




Overcoming Complexity: Percutaneous Embolization for Hepatic Artery Pseudoaneurysms in Hereditary Hemorrhagic Telangiectasia (HHT)

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Abstract

Osler-Weber-Rendu syndrome also known as hereditary hemorrhagic telangiectasia (HHT) is a rare vascular dysplasia affecting multiple organ systems in the body. Hepatic manifestations of HHT include telangiectasias, large confluent masses, hepatic perfusion abnormalities, and various types of intrahepatic vascular shunts. Spontaneous hepatic pseudoaneurysm (PA) in HHT is a less well-known phenomenon, with fewer than 20 cases reported in the literature so far. Even rarer is the use of percutaneous glue (N-butyl cyanoacrylate) embolization to manage multiple hepatic PAs in HHT syndrome. The purpose of this case report is to describe the successful management of multiple hepatic PAs by percutaneous embolization in a 56-year-old male diagnosed with HHT.

Keywords

- ▶ hereditary hemorrhagic telangiectasia
- ▶ pseudoaneurysms
- ▶ glue embolization

Introduction

Osler-Weber-Rendu syndrome (OWRS), also known as hereditary hemorrhagic telangiectasia (HHT), is a rare vascular disorder with multiorgan involvement. The “Curacao criteria” for diagnosing HHT include recurrent nosebleeds (epistaxis), mucocutaneous telangiectasias, visceral arteriovenous malformations (AVMs) affecting organs such as the lungs, liver, or brain, and a positive family history of HHT. If a patient satisfies three or more of the criteria, the diagnosis is confirmed. If two conditions are met, HHT is suspected; otherwise, HHT is doubtful.¹ Liver involvement occurs in 74 to 79% of patients with HHT manifesting as telangiectasia, arteriovenous shunting, perfusion abnormalities, and ischemic cholangitis.¹ Although uncommon, hepatic pseudoaneurysms (PSAs) associated with HHT have also been documented in the literature.² Nevertheless, there is

limited data on percutaneous management of hepatic PSAs in individuals with HHT.³ Herein, we present a case of HHT with multiple PSAs in the right hepatic artery (RHA) successfully treated by percutaneous embolization.

Case Presentation

Institutional review board exemption was granted for this study.

A 56-year-old male presented to our hospital with chief complaints of melena and chronic anemia along with a history of occasional nasal bleeding since childhood. Blood investigations revealed anemia (hemoglobin 5.7 g/dL) and elevated liver enzymes. Ultrasound (USG) of the abdomen revealed hepatomegaly along with two anechoic lesions in the right lobe. Color Doppler imaging revealed the classic

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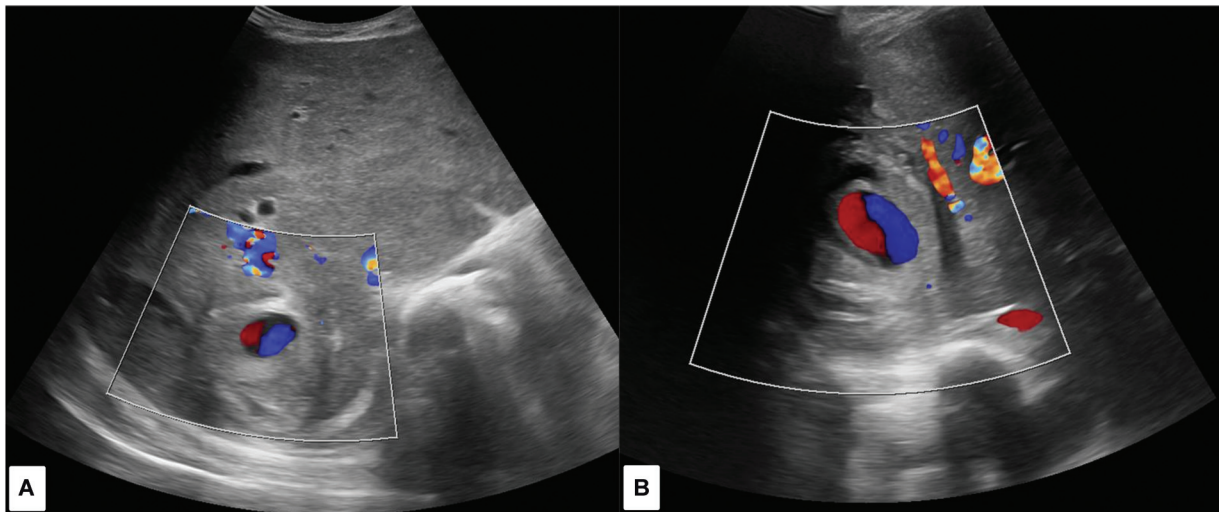


