

# World Journal of *Clinical Cases*

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**OPINION REVIEW**

- 1754 Gut-brain axis: Focus on gut metabolites short-chain fatty acids  
*Guo C, Huo YJ, Li Y, Han Y, Zhou D*

**MINIREVIEWS**

- 1764 Association between direct-acting antiviral agents in hepatitis C virus treatment and hepatocellular carcinoma occurrence and recurrence: The endless debate  
*Kamal A, Elsheaita A, Abdelnabi M*

**ORIGINAL ARTICLE****Retrospective Cohort Study**

- 1775 Effects of bilirubin on perioperative myocardial infarction and its long-term prognosis in patients undergoing percutaneous coronary intervention  
*Li Y, Li DB, Zhao LD, Lv QB, Wang Y, Ren YF, Zhang WB*
- 1787 Disease exacerbation is common in inflammatory bowel disease patients treated with immune checkpoint inhibitors for malignancy  
*Rubin SJS, Balabanis T, Gubatan J, Habtezion A*
- 1795 Multidrug-resistant organisms in intensive care units and logistic analysis of risk factors  
*Han Y, Zhang J, Zhang HZ, Zhang XY, Wang YM*

**Retrospective Study**

- 1806 Change and impact of left ventricular global longitudinal strain during transcatheter aortic valve implantation  
*Zhang H, Xie JJ, Li RJ, Wang YL, Niu BR, Song L, Li J, Yang Y*

**Observational Study**

- 1815 Early detection of noise-induced hearing loss  
*Meng ZL, Chen F, Zhao F, Gu HL, Zheng Y*
- 1826 Empathetic nursing with mindful cognitive therapy for fatigue, depression, and negative emotions in leukemia patients undergoing long-term chemotherapy  
*Lu YY, Lu XM, Shao CY, Wang CC, Xu TT, Zhang BL*

**Prospective Study**

- 1834 Superior pancreatic lymphadenectomy with portal vein priority *via* posterior common hepatic artery approach in laparoscopic radical gastrectomy  
*Zhang YJ, Xiang RC, Li J, Liu Y, Xie SM, An L, Li HL, Mai G*

**Randomized Controlled Trial**

- 1843 Systematic nursing interventions in gastric cancer: A randomized controlled study  
*He F, He RX*

**META-ANALYSIS**

- 1852 Impact of adding opioids to paravertebral blocks in breast cancer surgery patients: A systematic review and meta-analysis  
*Chen MH, Chen Z, Zhao D*

**CASE REPORT**

- 1863 Multiple different remote epidural hematomas after craniotomy: A case report  
*He Q, Tao CY, Fu RH, You C*
- 1869 Tuberculous pericarditis-a silent and challenging disease: A case report  
*Lucero OD, Bustos MM, Ariza Rodríguez DJ, Perez JC*
- 1876 Transileocolic endovascular treatment by a hybrid approach for severe acute portal vein thrombosis with bowel necrosis: Two case reports  
*Shirai S, Ueda T, Sugihara F, Yasui D, Saito H, Furuki H, Kim S, Yoshida H, Yokobori S, Hayashi H, Kumita SI*
- 1883 Efficacy of EGFR-TKI sequential therapy in patients with EGFR exon 19 insertion-positive non-small-cell lung cancer: A case report  
*Shan BB, Li Y, Zhao C, An XQ, Zhang QM*
- 1889 Novel compound heterozygous variants in the TAF6 gene in a patient with Alazami-Yuan syndrome: A case report  
*Lin SZ, Feng JH, Sun LP, Ma HW, Wang WQ, Li JY*
- 1896 Asymmetric limb weakness in Guillain-Barré syndrome: Three case reports  
*Hu M, Li X, Wong HY, Feng XG, Wang YZ, Zhang GR*
- 1903 Modified treatment of knee osteoarthritis complicated with femoral varus deformity: A case report  
*Xu SM, Li W, Zhang DB, Bi HY, Gu GS*
- 1909 Novel HNF1A gene mutation in maturity-onset diabetes of the young: A case report  
*Xu Q, Kan CX, Hou NN, Sun XD*
- 1914 Cerebral corridor creator for resection of trigone ventricular tumors: Two case reports  
*Liu XW, Lu WR, Zhang TY, Hou XS, Fa ZQ, Zhang SZ*
- 1922 Left abdominal wall proliferative myositis resection and patch repair: A case report  
*Xing RW, Nie HQ, Zhou XF, Zhang FF, Mou YH*
- 1929 Concurrent ankylosing spondylitis and myelodysplastic syndrome: A case report  
*Xu GH, Lin J, Chen WQ*

