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Stent-graft placement for delayed extrahepatic portal hemorrhage after surgical treatment for perihilar cholangiocarcinoma: A case report

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ABSTRACT

INTRODUCTION: Potential curative therapy for perihilar cholangiocarcinoma requires extensive surgical treatment, which can still be associated with significant morbidity and mortality. Postoperative hemorrhage from the portal vein is a rare but life-threatening complication. We herein report postoperative hemorrhage from an extrahepatic portal vein pseudoaneurysm successfully treated by stent graft placement late after surgical treatment for perihilar cholangiocarcinoma.

PRESENTATION OF CASE: An 83-year-old man was referred to our hospital with a chief complaint of jaundice. Based on radiological findings, we diagnosed the patient with hilar cholangiocarcinoma. After endoscopic retrograde biliary drainage, resection of the extrahepatic bile duct combined with extended left hemi-hepatectomy, including the caudate lobe, lymphadenectomy of the hepatoduodenal ligament, partial resection and reconstruction of the portal vein, and right hepaticojejunostomy was performed. Fourteen days postoperatively, bleeding through the abdominal drain around the portal vein was observed. Twenty days postoperatively, abdominal computed tomography revealed a portal vein pseudoaneurysm that had formed at the portion of reconstruction. Therefore, 24 days postoperatively, a stent graft placement of the pseudoaneurysm through the ileocolic vein was performed. Subsequently, the portal vein hemorrhage ceased.

DISCUSSION: Our present postoperative extrahepatic portal vein hemorrhage case was caused by an extrahepatic portal vein pseudoaneurysm that had formed at the reconstructed portion by erosion due to the chemical effect of the leaking bile and mechanical irritation of the surgical drain adjacent to the portal vein.

CONCLUSION: Stent-graft placement is a minimally-invasive, safe, and effective treatment option for hemorrhage from postoperative portal vein pseudoaneurysm.

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1. Introduction

Delayed postoperative hemorrhage after hepato-biliary-pancreatic surgery is a rare but life-threatening complication. Potential curative therapy for perihilar cholangiocarcinoma requires extensive surgical treatment, which can be associated with significant morbidity and mortality. We herein report the first case, to our knowledge, of postoperative hemorrhage from extrahepatic portal vein pseudoaneurysm successfully treated by stent graft placement late after surgical treatment for perihilar cholangiocarcinoma. This work has been reported in line with the SCARE 2018 criteria [1].

2. Presentation of case

An 83-year-old man was referred to our hospital with a chief complaint of jaundice. He had no relevant medical history. Abdominal contrast-enhanced computed tomography (CT) revealed a tumor at the hepatic hilum that involved the left main portal branch and associated with the dilation of the intrahepatic bile ducts (Fig. 1). Endoscopic retrograde biliary drainage was performed, and cytology of the bile revealed adenocarcinoma was examined. Based on the radiological findings, we diagnosed the patient with hilar cholangiocarcinoma. After the serum levels of bilirubin and other liver function tests returned to normal range, resection of the extrahepatic bile duct, combined with extended left hemi-hepatectomy, including the caudate lobe, lymphadenectomy of the hepatoduodenal ligament, partial resection and reconstruction of the portal vein, and right hepaticojejunostomy, followed by Roux-en-Y reconstruction, with an end-to-side jejunojunctionostomy in a retrocolic fashion was performed. A trans-anastomotic internal-external bil-

Abbreviation: CT, Computed tomography.

