Post-thrombotic syndrome of the lower extremity associated with arteriovenous fistula: Three case reports

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ABSTRACT

The post-thrombotic syndrome is a serious complication of deep vein thrombosis. Post-thrombotic iliac venous occlusion (PTIVO) is rarely associated with an arteriovenous fistula (AVF); however, the underlying mechanism remains unclear. We reported three PTIVO cases associated with an AVF, two symptomatic cases treated with venous stenting, and one asymptomatic case followed up conservatively. The essential imaging findings to diagnose PTIVO with an AVF were the presence of an arterial waveform on ultrasound examination and early opacification on contrast-enhanced computed tomography in the iliofemoral veins. Venous stenting resulted in the disappearance of the AVF, drastic improvement in symptoms, and an excellent long-term outcome. (J Vasc Surg Cases Innov Tech 2022;8:629-33.)

Keywords: Post-thrombotic syndrome; Arteriovenous fistula; Venous stenting; Chronic venous insufficiency

Acquired arteriovenous fistula (AVF) occurs in venous diseases such as acute deep vein thrombosis (DVT), varicose veins, and May-Thurner syndrome.[1-3] Post-thrombotic iliac venous occlusion (PTIVO) has also been reported to be associated with an AVF; however, most of these reports were case reports, and the pathogenesis and the mechanism remain unclear.[4-8] We reported three cases of PTIVO with an AVF: two symptomatic patients who underwent venous stenting (VS) and one asymptomatic patient who was followed up conservatively. This study reported the efficient examinations and optimal management of this condition. The patients consented to the publication of this case report.

CASE 1

A 91-year-old woman with a medical history of DVT in her left common iliac vein (CIV) and external iliac vein (EIV) 6 months earlier presented with worsening left lower extremity swelling and pain (Fig 1, *A*). Duplex ultrasound (DUS) examination demonstrated occlusion of the left CIV and high-velocity arterialized waveform (Fig 1, *B*) in the left femoral vein (FV). Contrast-enhanced computed tomography (CECT) showed enhancement of the left common FV (CFV) in the arterial phase and occlusion of the left CIV and EIV with highly developed collateral vessels in the venous phase, suggesting PTIVO with an AVF (Fig 1, *C-E*).

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Venography revealed findings similar to those in the venous phase of CECT (Fig 2, *A*). VS (SMART 14 × 60 mm, 14 × 40 mm, Cordis Corp; E-Luminexx 14 × 60 mm, 10 × 60 mm, Bard Inc) was performed in the left CIV, EIV, and CFV (Fig 2, *B*). After stenting, the collateral vessels remained undetected (Fig 2, *B* and *C*). During the VS, we measured the blood pressure in the left CFV (15/8 mm Hg), with an arterial waveform before stenting, which dropped to 3/0 mm Hg with a venous waveform after stenting (Supplementary Video, online only). The symptoms improved on postintervention day 1, and DUS examination showed that the arterialized waveform at the FV had changed to a venous waveform (Fig 2, *D* and *E*). Subsequently, the patient had good long-term patency with no symptom recurrence at the final follow-up 31 months after VS.

CASE 2

An 82-year-old man with a history of DVT 4 years before presented to another hospital with stasis ulcers on the left lower extremity (Fig 3, *A* and *B*) and bleeding at the scrotum. He was diagnosed with left CIV occlusion and underwent Palma-Dale surgery at the hospital; however, the bypass occluded a few days postoperatively. Subsequently, the patient was referred to our hospital because of worsening symptoms. DUS examination revealed occlusion of the left CIV and pulsatile flow in the left CFV. CECT revealed occlusion of the left CIV with highly developed collateral vessels and early opacification of the left iliac veins (Fig 3, *C* and *D*). These findings suggested that PTIVO with an AVF and the highly developed collaterals with venous hypertension were the cause of the scrotal bleeding. We performed VS (E-Luminexx 14 × 60 mm; Bard Inc) in the left CIV, and the developed collateral vessels were dramatically reduced (Fig 4, *A-D*). After VS, the scrotal bleeding resolved and the ulcers on the left lower extremity healed by postintervention day 10 (Fig 4, *E* and *F*). The patient showed no symptom recurrence at the last follow-up 52 months after VS.

