Endovascular Management of Postthrombotic Ilio-iliac Arteriovenous Fistula with Occluded Common Iliac Vein: A Case Report and Literature Review

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Abstract

Chronic venous hypertension as a result of May–Thurner syndrome (MTS) and subsequent venous thrombosis is a frequently observed clinical condition. However, a postthrombotic arteriovenous fistula (AVF) is a phenomenon not reported commonly. Such conditions have been treated surgically and by endovascular approach – embolization or stenting. We present a case of concomitant MTS and pelvic AVF in a patient with recurrent deep vein thrombosis, who also had lumbar laminectomy in the past, resulting in massive and debilitating left lower extremity edema and bleeding varices in the left lateral thigh, which was successfully treated with the placement of stent graft/stent in both arterial and venous systems.

Keywords: Pelvic arteriovenous fistula, May–Thurner syndrome, Ilio-iliac AVF

Introduction

May–Thurner syndrome (MTS), also known as Cockett syndrome or iliac vein compression syndrome, is an anatomical variant wherein an overriding right common iliac artery compresses the left common iliac vein (CIV). They described the presence of “spurs” in the wall of the iliac vein caused by chronic irritation and compression by the iliac artery, resulting in venous stenosis predisposing to thrombosis. It has rarely been reported with other vascular abnormalities. Acquired arteriovenous fistulas (AVFs) have been reported following thrombosis of lower extremity veins or cerebral veins. Varnagy and Labropoulos reported the occurrence of spontaneous AVF postvenous thrombosis in both humans and animals, with the latter being an initiating factor of angiogenesis. Link et al. have published case reports of peripheral arteriovenous malformations (AVMs) inside and around previously thrombosed veins as a result of angiogenesis secondary to a vascular stress mechanism.

We present a case of concomitant MTS and pelvic AVF in a patient with recurrent deep vein thrombosis (DVT), resulting in massive and debilitating left lower extremity edema and bleeding lateral thigh varices that was successfully treated with endovascular techniques.

Case Report

A 64-year-old female presented to the emergency department in the 1st week of December 2017, with a history of severe intrac table left lower limb swelling, gradually increasing over the past 1 year, and a sudden onset of continuous, uncontrollable bleeding from a small ulcer in the mid-lateral left thigh for 2 hours. Multiple superficial, dilated, and tortuous collateral veins were noted in the lateral aspect of the left thigh, abdomen, and suprapubic region. No thrill/bruit was present. Bleeding was controlled with compression dressing.

In 2014, she was diagnosed with acute DVT from left CIV to popliteal vein and anticoagulated for 3 months. Six months later,
she underwent complete laminectomy of L5–S1 spine for chronic back pain. She was asymptomatic till May 2016 when she was diagnosed with recurrent acute left lower limb DVT from the iliac to the tibial veins, treated with anticoagulation irregularly. She first presented to us in December 2016 with gradually increasing debilitating swelling of the limb. Venous duplex revealed acute thrombus in the superficial femoral and popliteal veins. Computed tomography (CT) venogram was suggestive of MTS with an occluded left CIV with no other findings. In view of acute DVT, no immediate intervention was planned and treated with anticoagulation and compression stockings. She again presented with bleeding varices in the lateral mid-thigh diagnosed as acute recurrent DVT in the distal superficial femoral and popliteal vein in October 2017. She was treated with injection sclerotherapy (with STD) to control and prevent rebleeding. She was then discharged on oral anticoagulation.

A CT venogram was repeated now which revealed a chronically occluded left CIV, dilated external iliac vein (EIV) (25 mm), and patent femoral and popliteal veins with multiple huge collaterals in the subcutaneous region of abdomen, pelvis, and left thigh (20 mm) [Figure 1]. She was taken up for stenting of the occluded left CIV. Venogram through the left common femoral vein (CFV) revealed similar findings as the CT. There was smooth passage of a Terumo 0.035″ guidewire into the presumed inferior vena cava, but the position of the wire was noted to be on anterior, left of the lumbar vertebrae. On angiogram, visualization of the aorta and bilateral iliac arteries and early visualization of the dilated left EIV and collaterals without extravasation were seen. Selective angiogram through a right common femoral artery (CFA) access revealed the presence of a fistulous communication between the mid-third of the left internal iliac artery (IIA) and the left EIV just below the occluded CIV [Figure 2]. Procedure was terminated for better planning.

Repeat duplex scan visualized the fistula, with reversal of flow in the left EIV and CFV with arterial pattern of waveforms [Figure 3]. A review of the previous CT venogram also revealed the presence of the AVF about 5 mm below the level of sacral promontory [Figure 2] and two small collaterals from the aortic bifurcation region and left distal external iliac. After considering available options, decision was made to place a stent graft in the left IIA since there were adequate landing zones. On December 23, 2017, a Lifestream 8 mm × 58 mm balloon-mounted stent graft was then deployed in the left IIA through right CFA approach which completely excluded arterial flow into the fistula [Figure 4].

