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Single Case

Portal Vein Aneurysm in a Patient with Cirrhosis Type C Controlled by Direct-Acting Antiviral Treatment

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Keywords
Portal vein aneurysm · Hepatitis C virus · Direct-acting antivirals

Abstract
Introduction: Portal vein aneurysm (PVA) is a rare saccular or fusiform portal vein dilatation. The management and optimal treatment of PVA remain unknown. ***Case Presentation:*** A 53-year-old man with hepatitis C virus (HCV) infection was diagnosed with PVA measuring 28 mm in diameter. Under observation, his liver fibrosis progressed, and the PVA diameter gradually increased to 52 mm. The patient was treated with elbasvir-grazoprevir for 12 weeks, and HCV disappeared. After achieving sustained virological response, liver fibrosis improved and the PVA progression ceased. ***Conclusion:*** HCV clearance by direct-acting antiviral treatment not only regressed liver fibrosis but may have also restrained the progression of PVA in a patient with cirrhosis type C and PVA.

Introduction

A portal vein aneurysm (PVA) is a saccular or fusiform dilatation of the portal vein that has a diameter of >19 mm and >15 mm in cirrhotic and non-cirrhotic patients, respectively. PVA has two types: congenital and acquired. The most frequent cause of acquired PVA is portal hypertension. PVA can present with several symptoms and complications, such as

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abdominal pain, thrombosis, or rupture. PVA is rare, and the number of reported PVA cases stands at about 280 all over the world, although the prevalence of imaging devices, such as magnetic resonance imaging (MRI) and computed tomography (CT), enables easy detection of PVA [1]. The management and optimal treatment of PVA have not been established because of its rarity and uncertainty of its pathogenesis.

Hepatitis C, caused by the hepatitis C virus (HCV), is one of the most common infectious diseases worldwide. Chronic inflammation induced by persistent HCV infection causes liver fibrosis, eventually resulting in liver cirrhosis. Portal vein pressure increases with liver fibrogenesis [2]. Esophagogastric varices, splenomegaly, and pancytopenia are major complications in patients with cirrhosis. As for anti-HCV treatment, type I interferon (IFN) was first approved for use in the 1990s. The sustained virological response (SVR) rate of IFN is 15–20%, and it can be accompanied by severe adverse events such as fever, fatigue, and depression. Although pegylated IFN and ribavirin therapy, and its combination therapy with protease inhibitors, which have been adopted since the 2000s, could increase SVR rates to 80–90%, IFN-related adverse events still occur [3]. Therefore, a considerable proportion of patients with chronic hepatitis C were previously unwilling to be treated with IFN-based therapy due to serious adverse events. Currently, IFN-free therapy, called direct-acting antiviral (DAA) treatment, directly inhibits viral enzymes and proteins, and has replaced IFN-based therapy because of its very high SVR rates of >90% and lack of toxicity. Both IFN-based therapy and DAA treatment have been shown to improve liver fibrosis and portal hypertension after achieving SVR [4, 5].

In the present report, a patient with type C cirrhosis was diagnosed with PVA using contrast-enhanced CT. The patient refused IFN-based therapy. PVA progressed and was accompanied by thrombosis, for which conservative thrombolytic therapy was adopted, followed by oral administration of warfarin. Thereafter, the patient decided to undergo DAA treatment. HCV disappeared at treatment week 4, and no relapse occurred. Consequently, PVA progression ceased after SVR was achieved. These findings suggest that in a PVA patient with HCV infection, DAA treatment may contribute not only to HCV clearance but also to PVA control.

Case Report

A 53-year-old man first visited our hospital because of liver dysfunction in 1992. The patient had no significant medical history other than diabetes. The patient was diagnosed with chronic hepatitis C genotype 1. His platelet count was 165,000/μL, and his albumin-bilirubin (ALBI) score was −3.07, which was classified as grade 1 [6]. He refused IFN therapy due to severe adverse events and underwent regular follow-up blood tests and abdominal ultrasound sonography (US) every 3 months. The clinical course of the patient is shown in Figure 1. His platelet count gradually decreased, and the ALBI score increased. Upper gastrointestinal endoscopy revealed the emergence of esophageal varices. In 2004, he first underwent contrast-enhanced CT to screen for hepatocellular carcinoma. The CT image did not reveal any tumor in the liver; however, there was an ovoid, homogenous, highly enhancing lesion with a diameter of 28 mm at the confluence of the splenic vein and superior mesenteric vein (Fig. 2a, b). Based on the CT image, the patient was diagnosed with PVA. At that time, abdominal US showed limited dilatation of the portal vein due to the PVA (Fig. 2c). As the patient did not present with any symptoms, no treatments were performed, and he was scheduled for a follow-up examination. In 2006 and 2007, the esophageal varices further enlarged, and combination therapies of endoscopic variceal ligation and endoscopic injection sclerotherapy were performed twice. His platelet count further decreased, and his ALBI score increased