CASE 3

A 46-year-old man with a history of DVT who had post-thrombotic occlusion of the left iliac veins was followed up

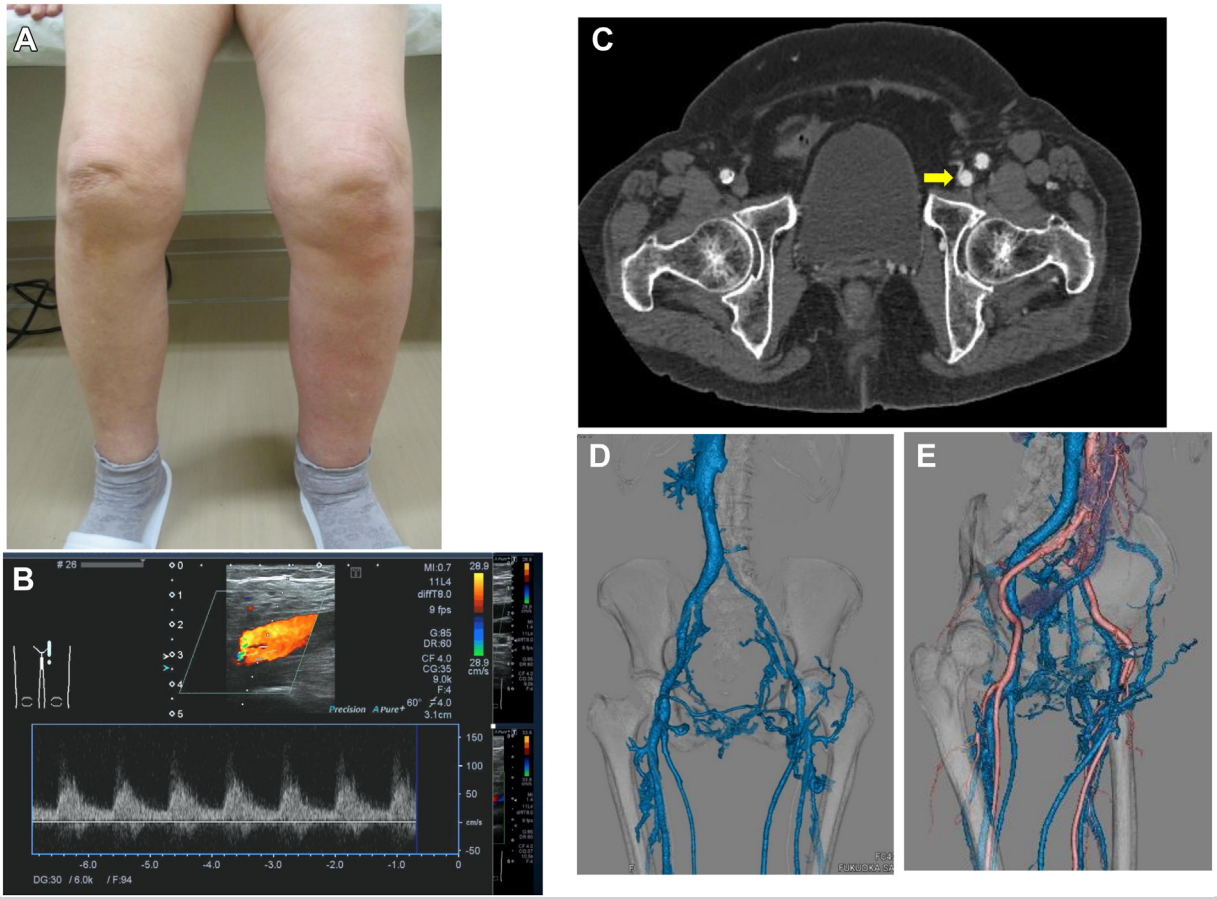

Fig 1. Preoperative clinical data of case 1. **A,** Photograph of the patient demonstrating left lower extremity swelling and inflammation. **B,** Duplex ultrasound examination demonstrated an arterialized waveform in the left common femoral vein (CFV). **C,** Contrast-enhanced computed tomography (CT) showed early opacification of the left CFV (*yellow arrow*). **D,** CT angiography three-dimensional (3D) reconstruction (venous phase). Occlusion of the left iliac vein with highly developed collateral vessels. **E,** CT angiography 3D reconstruction (arterial and venous phase). The exact locations of the arteriovenous fistula (AVF) were not identified.

without endovascular intervention because the lower extremities were asymptomatic (Fig 5, *A*). Four years after DVT, regular follow-up DUS examination showed an arterialized waveform at the distal part of the occluded left EIV (Fig 5, *B*), suggesting PTIVO with an AVF. CECT revealed occlusion of the left CIV and EIV with moderately developed collaterals and early opacification of the left EIV (Fig 5, *C-E*). His lower extremities were asymptomatic, and AVF was found incidentally during a routine follow-up examination. Therefore, no intervention was conducted, and the patient was currently under close surveillance and followed up without worsening symptoms for the next 18 months.

