Iliac May-Thurner syndrome coexisting with arteriovenous fistula treated by endovascular therapy

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Abstract
An 82-year-old woman presented with persistent left leg swelling despite 3 months anticoagulation for deep vein thrombosis. Ultrasound showed venous blood flowing back into the toe. Enhanced computed tomography scans and angiography showed occlusion of the left common iliac vein and a large number of arteriovenous fistulas. We made a diagnosis of May-Thurner syndrome coexisting with arteriovenous fistulas and we performed endovascular therapy only for the common iliac vein. We considered embolization for arteriovenous fistulas, but it seemed to be difficult to perform because of the possibility of development of collateral circulation and a large number of arteriovenous fistulas. After percutaneous balloon angioplasty, antegrade venous blood flow was restored, and stent implantation was not performed. After the procedure, swelling of the leg gradually reduced. She had no symptoms for the following 1 year.

Keywords
Angiography, arteriovenous fistula, endovascular therapy, deep vein thrombosis, venous occlusion

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Introduction
May-Thurner syndrome (MTS) is a rare condition that may exhibit swelling, pain, or deep vein thrombosis (DVT) of the left lower extremity when the left common iliac vein is compressed by the right iliac artery and the lumbar spine. An iliac arteriovenous fistula (AVF) is also a rare condition, mostly caused by traumatic, congenital, or iatrogenic origin. There have been some reports about favorable outcomes of MTS treated by endovascular therapy; however, MTS coexisting with iliac AVFs is rare, and appropriate treatment has not been established.

We report a case of MTS coexisting with AVFs appearing as edema of the left lower extremity, which was successfully treated by endovascular therapy.

Case description
An 82-year-old woman presented with subacute onset of swelling of the left leg. Her vital signs and physical examination results were unremarkable except for extensive edema of the left leg. She had no medical history of cardiovascular or pulmonary disease, malignant tumor, trauma, or surgery. Ultrasound of the lower extremities revealed DVT of the left superficial femoral vein to the popliteal veins. She was given a direct oral anticoagulant (rivaroxaban, 15 mg bid) and compression stockings for one month. Three months later, although DVT had completely disappeared, swelling of the leg remained. Ultrasound showed no DVT, but the arterial flow pattern indicated venous blood flowing back into the toe (Figure 1(a)) and diffuse iliac AVFs (Figure 1(b)). Enhanced computed tomography scans showed left external iliac vein dilation, some iliac AVFs, and occlusion of the common iliac vein.

Lower left extremity angiography showed the presence of multiple AVFs in the common and internal iliac arteries (Figure 2(a) and (b)) and the occluded common iliac vein (Figure 2(c)). Embolization for AVFs seemed to be difficult to perform because of the possibility of development of collateral circulation and a large number of AVFs. Endovascular
therapy was performed on the occlusion site. After the passage of a 0.014-inch guidewire, intravascular ultrasound revealed no DVT, with the left common iliac vein compressed by the iliac artery and sacrum, suggesting MTS. Balloon angioplasty was performed, and anterograde blood flow and sufficient acute gain were obtained (Figure 2(d)). The pressure of the external iliac vein was reduced from 42/38 mm Hg to 12/10 mm Hg. After the procedure, echocardiograms showed no pulmonary hypertension.

One year follow-up echocardiograms showed mild restenosis of the common iliac vein, but she had no symptoms (Figures 1(c) and (d)).

**Discussion**

MTS is a condition in which the left common iliac vein compresses by the right common iliac artery and bony structures, which may cause swelling or DVT of the left lower extremity. This initiates a localized focal stenosis of the left common iliac vein at the level of the crossover position of the right common iliac artery, with subsequent venous outflow obstruction/stenosis and venous hypertension in the ipsilateral limb. This syndrome was shown to occur most frequently between the second and fourth decade of life, more often in women, with a prevalence of 2% to 5% in the general population. The syndrome may cause swelling and DVT. Treatment for MTS usually involves clearing the thrombus and correcting the compression of the left iliac vein. Endovascular therapy with thrombolysis and stenting is considered the first-line treatment for MTS, because patients with extrinsic causes of obstruction usually tend to respond poorly to balloon angioplasty alone. Pogorzeski et al. also reported that endovascular therapy should be considered as a good method for MTS, which prevents severe disabilment in the patient, and stent placement for MTS resulted in 2-year long-term patency. The cause of AVFs is most likely to be an aortic aneurysm of traumatic, congenital, or iatrogenic origin. AVFs can present with concomitant venous dilation. Venous dilation is likely to increase inflow from the arterial portion of the AVFs, leading to venous hypertension that causes weakening and subsequent stretching of the vein wall.

MTS coexisting with left iliac AVFs is rare. In our case, there was no aneurysm, catheter insertion, or trauma history. At the time of onset, the patient was being treated at another hospital, and the exact cause was unclear. We speculated two possibilities: one possibility is that secondary AVFs were

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**Figure 1.** Venous duplex Doppler examination: (a) The blood flow shows an arterial flow pattern. (b) The white arrow indicates arteriovenous fistulas. (c) The blood flow shows an arterial flow pattern again. (d) The white arrow indicates mild stenosis in the common iliac vein.
created by thrombus formation in the common iliac vein due to MTS, and the other possibility is that MTS and AVFs were originally present and the symptoms worsened as MTS progressed. It is uncertain which possibility is more applicable in this case. AVFs can aggravate the symptoms of MTS causing arterial blood to flow to the interrupted venous blood. Furthermore, the AVFs in our case might have inhibited antegrade venous blood flow by the Venturi effect and worsened edema of the lower extremities.

Haijie et al. treated similar case of ours, embolization resulted in decreased venous pressure, and opening and stenting in the occlusion of iliac vein further reduced venous flow resistance. We considered embolization for treating the AVFs, but treating all the AVFs seemed to be difficult. Coil embolization is contraindicated because new collateral circulation might be formed, and N-butyl-2-cyanoacrylate embolization may pass through the shunt and flow further. In addition, the development of collateral circulation may lead to pelvic congestion syndrome and need to pay attention. In our case, plain old balloon angioplasty was performed for the iliac vein. Stent implantation was considered in the case of insufficient dilation; however, it was not performed because good expansion was obtained by plain old balloon angioplasty alone and stent implantation for a vein is not covered by insurance in Japan. If the iliac vein alone was treated and AVFs were not treated, pulmonary hypertension could have developed. Fortunately, no complications occurred, and good patency was obtained without progress of pulmonary hypertension. Endovascular therapy for MTS has recently become more popular. Huynh et al. and Raju et al. demonstrated that iliac vein stenting was safe and effective in a group of elderly patients aged 80–96 years. In our case, a good outcome was achieved 1 year after the treatment, but if the symptoms reappear, it will be necessary to consider stent implantation and pay attention to pulmonary hypertension.

**Conclusion**

We described a case of MTS coexisting with AVFs treated by endovascular therapy. Angioplasty for the iliac vein alone may result in improvement of the symptoms.

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