Fig. 1 Doppler ultrasound demonstrating pseudoaneurysms with adjacent hematoma in segments VI (A) and VII (B) of the right lobe of liver.

“yin-yang” sign, with a swirling, bidirectional flow pattern within the lesions, characteristic of turbulent blood flow typically seen in PSAs (►Fig. 1). Further evaluation with computed tomography (CT) angiography of the abdomen (►Fig. 2) revealed dilated tortuous celiac trunk and hepatic artery. The two PSAs observed on USG were seen to be arising from the RHA in segments VI and VII of the right lobe, respectively, along with subcapsular hematoma overlying these liver segments. There were multiple punctate as well as larger confluent telangiectasias and arteriohepatic venous shunting, but no arterioportal or portovenous shunting was

evident. Apart from this, an aberrant branch originating from the celiac artery was seen communicating with the superior mesenteric artery (SMA) near the point of origin of the inferior pancreaticoduodenal artery (►Fig. 2E). Dynamic CT scans were performed for the evaluation of potential AVMs in other organs, such as the brain and lungs, but no AVMs were detected. Based on the history of recurrent epistaxis and imaging findings, a diagnosis of possible HHT was considered, though definitive diagnosis was not possible due to the absence of mucocutaneous telangiectasias and a positive family history.

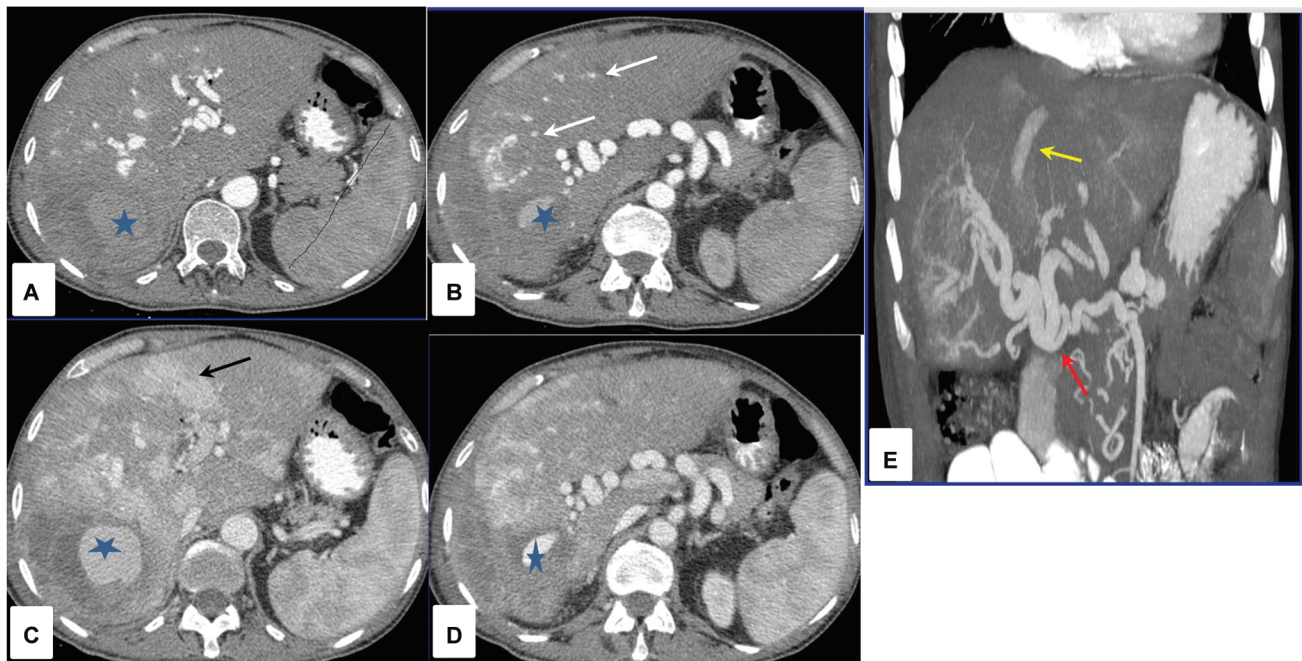


Fig. 2 Multiphase computed tomography (CT) abdomen in arterial (A, C) and venous phase (B, D) images show presence of contrast filled outpouching with surrounding hematoma in segment VII of the right lobe (A, C) suggestive of pseudoaneurysm from the right hepatic artery. Another pseudoaneurysm seen in segment VI of the right lobe (B). Multiple tiny telangiectasias (white arrow) and large confluent masses (black arrow) also seen. Maximum intensity projection (MIP) images (E) show abnormal communicating vessel between the superior mesenteric artery (SMA) and common hepatic artery (red arrow) and middle hepatic vein opacifying in the arterial phase (yellow arrow) indicative of arteriovenous shunt.