- 1937** Life-threatening subclavian artery bleeding following percutaneous coronary intervention with stent implantation: A case report and review of literature  
*Shi F, Zhang Y, Sun LX, Long S*
- 1946** Cryptogenic organizing pneumonia associated with pregnancy: A case report  
*Lee YJ, Kim YS*
- 1952** Eosinophilia complicated with venous thromboembolism: A case report  
*Su WQ, Fu YZ, Liu SY, Cao MJ, Xue YB, Suo FF, Liu WC*
- 1961** Neck and mediastinal hematoma caused by a foreign body in the esophagus with diagnostic difficulties: A case report  
*Wang LP, Zhou ZY, Huang XP, Bai YJ, Shi HX, Sheng D*
- 1966** Therapeutic endoscopy of a Dieulafoy lesion in a 10-year-old girl: A case report  
*Chen Y, Sun M, Teng X*
- 1973** Cavernous hemangioma of an intrapancreatic accessory spleen mimicking a pancreatic tumor: A case report  
*Huang JY, Yang R, Li JW, Lu Q, Luo Y*
- 1981** Surgery and antibiotics for the treatment of lupus nephritis with cerebral abscesses: A case report  
*Hu QD, Liao LS, Zhang Y, Zhang Q, Liu J*
- 1991** Median arcuate ligamentum syndrome: Four case reports  
*Kim JE, Rhee PL*
- 1998** Novel ABCB4 mutations in an infertile female with progressive familial intrahepatic cholestasis type 3: A case report  
*Liu TF, He JJ, Wang L, Zhang LY*
- 2007** Primary duodenal dedifferentiated liposarcoma: A case report and literature review  
*Kim NI, Lee JS, Choi C, Nam JH, Choi YD, Kim HJ, Kim SS*
- 2015** Implant site development using titanium plate and platelet-rich fibrin for congenitally missed maxillary lateral incisors: A case report  
*Zhang TS, Mudalal M, Ren SC, Zhou YM*
- 2023** Successful embolization of an intrahepatic portosystemic shunt using balloon-occluded retrograde transvenous obliteration: A case report  
*Saito H, Murata S, Sugihara F, Ueda T, Yasui D, Miki I, Hayashi H, Kumita SI*
- 2030** Bilateral pneumothorax and pneumomediastinum during colonoscopy in a patient with intestinal Behcet's disease: A case report  
*Mu T, Feng H*
- 2036** Acute kidney injury due to intravenous detergent poisoning: A case report  
*Park S, Ryu HS, Lee JK, Park SS, Kwon SJ, Hwang WM, Yun SR, Park MH, Park Y*

**2045** Vaginal enterocele after cystectomy: A case report

*Liu SH, Zhang YH, Niu HT, Tian DX, Qin F, Jiao W*

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## Transileocolic endovascular treatment by a hybrid approach for severe acute portal vein thrombosis with bowel necrosis: Two case reports

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### Abstract

#### BACKGROUND

Acute portal vein thrombosis (PVT) with bowel necrosis is a fatal condition with a 50%-75% mortality rate. This report describes the successful endovascular treatment (EVT) of two patients with severe PVT.

#### CASE SUMMARY

The first patient was a 22-year-old man who presented with abdominal pain lasting 3 d. The second patient was a 48-year-old man who presented with acute abdominal pain. Following contrast-enhanced computed tomography, both patients were diagnosed with massive PVT extending to the splenic and superior mesenteric veins. Hybrid treatment (simultaneous necrotic bowel resection and EVT) was performed in a hybrid operating room (OR). EVTs, including aspiration thrombectomy, catheter-directed thrombolysis (CDT), and continuous CDT, were performed *via* the ileocolic vein under laparotomy. The portal veins were patent 4 and 6 mo posttreatment in the 22-year-old and 48-year-old patients, respectively.

## CONCLUSION

Hybrid necrotic bowel resection and transileocolic EVT performed in a hybrid OR is effective and safe.

**Key Words:** Bowel necrosis; Endovascular treatment; Hybrid operating room; Hybrid treatment; Portal vein thrombosis; Transileocolic approach; Case report

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**Core Tip:** Acute portal vein thrombosis (PVT) with bowel necrosis is a fatal condition with no definite cure. This report describes successful endovascular treatment (EVT) in two severe PVT cases. Hybrid treatment (simultaneous necrotic bowel resection and EVT) was performed in a hybrid operating room. EVTs, including aspiration thrombectomy, catheter-directed thrombolysis (CDT), and continuous CDT, were performed *via* the ileocolic vein under laparotomy. By performing this procedure, we were able to achieve a good result for a disease with a low survival rate and did so with minimal intestinal resection.

