Case report

A case of ascites and SMV thrombosis due to an intrahepatic arterio-portal fistula after hepatectomy

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Abstract

Arterio-portal fistula (APF) is a rare vascular abnormality, characterized by communication between artery and portal vein. We describe a 69-year-old male patient with APF, who developed acute clinical symptoms 10 months after hepatectomy. The APF was accompanied by massive ascites and thrombosis in the superior mesenteric vein. The condition was successfully treated using anticoagulant, diuretics, and interventional coil embolization of the fistulas. This case indicates that APF is an important postoperative complication after hepatectomy, and that intrahepatic APFs could manifest acutely with ascites and superior mesenteric vein thrombosis.

Keywords: Arterio-portal shunt, fistula, communication, ascites, colorectal

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Introduction

Arterio-portal fistula (APF) describes an abnormal communication between artery and portal vein¹⁾, which can arise due to hepatic injury^{1,2)}, ablation therapy for tumors^{3,4)}, liver biopsies⁵⁾, and surgeries⁶⁾ in adults. Although the entity is well-known, its incidence is rare. One study into radio-frequency ablation therapy as a cause found asymptomatic APFs in only 9 patients (0.38%) among 2320 patients³⁾. After hepatectomy, only a small number of cases were described in literature^{7,8)}, with the incidence remaining unknown except that it is extremely rare. Clinical features and treatments vary among individuals, ranging from asymptomatic small fistulas that spontaneously resolve to severe symptoms derived from portal hypertension and necessitating urgent treatment¹⁻⁶⁾.

Herein, we describe a patient with intrahepatic APFs that developed 10 months after hepatectomy and radio-frequency ablation therapy. The location of APFs implicated hepatectomy as the causative intervention for this

rare disease. Massive ascites developed within 4 weeks based on the most recent surveillance by computed tomography demonstrating no sign of portal hypertension. The rapid onset of symptoms in the present case and the successful treatment course make it valuable to report.

Case presentation

A 69-year-old male patient was referred to our department for the treatment of sigmoid colon cancer and hepatic metastasis. After laparoscopy-assisted sigmoidectomy with lymphadenectomy, the patient underwent six courses of FOLFOX (5-FU and oxaliplatin) plus bevacizumab treatment. The patient then underwent partial resection of the liver for four tumors (S6, S5, S5, S3) and radio-frequency ablation therapy for one tumor (S8). After the operation, the patient received a further six courses of FOLFOX treatment as postoperative adjuvant chemotherapy.

Nine months after the hepatectomy and radio-frequency ablation therapy, enhanced abdominal computed tomography (CT) revealed no sign of recurrence. One month after this last surveillance, the patient presented at our department due to a three-week history of exacerbating abdominal distention. His serum level of carcinoembryonic antigen, which was elevated prior to surgery, was

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within normal range. Dual-phase abdominal CT demonstrated massive ascites and thrombosis of the superior mesenteric vein (SMV). In addition, the portal vein was strongly enhanced at the arterial phase (Fig. 1).

The patient was diagnosed as having arterio-portal shunt and portal hypertension. The APF site suggested that hepatectomy was the causative intervention. He was admitted to hospital for anti-coagulant therapy with 1 mg of warfarin and a diuretic. Subsequent angiography of the hepatic artery confirmed an APF sited at the periphery of A6 and A4 (Fig. 2). Radiological coil embolization of the shunts was performed after shrinkage of the SMV thrombosis and occlusion of the fistulas was confirmed by the lack of flow from artery to portal vein. The ascites gradually subsided following treatment, and after hospital discharge, the patient remains well two years later without recurrence of the fistula or tumor.