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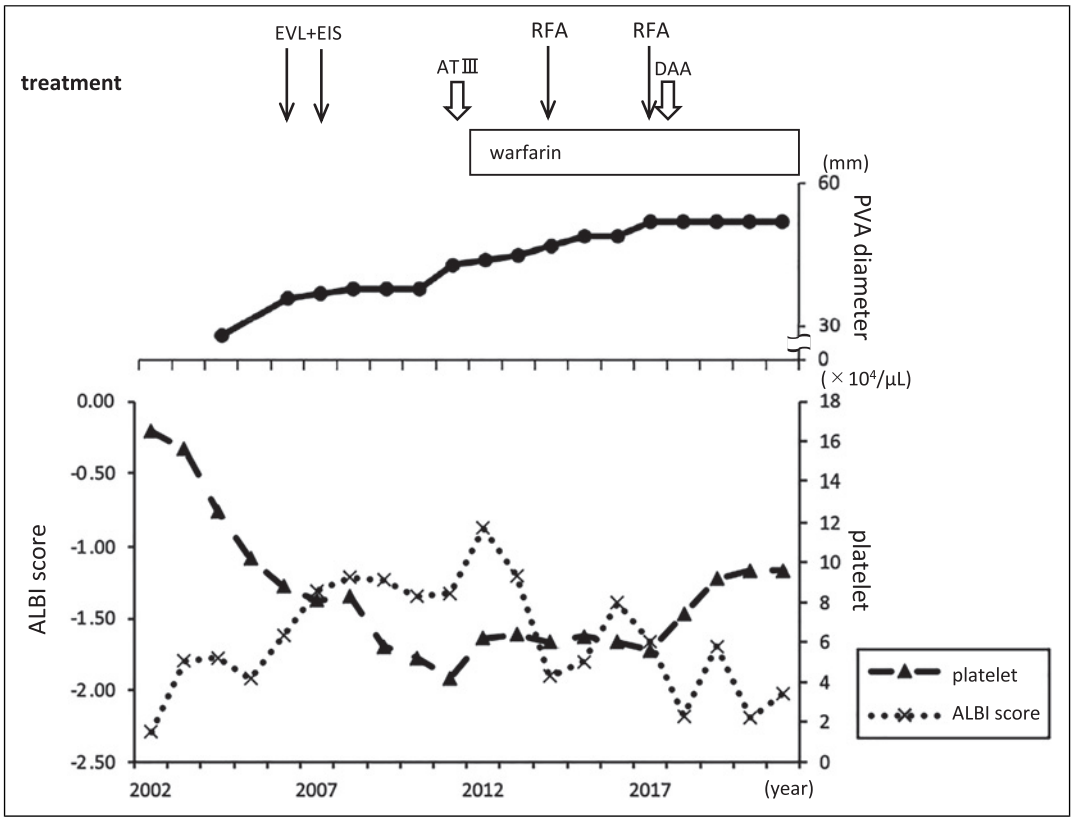

Fig. 1. Clinical course of the patient. Serial changes in the PVA diameter, platelet count, ALBI score, and medications are shown. EVL, endoscopic variceal ligation; EIS, endoscopic injection sclerotherapy; AT III, antithrombin III; RFA, radiofrequency ablation; DAA, direct-acting antiviral; PVA, portal vein aneurysm; ALBI, albumin-bilirubin.