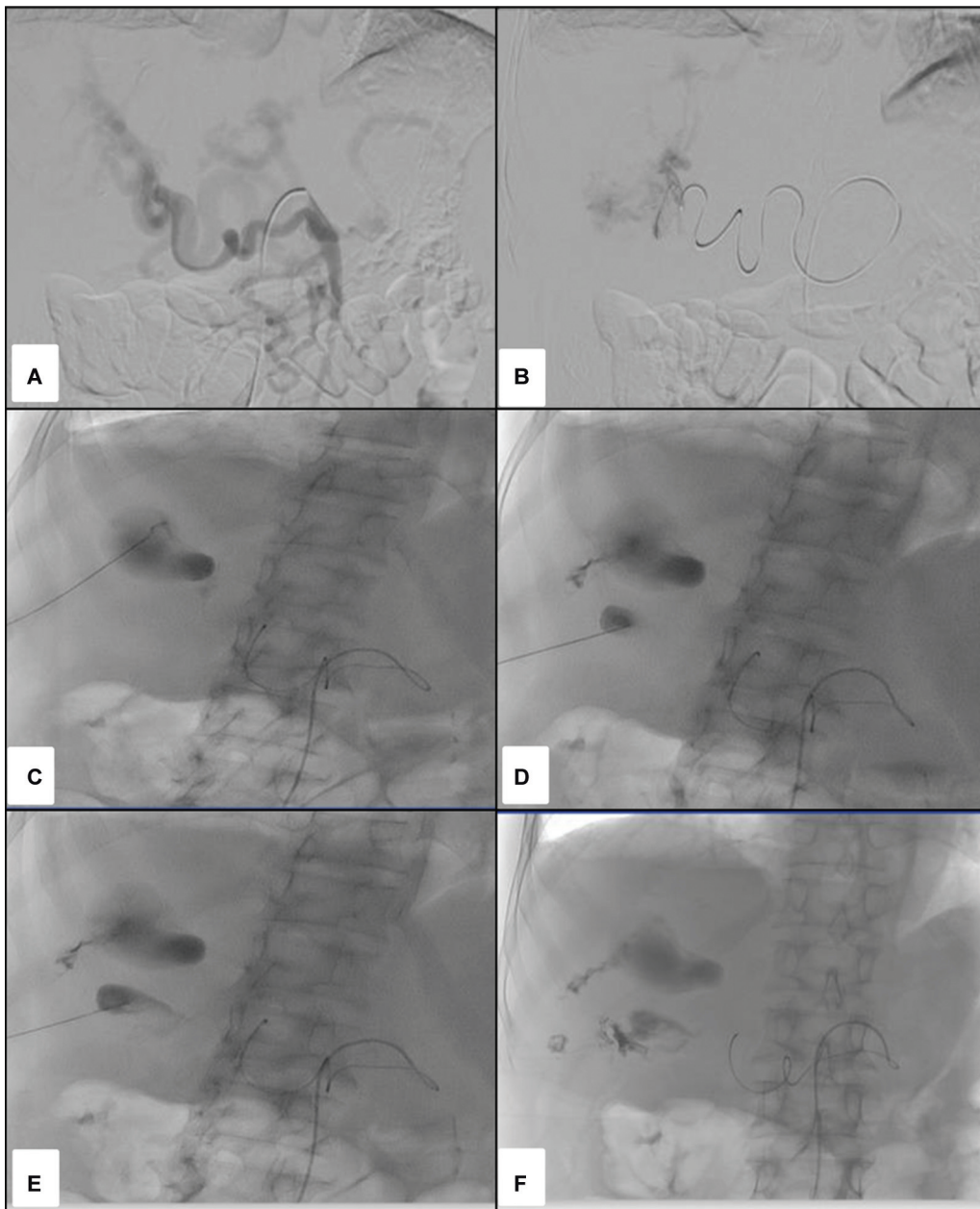


Fig. 3 Angiographic run through the superior mesenteric artery (SMA) showing opacification of branches of the celiac artery via an abnormal communicating vessel (A). Selective angiogram of the right hepatic artery (B) demonstrating nonopacification of both the pseudoaneurysms. Ultrasound (USG) and fluoroscopy-guided injection of glue mixed with lipiodol in both aneurysmal sacs (C–E). Glue cast seen within the aneurysmal sacs and along the needle tracts (F).

Considering the risk of spontaneous peritoneal rupture and high mortality, endovascular embolization of the PSAs was recommended. Digital subtraction angiography (DSA) depicted branches of the celiac artery being opacified during SMA runs alongside the presence of abnormal communicating artery between them (►Fig. 3A). Selective catheterization of the common hepatic artery and RHA failed to opacify the hepatic aneurysms (►Fig. 3B), likely due to arteriovenous shunting. Hence, endovascular coiling was deemed unfeasible, and it was decided to proceed with percutaneous embolization.

Under local anesthesia and USG guidance, each PSA was punctured using a 23G Chiba needle (Cook Medical, Bloomington, Indiana, United States). Iodinated contrast was injected to

visualize the PSA on fluoroscopy and to detect any reflux in the major arteries (►Fig. 3C). After flushing the needle with 5% dextrose solution, n-butyl cyanoacrylate glue (Trufill, Cordis, Miami Lakes, Florida, United States) mixed with lipiodol (Lipiodol Ultra-Fluid, Guerbet, Villepinte, France) in 1:1 ratio (50%) was injected targeting the neck of the PSA in multiple aliquots (►Fig. 3D). For the larger PSA in segment VII, a total of 1.4 mL of the glue-lipiodol mixture was administered, while 0.6 mL of the mixture was used for the smaller PSA in segment VI. The PSA was observed to thrombose in real time under USG guidance, with no flow detected within the aneurysm on color Doppler USG. Glue administration was continued as the needle was removed to plug the percutaneous tract (►Fig. 3E, F) and