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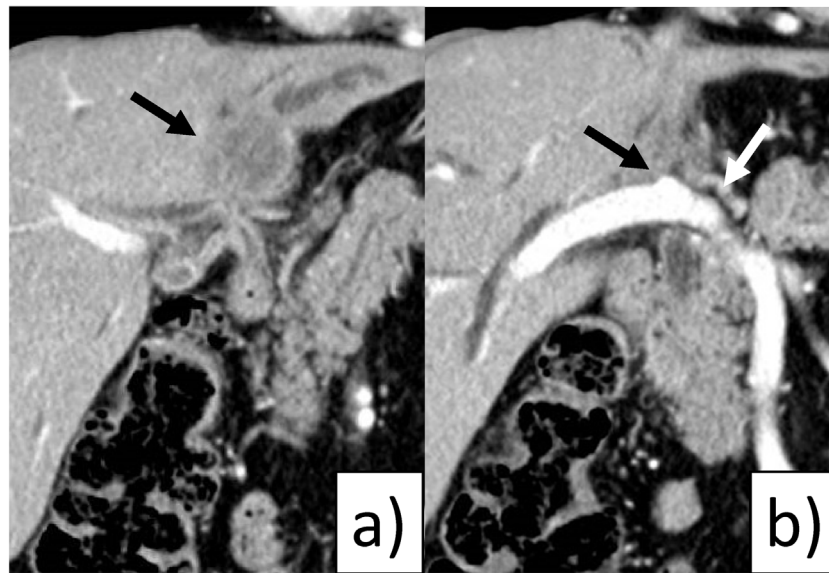


Fig. 1. Coronal abdominal contrast-enhanced computed tomography showing a low-density mass at the hepatic hilum (a), involving the left main portal branch (b, arrows), with dilatation of intrahepatic bile ducts.

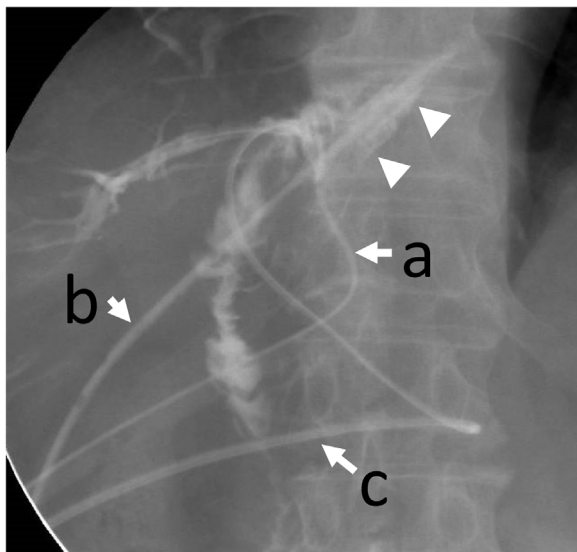


Fig. 2. Postoperative cholangiography through a trans-anatomic biliary drainage catheter (a), showing leakage of contrast medium from the hepaticojejunostomy (arrowheads) around the perioperatively placed drain behind the portal vein (b). (c): perioperative placed drain around the transected plane of the liver.

iary drainage catheter was placed (Fig. 2a), with the distal end in the bile duct and the proximal end pierced through the jejunal wall of the hepatobiliary limb of the Roux-en-Y reconstruction, leading through the abdominal wall. One abdominal drain was placed behind the portal vein (Fig. 2b), and another was placed around the transected plane of the liver (Fig. 2c). Histopathological diagnosis indicated perihilar cholangiocarcinoma with lymph node metastases invading the left portal vein and left hepatic artery, and the cut end of the right hepatic duct was microscopically positive for cancer.

Eight days postoperatively, bile leakage from the hepaticojejunostomy was observed (Fig. 2). A conservative therapy was initiated with prolonged utilization of intraoperatively placed drains. Fourteen days postoperatively, bleeding through the abdominal drain around the portal vein was observed. The drains were clamped temporarily for one day, after which bleeding