DISCUSSION

The following interesting findings were noted in this study: (1) PTIVO with an AVF was observed, but no AVF was observed in the acute DVT phase; (2) the location of the AVF was not identified, but the developed collateral vessels might be the source of the condition; (3) VS resulted in the disappearance of the AVF and a drastic improvement in symptoms; and (4) asymptomatic cases may also exist. The AVF develops during the transition from DVT to post-thrombotic syndrome (PTS); however, determining how the AVF develops over time and the mechanism remains unclear. According to the hemodynamics of PTS, collateralization and venous hypertension may contribute to the association with an AVF. Post-thrombotic remodeling and increased venous pressure may promote neovascularization,[9,10] and the latent micro-arteriovenous network may become more evident owing to vasodilation of the venous component caused by the development of collateral veins and venous hypertension. Interestingly, a reversal of the arterial waveform in the venous segment was observed immediately after VS.

In case 2, the patient had severe PTS symptoms, such as recalcitrant ulceration and highly developed collateral blood vessels with venous hypertension due to the association with AVF causing scrotal bleeding. We believed that PTS associated with AVF would be exposed to a

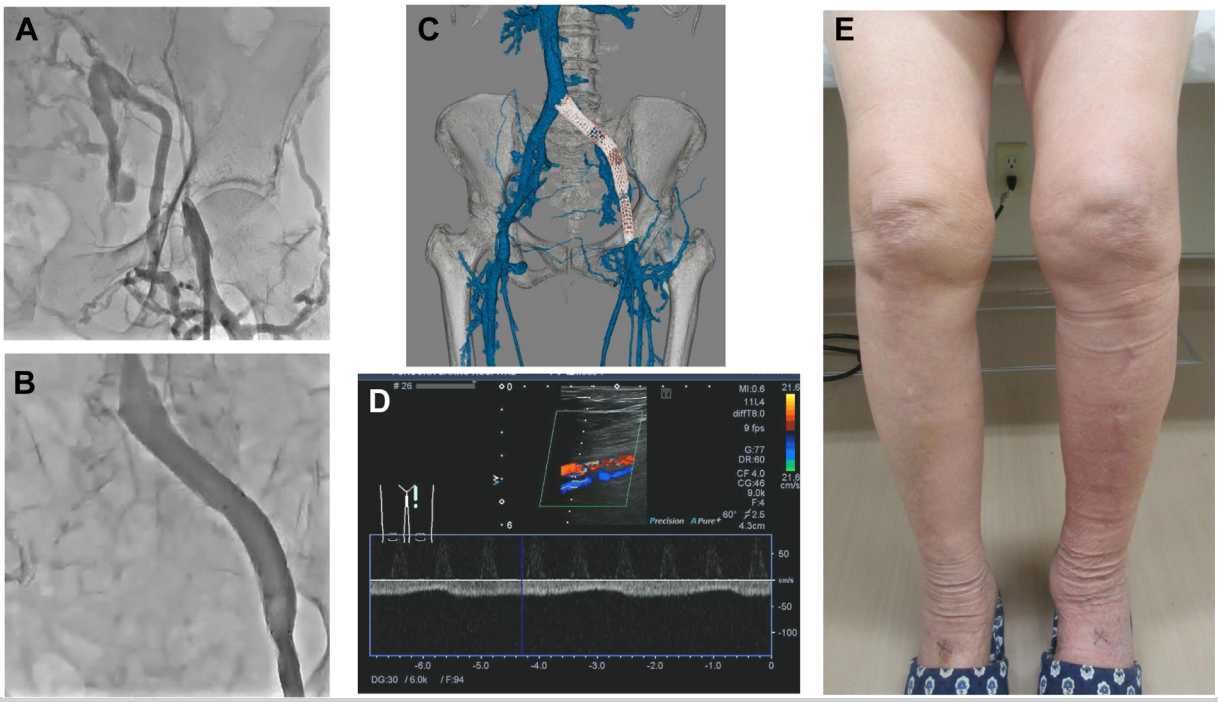