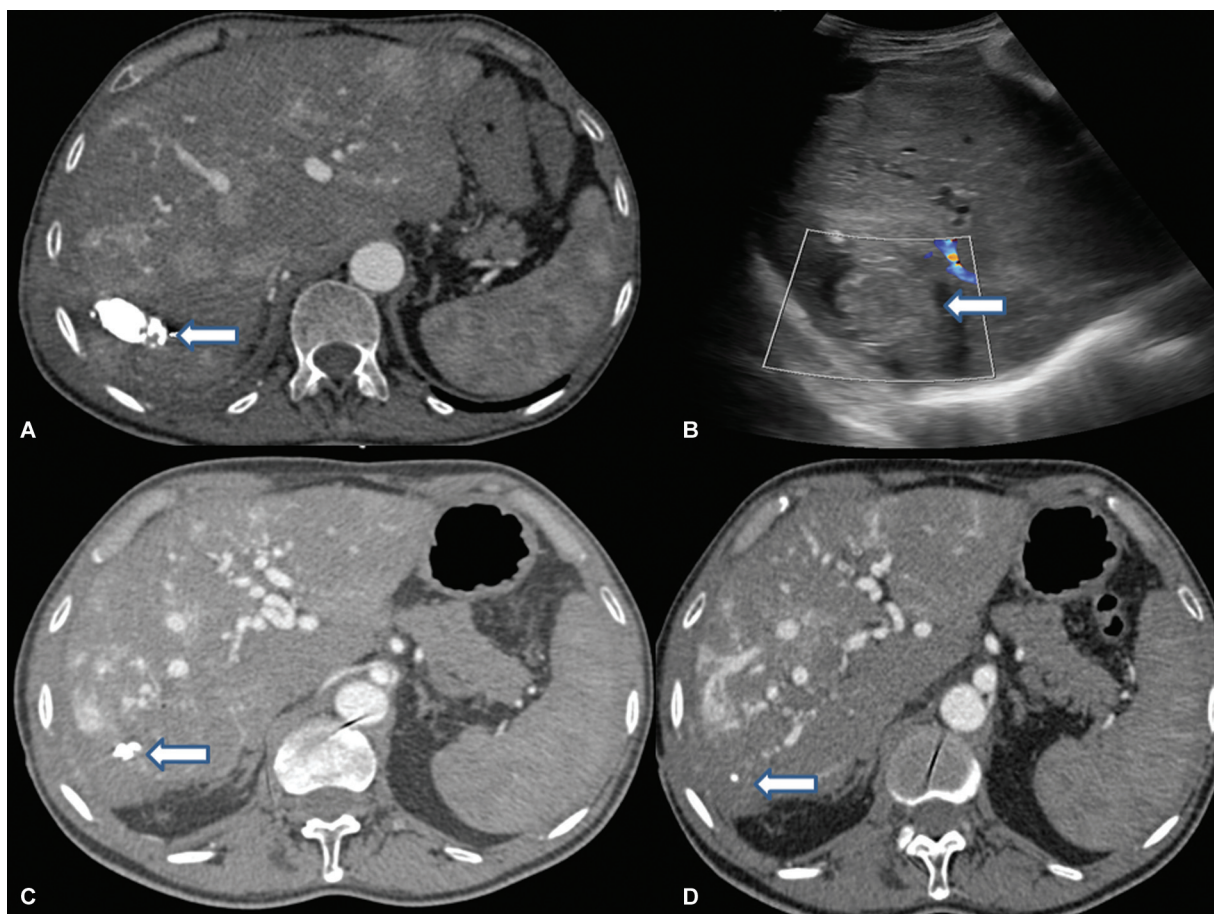


Fig. 4 Immediate postintervention computed tomography (CT) angiography image demonstrating a nonopacified aneurysm with high-density glue cast (arrow), confirming complete thrombosis (A). Follow-up ultrasound at 1 month postembolization demonstrating complete thrombosis of the pseudoaneurysm in segment VII with no detectable flow on color Doppler, confirming successful treatment (B). Follow-up CT at 6 months postembolization shows a significant reduction in the size of the glue cast in both the lesions (C, D).

prevent any possible bleeding from the parenchymal tract. There were no periprocedural complications and the patient's symptoms improved posttreatment. Postembolization CT angiography revealed a nonopacified aneurysm with high-density glue cast, indicating complete thrombosis (► Fig. 4A). Follow-up USGs at 1 and 3 months revealed no flow within the PSAs, confirming successful treatment (► Fig. 4B). A follow-up CT at 6 months demonstrated a significant reduction in the size of the glue casts in both PSAs, further supporting the effectiveness of the intervention (► Fig. 4C, D).