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## INTRODUCTION

Acute portal vein thrombosis (PVT) is a rare condition involving thrombus formation in the portal vein (PV) and its branches. Extension of the thrombus to the splenic vein (SV) and superior mesenteric vein (SMV) may lead to bowel necrosis, which has a reported mortality rate of 50%-75% [1-3]. Although systemic anticoagulation is the standard treatment for PVT, most thrombi (both intra- and extrahepatic) are not sufficiently dissolved using this therapeutic approach [4,5]. Recently, in addition to systemic anticoagulation, endovascular treatments (EVTs), including aspiration thrombectomy (AT) and catheter-directed thrombolysis (CDT), have been reported to be useful for PVT [6]. However, they have yet to be established as recommended treatment strategies for this condition. Additionally, in previous reports of EVT used for PVT, transjugular, transhepatic, or transsplenic approaches to the PV are relatively common, whereas the transileocolic approach under laparotomy is rarely reported [7]. Hybrid treatment [performed in a hybrid operating room (OR)], which consists of simultaneous surgical necrotic bowel resection and thrombus removal by transileocolic EVT, is an ideal treatment for PVT with bowel necrosis. Herein, we report the cases of two patients with PVT extending to the SV and SMV complicated with bowel necrosis. The patients were successfully treated using transileocolic EVTs, including AT and CDT, performed in a hybrid OR.

## CASE PRESENTATION

### Chief complaints

**Case 1:** A 22-year-old man presented with abdominal pain lasting 3 d.

**Case 2:** A 48-year-old man presented with acute abdominal pain lasting 1 wk.

### History of present illness

**Case 1:** This man had abdominal pain and visited another hospital but was diagnosed with gastroenteritis and returned home. After that, his symptoms worsened, and he was taken to the emergency room again. He was diagnosed with gastroenteritis by computed tomography and was hospitalized. Melena was observed at night on the same day, and intestinal ischemia was suspected based on contrast-enhanced computed tomography (CECT).

**Case 2:** This man was admitted to another hospital because of epigastric pain. He was diagnosed with reflux esophagitis and was prescribed an analgesic. He returned home and was followed up. Afterwards, the patient's abdominal pain and nausea worsened, and he again went to his local hospital. He was referred to our hospital on suspicion of acute peritonitis.

### **History of past illness**

**Case 1:** The patient had a history of acute pancreatitis.

**Case 2:** There was no significant past illness.

### **Personal and family history**

There was no significant personal or family history in two patients.

### **Physical examination**

**Case 1:** The abdomen had diffuse tenderness, with rebound tenderness in the entire area.

**Case 2:** The abdomen had diffuse distention and tenderness, with rebound tenderness in the entire area.

### **Laboratory examinations**

**Case 1:** Blood test results were as follows: white blood cell count,  $17.7 \times 10^3/\mu\text{L}$ ; neutrophil count, 86.0%; C-reactive protein (CRP), 9.03 mg/dL; aspartate aminotransferase, 499 U/L; alanine aminotransferase, 575 U/L; creatinine phosphokinase, 2888 U/L; lactate dehydrogenase, 1025 U/L; and lactate, 19 mg/dL.

**Case 2:** Blood test results were as follows: white blood cell,  $18.5 \times 10^3/\mu\text{L}$  and CRP, 20.56 mg/dL. Liver function and lactate analyses revealed no abnormal changes.

### **Imaging examinations**

**Case 1:** CECT showed massive PVT extending to the SV and SMV with non-enhancement of the bowel wall. The intestinal wall was edematous and thickened, accompanied by bloody ascites.

**Case 2:** CECT showed complete PVT extending to the SMV and SV without contrast medium enhancement in the jejunum walls. The wall of the small intestine was edematous and thickened, and the contrast effect was also diminished. The intestinal wall was edematous and thickened, accompanied by a large amount of ascites.

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## **FINAL DIAGNOSIS**

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### **Case 1**

The patient's condition was diagnosed as acute PVT extending to the splenic and SMVs with bowel necrosis.

### **Case 2**

The patient's condition was diagnosed as severe PVT extending to the splenic and SMVs with bowel necrosis.

