-up, after an improvement in laboratory values, there was a recurrence of elevated levels of WBC ($24.5 \times 10^3/\mu L$), CRP (133 mg/L), T.BIL (5.8 mg/dL), and D.BIL (4.7 mg/dL), prompting the planning of another ERCP due to suspected cholangitis. The pigtail stent was grasped and removed using a snare. Upon re-entering with a duodenoscope, intense bleeding from the duodenum into the stomach was observed. Bleeding was found to be due to severe hemobilia. Considering the recent history of ERCP, an 8-cm covered metal stent was placed in the common

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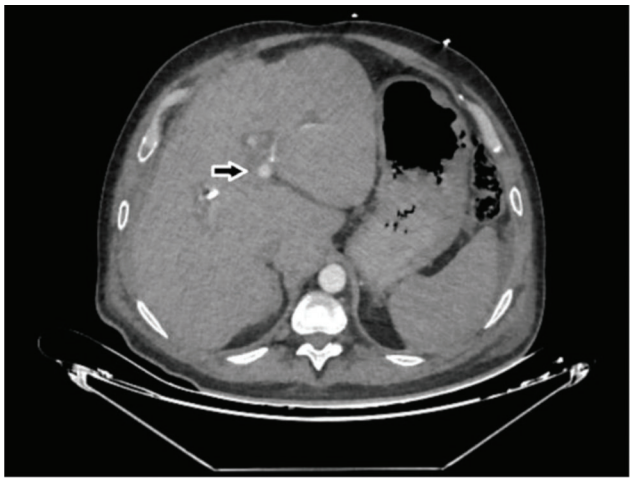

Fig. 1. CT angiography shows hepatic artery pseudoaneurysm (arrow).

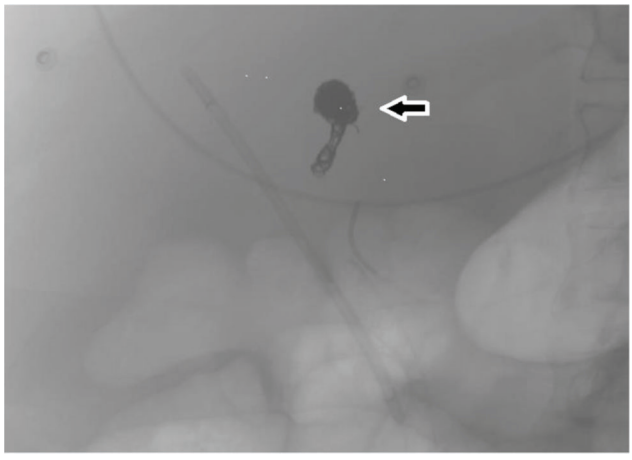

Fig. 2. Interventional radiology angiogram shows coil embolization (arrow) and the migrated stent.

bile duct. Subsequently, it was observed that the bleeding had stopped. Due to uncertainty about the diagnosis, a CT angiography was performed after the ERCP. HAP was detected on CT angiography, and the patient was urgently referred to interventional radiology (Figure 1). Interventional radiology angiograms showed pathological contrast enhancement corresponding to a 16 × 19 mm pseudoaneurysm in the midportion of the left hepatic artery, and coil embolization was performed (Figure 2). During the procedure, due to the gallbladder being filled with dense hematoma, the patient also underwent cholecystostomy. It was found that the pseudoaneurysm was located away from the covered metal stent that had been placed. Therefore, it was considered that the cessation of bleeding during ERCP occurred spontaneously and not due to the effect of the covered metal stent. The patient, who had no issues during follow-up, underwent another ERCP 14 days later, and the metal stent was removed. On fluoroscopic examination, it was observed that the stricture appearance described in previous ERCP procedures had likely resolved due to the presence of the metal stent. However, as a precautionary measure, a final plastic stent was placed. One day later, the cholecystostomy was also terminated, and the patient was discharged with plans for outpatient follow-up.

DISCUSSION

Hemobilia occurs due to a fistula forming between the biliary system and a vessel of the splanchnic circulation (portal vein or hepatic artery); it is a rare condition [4]. The most common cause of hemobilia was believed to be penetrating liver trauma, but it occurs primarily due to radiological, surgical, and endoscopic procedures [5]. Other causes of hemobilia include malignancies of the gallbladder, bile duct, pancreas, and liver; inflammatory conditions such as cholangitis, pancreatitis, cholecystitis, and hepatic abscess; renal aneurysms; gallstones; and coagulopathy [6].

The diagnosis of hemobilia is challenging. Rapid bleeding can occur into the duodenum and can obstruct the bile ducts, leading to abdominal pain, jaundice, and abnormal liver tests. Acute biliary obstruction caused by a clot can lead to pancreatitis, cholangitis, and cholecystitis [5].

HAP is rare and not commonly reported in the literature. Approximately 50% of hepatic artery aneurysms are pseudoaneurysms [7]. Such aneurysms are at high risk of spontaneous rupture. Among all visceral aneurysms, hepatic aneurysms have the highest rupture rate of 44% [8]. Presentation of HAP ranges from asymptomatic incidental findings to life-threatening bleeding. HAP can resolve spontaneously by thrombosis. However, there is a high risk of rupture associated with a high mortality rate. Therefore, early diagnosis and intervention are very crucial [9]. Selective angiography (SA) is the most sensitive test for the detection of HAP. If HAP is suspected, CT angiography is recommended first, followed by SA, if necessary [8-10].

Interventional angiography is considered the gold standard for diagnosing hemobilia and treating the underlying cause of bleeding [11]. Coil embolization is the first-line interventional method to stop bleeding and its success rate varies between 80% and 100% [6]. Surgical treatment should be considered only if embolization is insufficient [12].

The most common causes of HAP are trauma and iatrogenic factors. Iatrogenic causes include procedures such as liver transplantation, liver biopsy, laparoscopic cholecystectomy, and percutaneous transhepatic biliary drainage [13]. Additionally, HAP very rarely develops due to biliary stent placement, but can lead to hemobilia. There are very few case reports in the literature related to this [14,15]. Among these, only a few are related to pigtail stents [16,17]. The case presented in this paper is one of them.

CONCLUSION

In cases of unexplained upper gastrointestinal bleeding, hemobilia, although rare, should always be considered, and potential past iatrogenic causes should be thoroughly investigated. In cases of suspected HAP, CT angiography should be rapidly performed, and if necessary, interventional angiography should be planned for treatment.

Conflict of Interest

The author declares that he has no conflict of interest.

Informed Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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