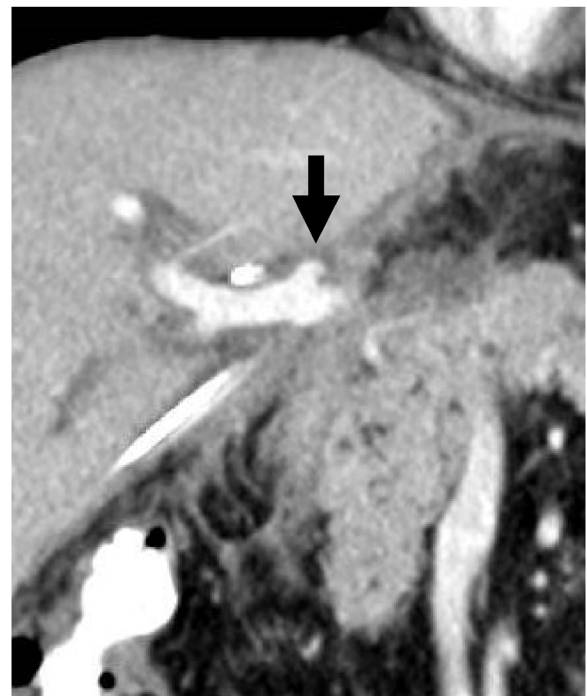


Fig. 3. Coronal abdominal contrast-enhanced computed tomography, showing portal pseudoaneurysm at the reconstructed portion (arrow).

through the drains ceased. Because the tip of the drain around the transected plane of the liver connected to the portal vein, it was removed. The drain behind the portal vein was kept indwelling and open for drainage of the bile leakage. However, bleeding through the drain behind the portal vein was observed three days later, thus, the drain was temporarily clamped again. Twenty days postoperatively, abdominal CT revealed a portal vein pseudoaneurysm at the portion of the anastomosis (Fig. 3). Therefore, 24 days postoperatively, a stent graft (GORE VIABHN endoprosthesis, W.L. Gore & Associates, Inc.) of 10 mm in diameter and 5 cm in length was inserted at the portion of the pseudoaneurysm through the ileocolic vein via mini-laparotomy under spinal anesthesia (Figs. 4, 5).

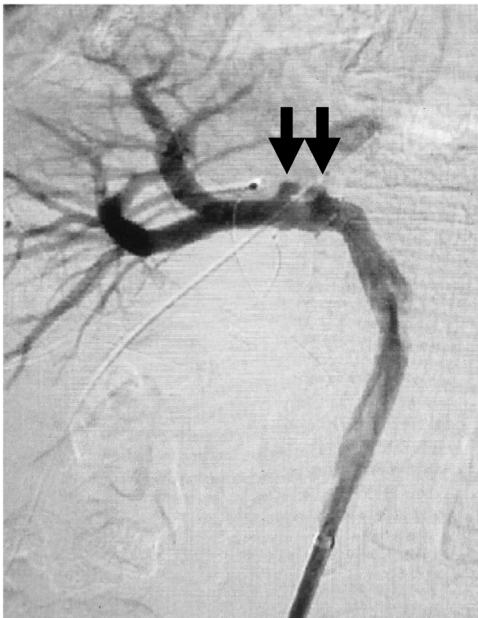


Fig. 4. Portography before stent placement, showing portal vein pseudoaneurysm (arrows).



Fig. 5. Portography after stent placement, showing the elimination of the portal vein pseudoaneurysm.

Subsequently, the portal vein hemorrhaging ceased. The bile leakage from the hepaticojejunostomy was treated with a drain that was removed 14 days after portal vein stenting. The patient died of local recurrence 5 months postoperatively; however, the patency of the stent-graft was maintained with a good portal flow.

3. Discussion

Perihilar cholangiocarcinoma is relatively rare but devastating, and it still qualifies for resection in a small subset of patients. Poten-

tial curative therapy for perihilar cholangiocarcinoma requires complete operative resection, which consists of extrahepatic bile duct resection with (extended) hemi-hepatectomy, including the caudate lobe, lymphadenectomy of the hepatoduodenal ligament, sometimes with vascular resection and reconstruction, following biliary reconstruction. These extensive surgical procedures pose a major risk of substantial postoperative morbidity and mortality [2–4]. The main postoperative complications are bile leakage, hemorrhage, septic events, and eventually liver failure, which is the most life-threatening event.

Bile leakage after surgery for perihilar cholangiocarcinoma can occur from liver resection surface or hepaticojejunostomy insufficiency, and sometimes the two are not easily distinguishable. While bile leakage from the transection surface of the liver can sometimes heal spontaneously without radiological intervention, biliary leakage can be managed with perioperatively or newly placed subhepatic drains as the first step of percutaneous drainage of the biloma. Even bile leakage due to the disruption of biliary-enteric anastomosis can be managed by interventional radiology; therefore, early relaparotomy has seldom been performed in this situation [5,6].