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## **TREATMENT**

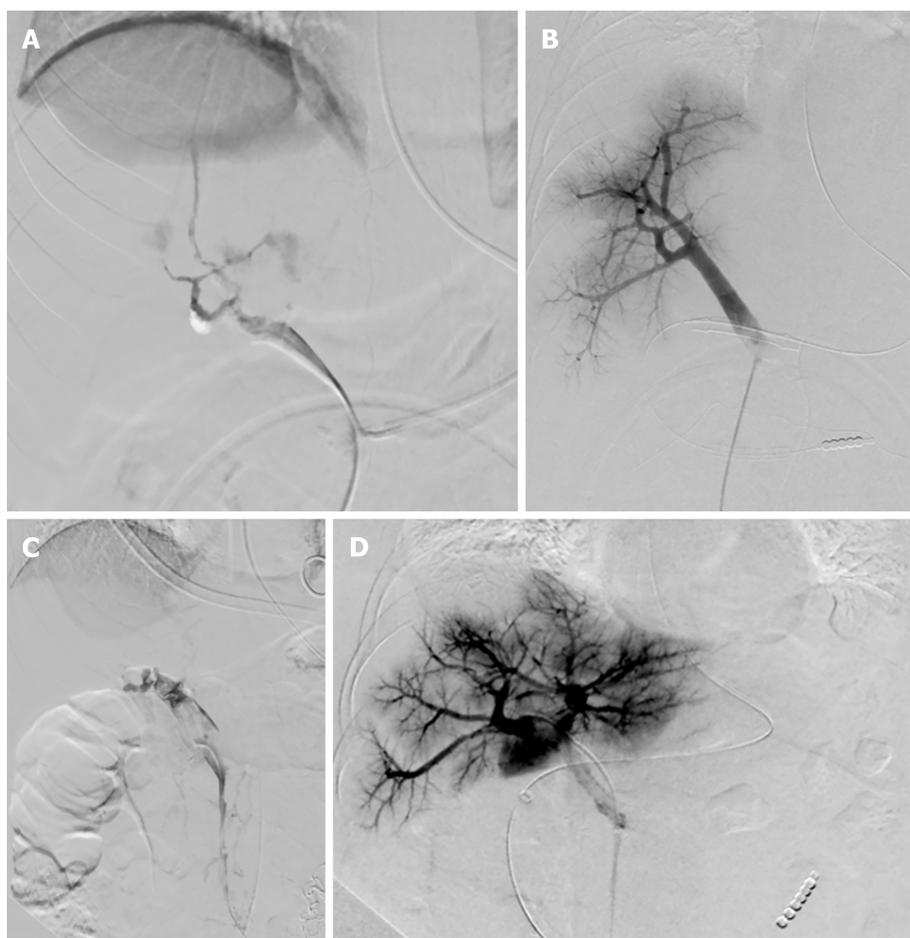
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### **Case 1**

A decision was made to perform an emergency hybrid treatment including simultaneous surgical necrotic bowel resection and EVT in a hybrid OR. Under general anesthesia, approximately 340 cm of necrotic bowel, which appeared as irreversible bowel ischemia at the time of laparotomy, was resected from the region between the distal ileum and the proximal jejunum. EVT *via* the ileocolic vein was performed immediately after the surgery (Figure 1A) over three sessions within a period of 4 d. The blood flow from the main PV to the right PV was completely recovered by the end of the final EVT (Figure 1B).

### **Case 2**

Hybrid treatment was performed in a hybrid OR with the patient under general anesthesia over a total of four sessions of EVT. In the first session, considering that bowel ischemia could be improved by EVT, open abdomen management without resection was adopted. However, the next day, there was no improvement in the ischemic bowel, indicating that the change was irreversible. In the subsequent session, approximately 210 cm of necrotic bowel was resected in total, and the thrombus was removed using the same method as that used in case 1 (Figure 1C). A total of four EVT sessions took place over a period of 5 d. The final angiogram showed good PV flow at the end of the fourth EVT session (Figure 1D).



**Figure 1 Endovascular treatment.** A and B: Endovascular treatment in a 22-year-old man with portal vein thrombosis. Pretreatment angiogram *via* an ileocolic vein showing a massive thrombus in the entire portal vein (A); Posttreatment angiogram showing the recovery of blood flow from the main portal vein into the right portal vein (B). C and D: Endovascular treatment in a 48-year-old man with portal vein thrombosis. Pretreatment angiogram *via* an ileocolic vein showing complete occlusion of the portal vein (C); Posttreatment angiogram showing the disappearance of the portal vein thrombus (D).