Fig 2. Operative and postoperative data of case 1. **A,** Venography before stenting showed occlusion of the left iliac veins with highly developed collateral vessels. **B,** Venous stenting (VS) was performed in the left common iliac vein (CIV), external iliac vein (EIV), and common femoral vein (CFV). The developed collateral vessels were undetectable. **C,** Computed tomography (CT) angiography three-dimensional reconstruction (venous phase) showing patent venous stent with decreased collaterals. **D,** Postoperative ultrasound examination showed blood flow of the left CFV with a venous waveform. **E,** Postoperative photograph. No swelling was observed in the left lower extremity.

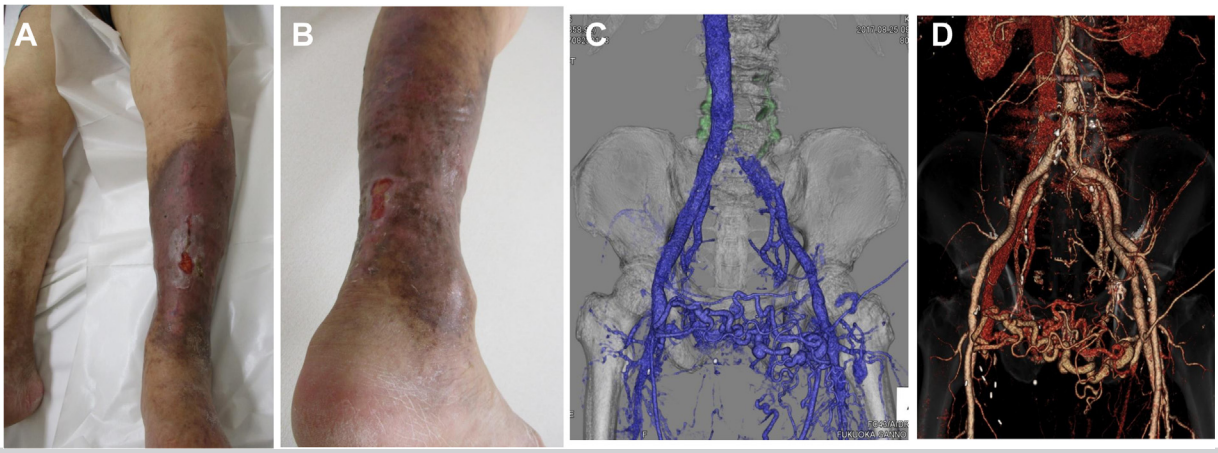

Fig 3. Preoperative clinical data of case 2. **A,** Stasis ulcer on the anterior tibial region with extensive hyperpigmentation of the left lower extremity. **B,** Stasis ulcer on the dorsal surface with extensive hyperpigmentation of the left lower extremity. **C,** Computed tomography (CT) angiography three-dimensional (3D) reconstruction (venous phase) showing occlusion of the left common iliac vein (CIV) with highly developed collateral vessels. **D,** CT angiography 3D reconstruction (arterial and venous phase) showing early opacification of the left iliac veins. The exact locations of the arteriovenous fistula were not identified.

much higher venous pressure, resulting in higher clinical severity. However, the actual venous pressure measured in case 1 was 15/8 mm Hg, which was not very high. Surprisingly, there was also an asymptomatic case (case 3). The drastic decrease in collateral veins and the simultaneous disappearance of the AVF immediately after VS