to −1.77, which was classified as grade 2b. In 2010, contrast-enhanced CT showed the PVA with a diameter of 38 mm and an internal thrombus (Fig. 2d, e). He was treated with intravenous administration of antithrombin III (1,500 units/day) for 10 days. The internal thrombus completely disappeared; however, the diameter of the PVA increased to 43 mm 1 year later. Oral administration of warfarin was initiated to prevent recurrence of thrombosis. The diameter of the PVA continued to gradually increase. In 2014 and 2017, radiofrequency ablation was performed for hepatocellular carcinoma in segments 6 and 7 that measured 17 mm and 15 mm, respectively. In 2017, the diameter of the PVA measured 52 mm. He then decided to undergo DAA treatment. The patient was treated with elbasvir-grazoprevir for 12 weeks [7]. HCV disappeared at treatment week 4, and no adverse events were observed during the duration of elbasvir-grazoprevir therapy. The patient continued regular follow-up after DAA treatment. HCV did not relapse, and the viral response was sustained. Upper gastrointestinal endoscopy showed no recurrence of esophageal varices. The platelet count increased, and the ALBI score decreased after achieving SVR. In 2021, his platelet count was 96,000/μL, and the ALBI score was −2.64, classified as grade 1. The diameter of the PVA remained at 52 mm (Fig. 3a, b). Abdominal US showed an anechoic lesion with no recurrence of thrombus (Fig. 3c). Using the color Doppler method, the blood flow in the aneurysm was seen as bidirectional due to the swirling of blood, which is the so-called yin-yang sign (Fig. 3d), while a flat venous flow was observed inside the aneurysm using the pulsed-wave Doppler method (Fig. 3e). Thus, the PVA did not enlarge 4 years after achieving SVR following DAA

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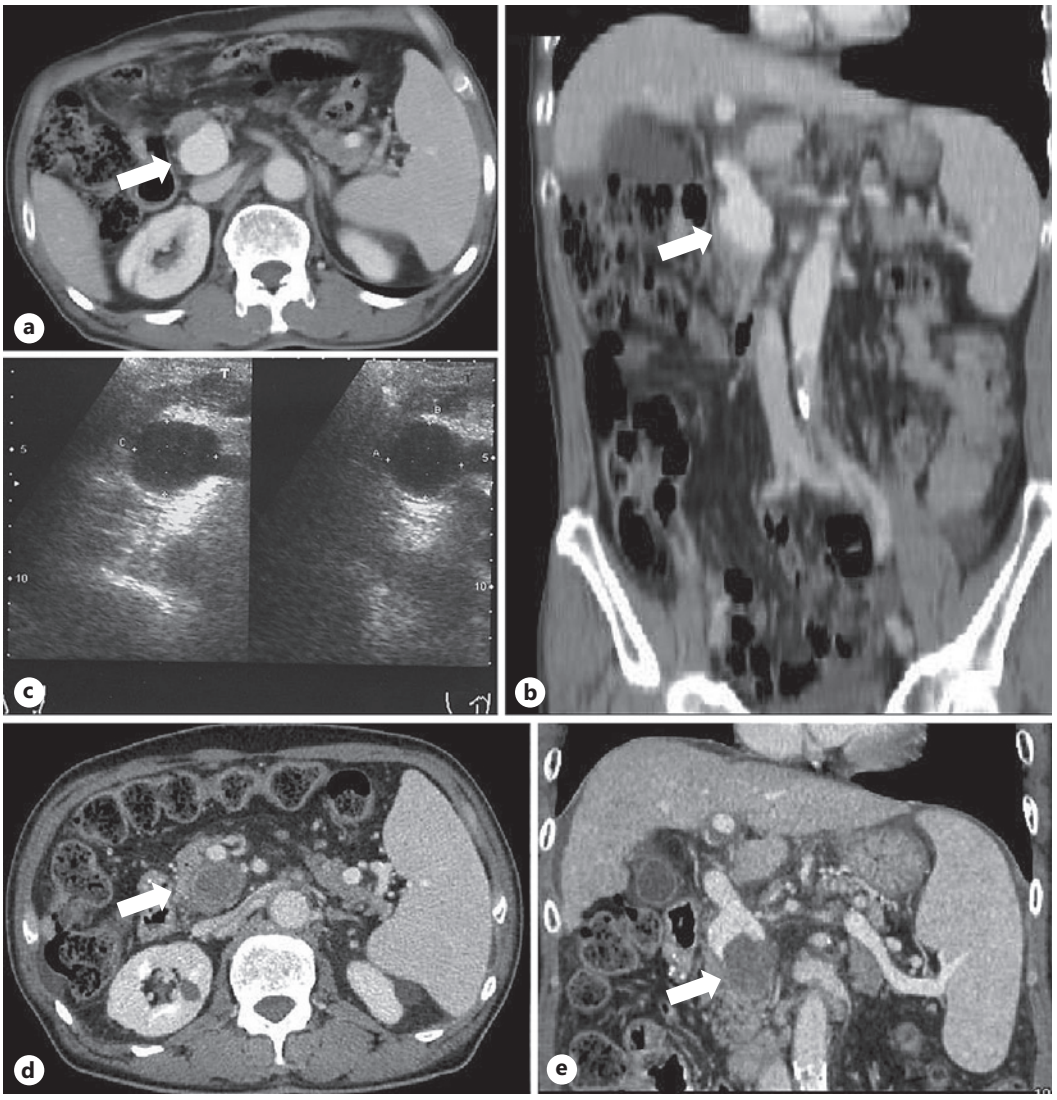