Although there was rapid decrease in the limb girth, bleeding from thigh varix continued to be significant. Hence, 2 days later, a 20 mm × 18 mm Wallstent (Boston Scientific) was deployed with access from the left CFV and right internal jugular vein using “railroad” technique [Figure 4]. There was near-complete disappearance of venous collaterals. STD was used to sclerose the branches around the bleeding ulcer. Limb girth reduced to 32 cm from a preoperative 66 cm [Figure 5]. Duplex revealed no arterial flow in the left EIV [Figure 3]. She was then discharged on anticoagulation and compression stockings. There was no further bleeding at 1-month follow-up and the patient was symptom-free. CT angiogram revealed both stents to be patent, no leak through the stent graft, collaterals from the aorta and external iliac artery not visualized, and complete thrombosis of the venous collaterals [Figure 6].

**DISCUSSION**

The process of revascularization of a thrombus may result in a biochemical environment where the resultant lesions may rapidly shunt arterial flow to the venous side, resulting in AVMs. This phenomenon has been observed in the transverse sinuses[8,9] and in the iliac and femoral veins.[10] The robust response of vasculogenesis of the vasa vasorum in diseased arteries could also be observed in venous vasa vasorum.[11,12] High venous pressure opens the physiological arteriovenous shunt in a retrograde fashion. This has been demonstrated in surgically induced venous hypertension model in rats where it was confirmed that venous hypertension without thrombosis can cause dural AVF.[13,14]

There have been only four reports of similar cases of MTS with pelvic AVF published in English literature with different modalities.
Sravan, et al.: Ilio-iliac AVF endovascular management

used to treat each of them as all cases reported had multiple minor fistulae. Yuan et al.\(^{15}\) described the use of only stenting of the occluded left CIV as a treatment to relieve the venous hypertension. Huynh et al.\(^{16}\) described the use of concomitant coil embolization (azur coils) of the fistula and stenting of the occluded CIV as the method of choice. Mihmanli et al.\(^{17}\) described a case of MTS and AVF diagnosed by CT, but further details of treatment are not described. Link and Granchi\(^{18}\) treated a patient presenting with nonhealing ulcers with embolization of the fistula using coils and Onyx embolic agent. Further sclerotherapy of the veins near the ulcer was needed as the ulcer remained in an unhealed state. Recanalization of the occluded iliac vein was not done.

Confounding factor in this patient is the previous lumbar laminectomy, which could have also contributed to this AVF (though unlikely, in our opinion). Linton and White first reported a pelvic AVF after intervertebral disk surgery in 1945.\(^{19}\) Lumbar disk surgeries performed through a posterior approach may perforate the anterior spinal ligament (usually by a pituitary rongeur) and may be associated with vascular injuries at a frequency of up to 5/10,000 with 67% presenting as an AVF.\(^{20,21}\) Seventy five percent of iliac AVFs after laminectomy is at the L4-L5 level.\(^{22}\) van Zitteren et al.\(^{23}\) in a review of literature from 2002 to 2011 reported that only 2.3% of cases involved injury of the left IIA. Fistulas occurring at L5–S1 level are the most anatomically variable.\(^{24}\) Although congestive heart failure is common, pathognomonic features are absent in up to 50% of patients.\(^{25}\) A machinery type of bruit is noted in the most of patients.\(^{26}\) About 25% of AVFs remain undiagnosed even after 1 year of index surgery. This may be due to the absence of characteristic findings, rare occurrence, and lack of awareness. Failure to make the diagnosis has been emphasized by Baker et al.\(^{27}\) and Dardik et al.\(^{28}\) Recently, Pawar et al. reported two cases with succinct review of literature.\(^{29}\)

Diagnosis of a pelvic AVF by duplex has reported to have 98% accuracy.\(^{30}\) Computerized tomography is reliable with sensitivity up to 95% and specificity up to 90%. It is especially useful in diagnosing in anatomically inaccessible pelvic locations.\(^{31}\) Conventional angiography remains the gold standard for diagnostic purposes and is also a therapeutic option for endovascular exclusion. Treatment may be surgical or endovascular exclusion of the fistula. In spite of availability of modern surgical equipment, surgery is still associated with high mortality rates of 5%–10%.\(^{32}\) Hence, endovascular techniques have taken precedence in the last decade. Zajko et al.\(^{32}\) performed the first reported endovascular repair of an iliac AVF. Similarly, several authors have shown the effectiveness of endovascular stent grafts to exclude pelvic AVFs, such as Ventura et al.,\(^{33}\) Schneider et al.,\(^{34}\) and Cronin et al.\(^{35}\) Larger series emphasize the high success rate and low morbidity and mortality related to endovascular approach.\(^{36,37}\)

The present patient has presented with severe venous hypertension due to the combined effect of the pelvic AVF, MTS,
and postthrombotic syndrome. Spine surgery resulting in the fistula is less likely in this patient as previous records suggest only laminectomy and no additional discectomy. Her present imaging also showed intact anterior and mid segments of the L5–S1 intervertebral disc and the fistula was 5 mm distal to sacral promontory. On contrary to the previous reported cases wherein multiple feeder arteries were present, the size of the fistula is quite large in the present case. Such large communication in the iliac level would usually result in right heart failure in view of the increased preload, but due to the occluded left CIV, the pressure was diverted to the left lower limb and pelvis, resulting in a large number of collateral channels.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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