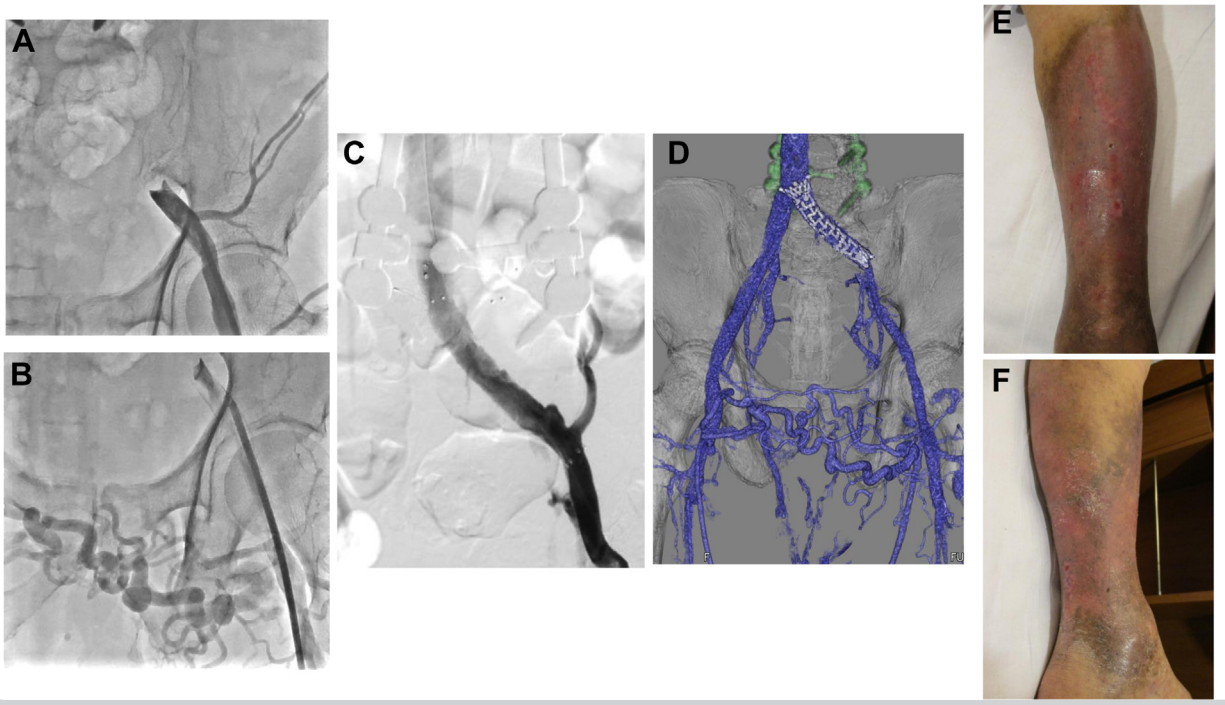

Fig 4. Operative and postoperative data of case 2. **A,** Venography before stenting showed occlusion of the left common iliac vein (CIV) with highly developed collateral vessels **(B)**. **C,** Venous stenting (VS) was performed in the left CIV. The developed collateral vessels were undetectable. **D,** Computed tomography (CT) angiography three-dimensional reconstruction (venous phase) showing patent venous stent with decreased collaterals. **E,** *and* **F,** Healing of the skin ulcers after VS.

suggest that collateralization and venous dilatation rather than venous hypertension are essential factors in the pathogenesis of this condition.

DUS examination, which can assess the occluded vein and the existence of an AVF according to the waveform, should be the initial examination because of its low invasiveness. CECT is useful in determining the precise location of the stenosis, and for optimal VS treatment, and necessary to rule out differential diagnoses such as iatrogenic injury, neoplasms, and erosion of an arterial aneurysm, which are the most common causes of an AVF. Obtaining images in the arterial and venous phases is a key to evaluating this condition efficiently. We performed arterial phase imaging immediately after contrast injected from the arm was identified in the popliteal artery, and venous phase imaging was performed 90 seconds later. Owing to its effectiveness, we used this protocol to evaluate all PTS cases, not only those with an AVF.

Treatments for this condition have been reported, including VS, transarterial embolization, arterial stent-graft, and venous bypass (Palma-Dale surgery).[1,4-8] Treatments on the arterial side, such as transarterial embolization and arterial stent-graft, often require additional treatment on the venous side[6,8] and may not be necessary because the main pathogenesis of this condition is in the venous component, and it is difficult to identify a definite AVF location in the preoperative evaluation. Although surgical venous reconstruction used to be the only treatment option, the outcomes were suboptimal,[11] and it is not the first-line treatment in this endovascular era.[12] VS in chronic iliocaval venous outflow obstruction has been well accepted because of its safety and efficacy.[13,14] Similar to other case reports, VS showed a drastic improvement in symptoms and continued curability in the present study.[1,6,7] However, the indication for VS should be limited to symptomatic cases because there are asymptomatic cases of PTIVO with an AVF.