Fig. 2. Images at initial diagnosis of the PVA in 2004 (**a–c**) and thrombus in the PVA in 2010 (**d**, **e**). Axial (**a**) and coronal (**b**) plane images of contrast-enhanced CT show an enhancing ovoid lesion at the confluence of the splenic vein and superior mesenteric vein (arrows). The abdominal ultrasound image shows an anechoic dilatation of the portal vein through the short (left) and long (right) axes on subcostal view (**c**). Axial (**d**) and coronal (**e**) plane images of the contrast-enhanced CT show the enlarged PVA with internal thrombus (arrows).

treatment. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000535957).

Discussion

PVA is defined as a localized dilatation of the portal vein that has a diameter of >19 mm and >15 mm in cirrhotic and non-cirrhotic patients, respectively. The most frequent site of PVA is the confluence of the splenic vein and superior mesenteric vein, as

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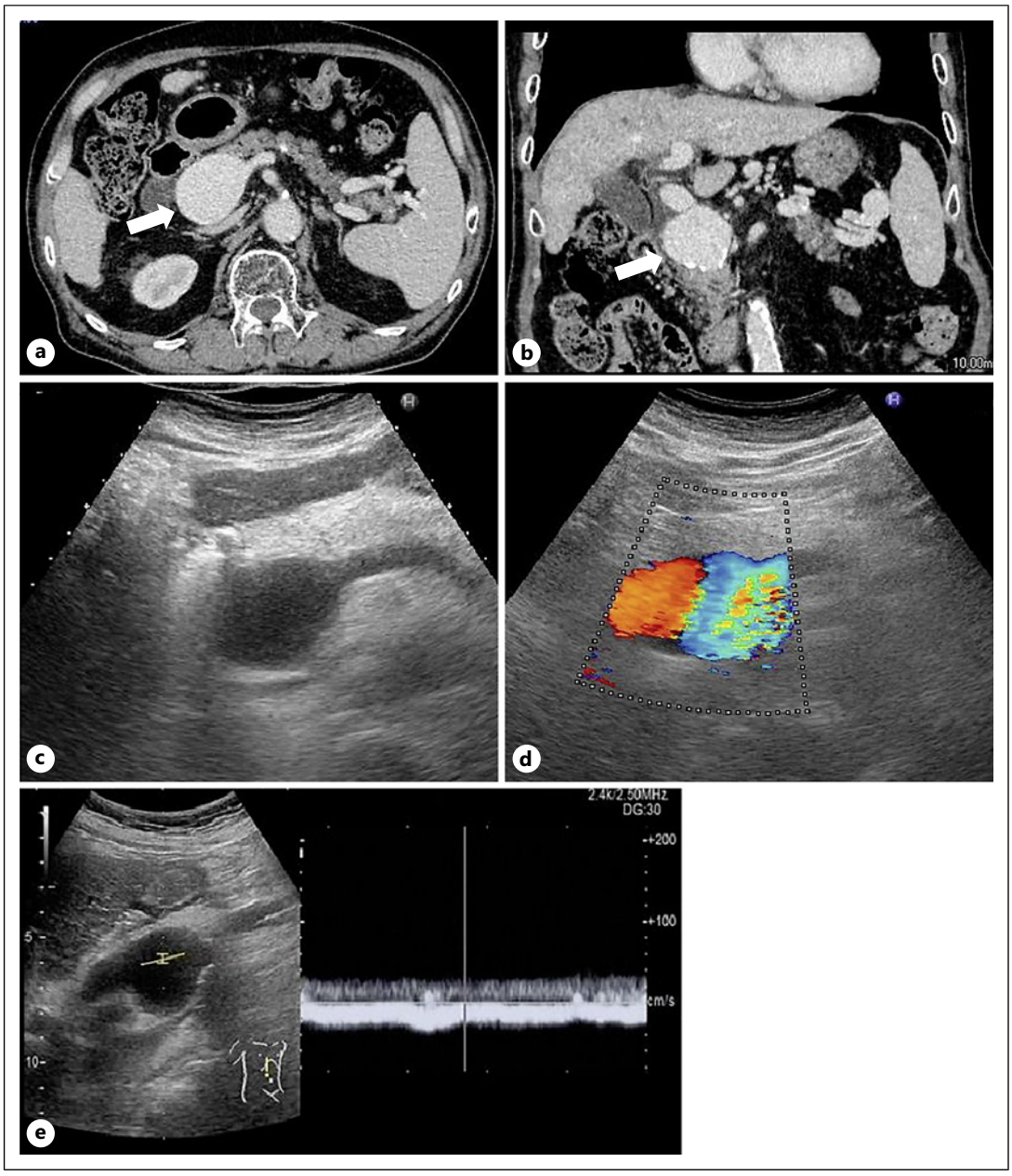