Discussion

Liver involvement can be seen in approximately 74% of all cases of HHT; however, only 8 to 10% of patients are symptomatic.⁴ Hepatic manifestations can range from small telangiectasias to large confluent masses, transient perfusion abnormalities, and AVMs.⁵ Hepatic arterial aneurysms are less commonly seen, but can occur due to high flow secondary to the arteriovenous or arterioportal shunting.

While there is no permanent cure for HHT, treatment aims at managing symptoms and complications associated with it. Hepatic PSAs had previously been treated mostly with surgery. However, advances in interventional radiology

have made endovascular interventions, such as embolization and coiling, the preferred therapeutic options for hepatic AVMs and PSAs because of their minimally invasive nature and relatively low morbidity rates.⁶ However, successful endovascular embolization may be difficult in cases with complex vascular anatomy or extensive arteriovenous shunting, as seen in our case, in which the hepatic PSAs did not opacify even on superselective runs of the RHA and its branches. Hence, percutaneous embolization was pursued for exclusion of the PSAs.

Several studies have reported the management of hepatic PSAs in HHT patients, although the difficulty in visualizing these aneurysms on DSA is not commonly emphasized. For example, Miyabe et al⁷ discussed a case of hepatic aneurysm in an HHT patient, where angiographic visualization was challenging due to the vascular malformations associated with HHT. However, they successfully treated the aneurysm using coil embolization, demonstrating the efficacy of this approach even in the presence of complex vascular anatomy. Similarly, Çıldıg et al⁸ reported on a patient with OWRS and an aberrant RHA originating from the SMA, where they used coil embolization and stent grafting to manage a giant hepatic aneurysm.

To the best of our knowledge, there is no existing data on the management of hepatic artery PSAs in complex anatomical

cases, such as those seen in OWRS, using percutaneous glue embolization. Percutaneous embolization is a cheaper and time-effective alternative to endovascular embolization. When treating visceral PSAs by percutaneous approach, glue and thrombin offer distinct advantages and disadvantages. Glue offers immediate and permanent occlusion of the PSA. Moreover, its viscosity can be adjusted for precise control, and its radiopacity when mixed with lipiodol permits real-time monitoring during the treatment, reducing the risk of incomplete embolization.⁹ However, glue handling requires expertise to minimize nontarget embolization. On the other hand, thrombin is easier to handle and has a lower risk of causing irreversible damage. However, it is associated with higher risk of recanalization, thromboembolic complications, as well as allergic reactions.⁹ Moreover, its effectiveness can be limited in high-flow conditions due to rapid washout, and it might not provide the same level of endurance as glue, often necessitating repeat interventions. Therefore, the choice between glue and thrombin depends on the specific clinical scenario, with glue being favored for its stability and thrombin for its ease of use and safety profile in certain contexts. In view of the high-flow resulting from arteriovenous shunting in our case, glue was preferred over thrombin injection.

The long-term management of AVMs in HHT patients who undergo glue embolization or other interventions remains challenging due to the disease's inherent vascular instability and tendency for recurrence. Glue embolization, while effective initially, may only provide temporary control, as recurrent AVM growth is common due to ongoing angiogenesis and revascularization driven by HHT's underlying genetic mutations. This leads to the need for repeat interventions, especially in high-flow pulmonary or hepatic AVMs, where revascularization and new malformations are frequently observed.

While percutaneous embolization has been successfully used to treat pulmonary and nasal AVMs in HHT, literature on its application in liver PSAs is limited.^{10,11} This case report emphasizes the need for further research to illustrate the efficacy of percutaneous embolization in this context.

Conclusion

Our case report illustrates a rare HHT manifestation including ruptured intrahepatic PSAs with subcapsular hematoma. It also highlights the complexities of endovascular emboli-

zation due to the complex vascular anatomy and the role of percutaneous embolization for hepatic PSAs in HHT with successful outcome in our patient.

Conflict of Interest

None declared.

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