Discussion

Herein, we described a case of intrahepatic APF causing symptomatic ascites and SMA thrombosis 10 months after surgical treatment. Vauthey et al¹⁾ presented 12 cases of extra/intra hepatic APF, among which 5 cases were caused by liver injury. While the development of APFs immediately after trauma has been described, one case of intrahepatic APF was diagnosed 2.5 years after hepatic injury. A review of other reports also revealed that some described APFs becoming symptomatic soon after the causative event(s)^{3, 9)}, while others suggested that APF could gradually exacerbate^{2, 6, 11, 12)}. Reportedly, APFs have been diagnosed 18-20 years after the causative event^{10, 11)}. From our experience and others, intrahepatic APF could become symptomatic late after liver injuries or invasive testing/treatment. Another uniquely valuable point of the present case is that our case suggested symptom of APFs could also develop within a few weeks.

Kimura et al., in 2018⁷⁾, reported a case of APF in a 62-year-old male patient who underwent anterior segmentectomy of the liver. They concluded that APFs after hepatectomy are rare because only one case of APFs after hepatectomy had been published before their case presentation. Indeed, our search through PubMed by using the term ‘((arterio-portal) OR (arteriportal) OR (arterial-portal)) AND ((shunt) or (fistula)) (arterio-portal) OR (arteriportal) OR (arterial-portal)) AND ((shunt) or (fistula))’ identified no additional case between 2018 and November 2019, confirming their claim. They also discussed the relationship between the surgical technique and development of APFs, whereby perihilar fully simultaneous transection of the Glissonean pedicle with transfixion suture could be a potential cause of the APFs, although they mentioned that no conclusion could be drawn due to the lack of evidence⁷⁾. Such a hypothesis is not applicable in the present case because APFs in our

patients are attributable to partial hepatectomy with ligation of peripheral ducts and vessels.

A potential factor influencing the development of symptomatic APF is the perioperative use of oxaliplatin, which is known to cause sinusoidal injury. Vauthey et al¹⁾ discussed that young patients with APFs often suffered from high-output (hyperdynamic) heart failure because there was practically no sinusoidal resistance and APFs demands more ejection fraction. In contrast, adult patients with APFs tended to suffer from variceal bleeding and ascites due to portal hypertension¹⁾. In our patient, the use of oxaliplatin might have caused sinusoidal injuries, and worsened the portal hypertension, resulting in acute and massive ascites with thrombosis. Again, the relationship between perioperative use of chemotherapy and postoperative APF formation has not been clearly demonstrated in clinical studies, with the exception of one case presentation¹³⁾, and further accumulation of data is necessary to prove the hypothesis.

Although the mainstream treatment for APF had been surgical resection of the shunt¹⁾, embolization of the responsible arteries by radiological intervention are also frequently reported. The radiological intervention is less invasive than surgical intervention and effective with fewer complications¹⁾; however, as Roux et al noted¹⁴⁾, potential complications such as ischemic change to the liver parenchyma, development of abscess, and coil migration warrant consideration of the risks of endovascular treatment. After treatment, the clinical prognosis of APFs is favorable^{1, 2, 4)}; however, the long-term effect of the APFs may have negative impacts on the regeneration of hepatic parenchyma or the parenchymal structures^{15, 16)}. Thus, we suggest that asymptomatic APFs or remnant fistulas after embolization of large/multiple APFs should be monitored.

In conclusion, the present case suggested that APF symptoms might acutely develop late after intrahepatic interventions. Thus, although rare, hepatectomy is an important potential cause of intrahepatic APFs. In addition, radiological interventional embolization seems a safe and effective treatment option for APFs.

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Fig. 1 Enhanced abdominal computed tomography (A) The portal vein (white arrowhead) is strongly enhanced at the early phase of dual-phase CT. The arrow indicates the arteries. (B) Thrombotic stenosis of superior mesenteric vein (white arrow). (C) Development of ascites is shown.

Fig. 2 Angiography of hepatic artery and CT (A) Angiography of the right hepatic artery clearly demonstrates the existence of fistula between artery and portal vein (white arrowhead). (B) Selective angiography of A6 (arrow) shows the arterio-portal fistula with backflow from the artery going to the portal vein branch. (C) Angiography of the common hepatic artery (arrowhead) shows the disappearance of flow from artery to portal vein. Multiple coils at A6 and A4 occluded the fistula (white arrows).

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