Fig. 3. Images in 2021. Axial (**a**) and coronal (**b**) plane images of contrast-enhanced CT show the diameter of the PVA is 52 mm with no thrombus (arrows). An abdominal ultrasound image shows an anechoic lesion (**c**) with color flow (**d**) and flat venous flow (**e**).

in our case. PVA is classified into congenital and acquired types. Congenital PVA occurs due to failure in the regression of the primary right vitelline vein or an inherent weakness of the vessel wall. For the acquired type, PVA can occur in chronic liver disease, pancreatitis, or trauma. The most frequent cause of acquired PVA is portal hypertension, because continuous high pressure makes the vessel wall more susceptible to dilation [1]. In the present case, PVA progressed as the platelet count decreased and the ALBI score increased, and enlarged esophageal varices were observed during surveillance. These

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findings suggest the relevance of portal hypertension due to liver fibrosis and progression of PVA. Thus, the present case is thought to be of the acquired type.

PVA is usually detected in surveillance studies using abdominal US On grayscale US, the typical finding of PVA is an anechoic mass near the portal hepatis or around the pancreas, which resembles a cyst [8]. The presence of color flow by color Doppler method or a non-pulsatile waveform indicating portal vein blood flow by pulsed-wave Doppler method in an anechoic mass suggests the existence of PVA. However, the sensitivity of abdominal US for the detection of PVA ranges from 80% to 100% [9]. Therefore, contrast-enhanced CT or MRI should be considered to detect PVA when the portal vein cannot be depicted clearly by US in a patient with liver cirrhosis.

Although the prevalence of imaging devices, such as MRI and CT, has resulted in a high diagnostic capability, PVA is still rare, and its pathogenesis remains unclear. An optimal consensus on the management of PVA has not been established. A larger PVA diameter or a tendency to progress has been considered as having clinical significance in terms of complications and mortality [1]. In the present case, portal hypertension due to liver fibrogenesis was thought to be involved in the enlargement of the PVA. Amelioration of chronic liver inflammation after HCV clearance was followed by improvement of liver fibrosis and portal hypertension [4]. In this case, the platelet count increased, and the esophageal varices improved after achieving SVR by DAA treatment, which certainly suggested improvement of liver fibrosis and portal hypertension. Progression of PVA ceased after the achievement of SVR. According to these results, clearance of HCV by DAA treatment not only regressed liver fibrosis but may have also restrained the progression of PVA. Six PVA cases with persistent HCV infection were reported previously; however, there were no cases who received antiviral treatment [8, 10–14]. As far as we know, this is the first report of PVA in a patient with cirrhosis type C treated with antiviral therapy. Therefore, further investigations are required to consider the effects of elimination of HCV on the progress of PVA. PVA is asymptomatic in most cases, but sometimes results in several complications such as thrombosis, rupture, and compression of inferior vena cava, duodenum, or bile duct due to its mass effect. The most frequent PVA complication is thrombosis, which accounts for 20% of cases [15]. Anticoagulation therapy is recommended for PVA thrombosis, while percutaneous thrombolysis or thrombectomy is indicated for anticoagulation therapy-refractory cases [1]. In the present case, the thrombosis disappeared after therapy with antithrombin III, and thrombosis did not recur following anticoagulant therapy.

In summary, this report describes a PVA in a patient with cirrhosis type C, in which progression ceased after achieving SVR by DAA treatment. PVA can occur and progress in a cirrhotic patient along with aggravation of liver fibrosis and portal hypertension. DAA treatment is proposed not only for the clearance of HCV but also for the control of PVA in a patient with HCV infection. Further investigations are required to validate our findings.

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Study concept and design and drafting of the manuscript: S.H. and T.N. Acquisition of data and critical revision of the manuscript for important intellectual content: S.H., T.N., and K.O. Analysis and interpretation of data: S.H., T.N., K.M., Y.S., K.W., T.H., Yasu.Kawa., M.U., Yu.Kai., R.T., T.Y., K.I., H.U., and K.O. Final approval of the manuscript: all authors.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material files. Further inquiries can be directed to the corresponding author.

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