### **EVT procedure**

During the laparotomy, the ileocolic vein was divided, isolated, and punctured with an 18-G needle (Catheter Introducer; Medikit, Tokyo, Japan). After ligation of the proximal end of the ileocolic vein, a 0.035-in guidewire (Radifocus Guide Wire M; Terumo, Tokyo, Japan) was inserted, and an 8-Fr sheath (Supersheath; Medikit) was inserted. At this point, heparin (5000 U) was administered. A further 1000 U was then administered every hour to keep the activated clotting time within the range of 200-300 s. Next, the 0.035-in guidewire and a 4-Fr guide catheter (GLIDECATH; Terumo) were advanced into the PV, SMV, and SV *via* the sheath. The 4-Fr guide catheter was then replaced with a 6-Fr catheter (Aspiraircass; Medikit) and an 8-Fr guiding catheter (Launcher; Medtronic, Dublin, Ireland) for the patient in case 1 and the patient in case 2, respectively. Manual AT using these catheters was repeatedly performed. Additionally, to remove the residual thrombus found after AT, CDT using 30000 U of urokinase (Mochida, Tokyo, Japan) for the patient in case 1 and 24000 U for the patient in case 2 was performed *via* a 5-Fr multiple-side-hole infusion catheter (Fountain Infusion System; Merit Medical, South Jordan, UT) within the same session. This was repeated almost every day for 3 d in the patient in case 1 and for 4 d in the patient in case 2 until peripheral PV circulation was completely restored. During the interval between each session, a 5-Fr sheath (Medikit) and a heparin-coated catheter (Anthon; Toray Medical, Tokyo, Japan) were kept in place within the SMV and PV. Continuous CDT using urokinase was performed *via* the sheath (120000 U/d) and catheter (240000 U/d) with an open abdomen and continuous negative pressure.

## **OUTCOME AND FOLLOW-UP**

### **Case 1**

After the treatments, an intravenous injection of systemic heparin (20000-50000 U; Mochida) was administered daily for 45 d, while the activated partial thromboplastin time was maintained at 50-60 s.

Heparin was switched to warfarin (Eisai, Tokyo, Japan) 3 mg/d on postfinal EVT Day 40. Although the patient experienced short bowel syndrome, he was discharged 4 mo after the treatment. CECT showed that the patient's main and right PVs were patent 4 mo after the treatment.

## Case 2

Heparin (15000 U) was replaced with edoxaban (Daiichi-Sankyo, Tokyo, Japan) 60 mg/d on postfinal EVT Day 24. The patient was discharged 33 d after the final EVT. CECT showed that the PV was patent 9 mo after the treatment.

## DISCUSSION

PVT is a rare condition, with an incidence of approximately 0.7 per 100000 people per year[1]. Acute PVT is even more infrequent and is difficult to diagnose. In this case report, acute PVT was diagnosed as gastroenteritis in one patient and peritonitis in another patient. Thus, infections might have induced three factors of Virchow's triad, including intravascular vessel wall damage, stasis of flow, and the presence of a hypercoagulable state, and caused PVT[4]. Focusing on risk factors, such as thrombophilia and portal hypertension, may be effective for diagnosing PVT; however, it is difficult to distinguish PVT from enteritis and other ischemic intestinal diseases, such as arterial thrombosis or nonocclusive mesenteric ischemia, based on clinical symptoms without imaging evaluation[4].

Nevertheless, early diagnosis is important because PVT can be quite severe and is associated with a high mortality rate[1-3]. In severe PVT, such as in these cases, venous congestion is initially induced owing to various etiologies. Hemorrhagic infarction then occurs, and arteriovenous obstruction finally leads to bowel necrosis. Yerdel *et al*[8] classified PVT into four grades of severity (1 to 4), with Grade 4 being the most severe stage, defined as "complete PV and entire SMV thrombosis." According to their study, the in-hospital mortality of Grade 4 PVT was 50%. In the present case report, both patients had Grade 4 PVT; nonetheless, despite the severity of their conditions and the high associated mortality rate, positive outcomes were achieved using hybrid treatment including necrotic bowel resection and multiple EVT sessions for thrombi until the intrahepatic blood flow became stable.