CONCLUSION

We reported three cases of PTIVO with an AVF: two symptomatic cases treated with VS and one asymptomatic case followed up conservatively. The essential imaging findings to diagnose PTIVO with an AVF were the presence of an arterial waveform on DUS examination and early opacification on CECT in the iliofemoral veins. VS resulted in the disappearance of the AVF, a drastic improvement in symptoms, and an excellent long-term outcome.

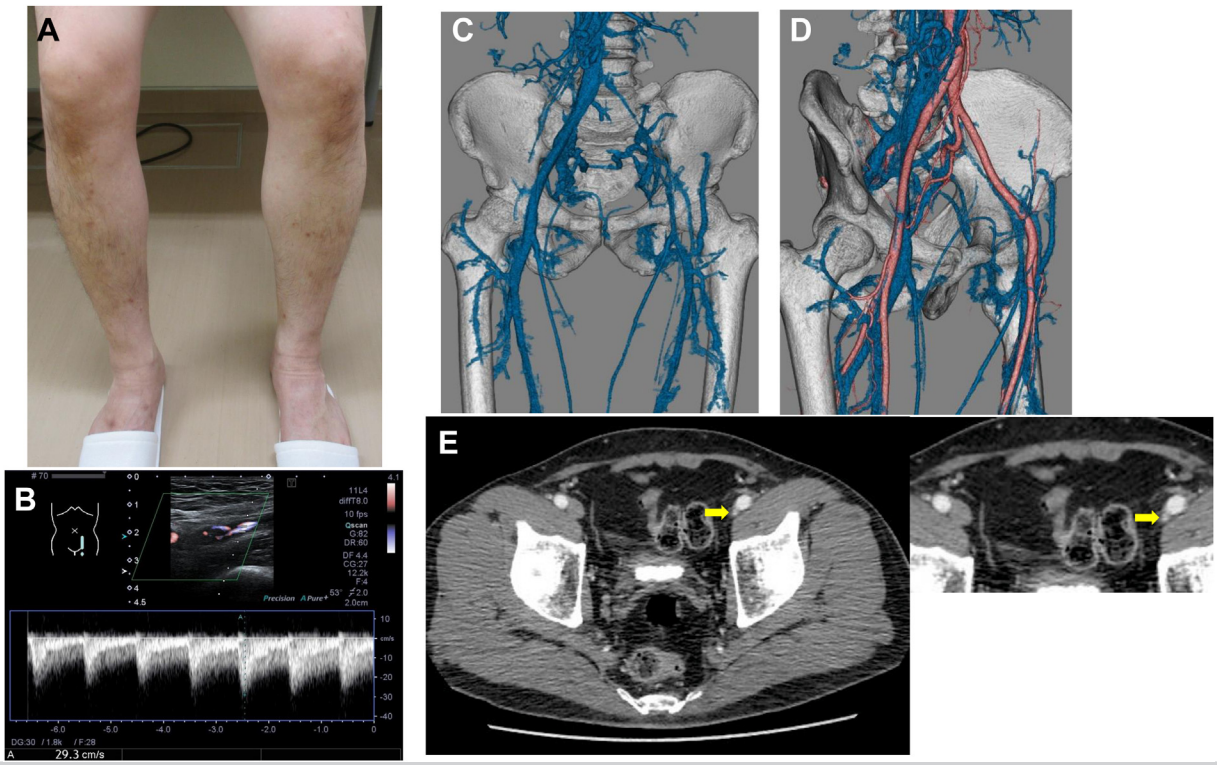

Fig 5. Clinical data of case 3. **A,** No signs of swelling or dermatitis symptoms in the left lower extremity. **B,** Duplex ultrasound examination showed an arterialized waveform at the distal part of the occluded left external iliac vein (EIV). **C,** Computed tomography (CT) angiography three-dimensional (3D) reconstruction (venous phase). Occlusion of the left common iliac vein (CIV) and EIV with moderately developed collateral vessels. **D,** CT angiography 3D reconstruction (arterial and venous phase). The exact locations of the arteriovenous fistula were not identified. **E,** Contrast-enhanced CT showing early opacification (*yellow arrow*) of the distal part of the occluded left EIV (with its enlarged view).

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