Postoperative hemorrhage after surgery for perihilar cholangiocarcinoma occurs at a rate of 4–8% [7,8], and postoperative bile leakage has the highest risk of postoperative hemorrhage [6]. Although the origin of the hemorrhage has not been reported in detail, conservative treatment is performed by blood transfusion or radiological interventional treatment (such as embolization) or relaparotomy to control the bleeding, which depends on the bleeding situation. Pseudoaneurysm of the hepatic artery causes life-threatening intra-abdominal bleeding, which can be controlled by embolization or stent-graft placement [9]. Postoperative extrahepatic portal vein hemorrhage is quite rare, and our present case is the first report, to our knowledge, to be successfully treated with stent-graft deployment. In the current case, delayed postoperative hemorrhage was caused by extrahepatic portal vein pseudoaneurysm that was formed at the reconstructed portion by erosion due to the chemical effect of the leaking bile and mechanical irritation of the surgical drainage adjacent to the portal vein. Bleeding was observed directly and only through the drain formed by a fistula to the portal vein pseudoaneurysm; therefore, clamping the drains resulted in transient hemostasis without intra-abdominal bleeding. The drain was necessary to treat the bile leakage, which caused a relapse of the pseudoaneurysm hemorrhage, thus, direct repair was necessary. While the standard treatment for extrahepatic portal vein hemorrhage is surgical repair [10,11], in our case, it would have been difficult to dissect severe postoperative intra-abdominal adhesions controlling intraoperative bleeding and repair the portal vein of the brittle wall due to infection and chemical action of the bile. Additionally, the postoperative course is not always good [10]. Stent-graft placement for pseudoaneurysm hemorrhaging of the splanchnic artery has been widely accepted and recognized as the first choice of treatment [9]. However, stent-grafting for hemorrhage from the extrahepatic portal vein has been reported in only three cases [11–13], two of which were for postoperative hemorrhage after pancreaticoduodenectomy and the other was for a portal vein tear due to acute necrotizing pancreatitis. In all four cases, including our case, stent-graft treatment controlled the portal vein hemorrhage without treatment-related complications.

Portal vein stenting has been widely used for portal vein stenosis due to malignant tumors [14] after either liver transplantation [15] or surgery for hepatobiliary pancreatic malignancies [16]. The used stents are bare, i.e., noncovered with relatively good patency. For portal hemorrhage, stent-grafts, i.e., covered metallic stent, are used for sealing the suspected bleeding site. In our case, we used the GORE VIABHN endoprosthesis, which is a self-expandable metallic stent covered with heparin-bonded ePTFE. We successfully

achieved hemostasis of the portal hemorrhage, and portal flow was maintained during the patient's life, for five months.

Portal stent graft can be placed either via the percutaneous transhepatic route, trans-ileocolic vein route under mini-laparotomy, or trans-jugular intrahepatic portosystemic shunt route. In the present case, a 12-Fr vascular sheath was needed for the stent-graft device; therefore, we selected the trans-ileocolic vein route to avoid the risks of complications around the access.

4. Conclusion

Stent-graft placement is a viable treatment option for minimally invasive, safe, and effective treatment of hemorrhage from postoperative portal vein pseudoaneurysm.

Declaration of Competing Interest

The authors report no declarations of interest.

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This report was not supported by any funding.

Ethical approval

Our institutional review board accepted this case report for publication.

Consent

Written informed consent was obtained from the patient for the operation and the therapeutic procedures and for publication of this case report and any accompanying images before treatment. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Authors contribution

The report concept and design were conceived by T. Tsukamoto, C. Nobori, T. Nishiyama, T. Kunimoto, R. Kaizaki and T. Inoue. T. Tsukamoto, C. Nobori, T. Nishiyama, T. Kunimoto, R. Kaizaki, T. Inoue, and Y. Nishiguchi were responsible for the interpretation of the report. Drafting of the manuscript was completed by T. Tsukamoto, C. Nobori, and T. Nishiyama.

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