Although there are no reports that clearly define the treatment endpoint for PVT, we believe that it is important to improve the inflow and outflow systems of the liver. Sharma *et al*[4] revealed that different treatments were shown according to the etiologies. It is reasonable to monitor conservatively, without anticoagulation, for reversible risk factors such as pancreatitis or abdominal infections in patients with minimal thrombus in PV. In cases of noncirrhotic, nonmalignant, and symptomatic PVT, anticoagulation is recommended. For asymptomatic PVT, especially with extension to the mesenteric veins, nonreversible risk factors, and hypercoagulable states, anticoagulation is reasonable for preventing or reducing symptoms such as abdominal pain, nausea or vomiting, or ischemic complications, including bowel necrosis and portal hypertension-related symptoms. In patients with PVT accompanied by malignancy, anticoagulation is also recommended in the majority of cases. Thrombolysis can be considered in those with thrombus extension or worsening pain while on anticoagulation and in patients with impending or ongoing bowel necrosis owing to thrombosis. For these, anticoagulation is often recommended.

According to the guidelines for PVT treatment[9], systemic anticoagulation is considered the first-line therapy for this condition. However, collective data from retrospective studies show that anticoagulation alone might be insufficient. A report showed that in cases of extensive PVT in which anticoagulation was initiated early and maintained for 6 mo, 50% of patients recanalized completely, 40% did so partially, and 10% did not recanalize at all[6]. This result supports the idea that combining systemic anticoagulation with EVT might be beneficial, especially in cases of imminent bowel infarction. Furthermore, EVT may become the sole effective therapy for PVT in patients with a contraindication to anticoagulation.

Recently, several EVT techniques, including CDT and AT, have been described for acute PVT[6]. Wang *et al*[10] reported the successful treatment of 12 patients with acute PVT by aspiration combined with CDT. Eight patients achieved complete recanalization, and four patients achieved partial recanalization, which shows that AT combined with CDT was more effective than monotherapy. Kennoki *et al* [7] reported a case of successful recanalization of acute extensive PVT by AT and continuous CDT. The present case report involved EVTs, including AT, CDT, and continuous CDT. Although CDT is effective for acute thrombosis and enhances the efficacy of thrombolytics, it generally takes time and may cause clot migration and hemorrhagic complications[11]. In contrast, high recanalization rates have been achieved using AT, which has frequently allowed for the removal of large sections of thrombus all at once. This reduces the time required for recanalization, is not associated with hemorrhagic risk, and may prevent distal embolization, which is not the case in CDT[11]. Nevertheless, a large sheath is generally required to avoid vessel injury, and this cannot be applied to tortuous vessels.

In terms of the approach to the PV, the percutaneous transhepatic, percutaneous transsplenic, and transjugular approaches, including transjugular intrahepatic portosystemic shunt (TIPS), are known, along with the transileocolic approach under laparotomy. The percutaneous approach is the least

invasive. However, it may increase the risk of bleeding. In one report, out of 46 patients who underwent the percutaneous transsplenic approach, 3 (6.5%) had severe bleeding, and 6 (13%) had mild bleeding [7]. Moreover, although TIPS using covered stents is a common procedure in Europe and the United States when approaching the PV, TIPS has not been allowed by the health insurance system in Japan and is only performed in a limited number of institutions. In the present cases, there were a few reasons for the choice of the transileocolic approach. First, the percutaneous approach would have been difficult to perform because of the large amount of ascites, especially in case 1, and the complete occlusion of the intrahepatic PV. Second, considering that both patients required necrotic bowel resection, it was reasonable to perform EVT under laparotomy. Third, the transileocolic approach is associated with a low risk of bleeding compared with other approaches because the ileocolic vein is punctured under direct observation[12]. Finally, this approach enables the use of larger devices for AT. Therefore, the transileocolic approach is considered a useful method that does not cause serious complications in the short term; however, further research and evaluation of the complications and long-term prognosis of patients is required.

As shown in the present cases, surgery and EVT should be performed in a hybrid OR to optimize the treatment of PVT with bowel necrosis. Indeed, the hybrid OR enabled us to reduce the transfer time from the intervention room to the OR and, consequently, the risks related to this transfer. Moreover, these patients' abdomens were left open with negative pressure applied during the intervals between EVT sessions. The open abdomen allowed for the bowel condition to be monitored and permitted a sheath to be left with a catheter in the PV during the intervals between EVT sessions. Multiple EVT procedures in combination with an open abdomen enabled us to perform minimal bowel resection while visually observing the condition of the bowel.

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## CONCLUSION

Herein, two patients with severe acute PVT extending to the SV and SMV with bowel necrosis were successfully treated with EVTs. Overall, hybrid necrotic bowel resection and transileocolic EVT with AT and CDT performed in a hybrid OR were found to be effective and safe.

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## FOOTNOTES

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