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Case Report

Endovascular strategy for deep vein thrombosis caused by a huge uterine myoma accompanied by May–Thurner syndrome: A case report ☆,☆☆

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ABSTRACT

We report the case of a 71-year-old female presenting with deep vein thrombosis (DVT) of the left lower extremity secondary to a huge uterine myoma, who was successfully managed by hysterectomy and staged endovascular treatment. Her DVT was caused by left common iliac vein compression as a result of both the huge uterine myoma and preexisting May–Thuner syndrome. Although reported to put patients at high risk for DVT, coexisting large uterine myomas and May–Thurner syndrome are considered extremely rare.

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Introduction

Uterine myoma is a common gynecological disease that rarely causes deep vein thrombosis (DVT). However, patients with huge uterine myomas weighing over 1000 g are reportedly at high risk for DVT. As a risk factor for pulmonary embolism (PE), which is a life-threatening condition, the presence of DVT warrants the need to take preventative measures such as intra- or post-operative use of compression stockings and initiation of appropriate anticoagulant therapy (AT).

On the other hand, May–Thurner syndrome, characterized by the pathological compression of the left common iliac vein by the overlying right common iliac artery, is generally known as the anatomical cause of DVT. Recently, endovascular treatment with venous stents has reportedly proven effective for DVTs caused by May–Thurner syndrome.

Here, we report the case of a DVT secondary to a huge uterine myoma coexisting with May–Thurner syndrome, which was successfully managed by hysterectomy and staged endovascular treatment.

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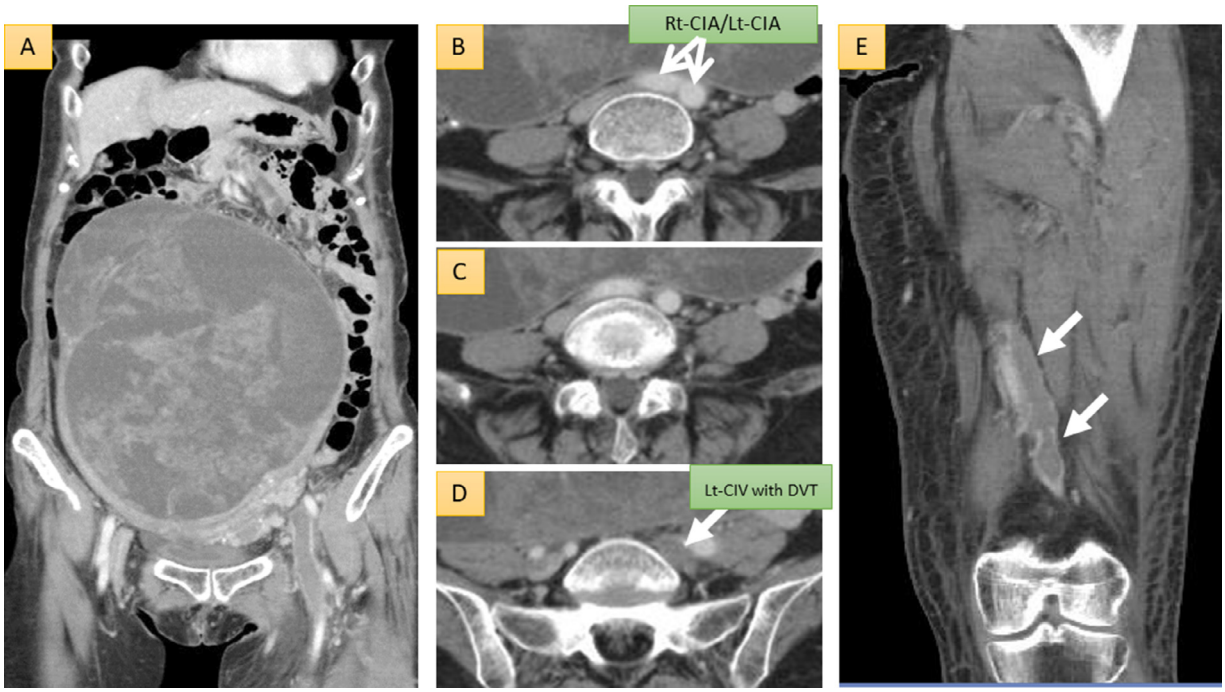

Fig. 1 – Enhanced computed tomography on admission. (A) Coronal view of the whole abdomen, (B-D) axial views, demonstrating the common iliac vein and artery at the level of the fifth lumbar vertebra as well as an occlusive thrombosis of the left common iliac vein (white arrow), and (E) coronal view of the left thigh, revealing an occlusive thrombosis of the left superficial femoral vein (white arrow).

Case report

A 71-year-old female was transferred to the emergency department of our hospital because of a painful, swollen left leg and severely distended abdomen which had been gradually worsening in the last 6 months. She reported no history of smoking, drug abuse, recent air travel, or familial thrombotic disorders.

On admission, her height and weight were 153 cm and 56 kg, respectively. She had a distended abdomen almost the size of a 30-week pregnant woman's belly and severe edema in the left leg. Her vital signs indicated a blood pressure of 155/105 mmHg, heart rate of 108 beats per minute, respiratory rate of 15 breaths per minute, and room air oxygen saturation of 97%. Electrocardiogram revealed sinus tachycardia without ST-T abnormalities. Chest X-ray showed no cardiac enlargement or abnormal shadow in the lung fields. Laboratory findings were remarkable for mild anemia (serum hemoglobin, 9.9 g/dL) and an elevated D-dimer level (30.6 pg/μL). Additional coagulation examinations demonstrated normal levels of protein C and protein S activity/antigen and anticardiolipin antibody.

Transthoracic echocardiography displayed normal findings except the collapse of inferior vena cava (IVC) compressed by a pelvic tumor. Duplex ultrasonography and enhanced computed tomography (CT) showed firm compression of the left common iliac vein by the pelvic tumor and thrombotic occlusion of the left iliofemoral venous system (Fig. 1). After PE was ruled out by enhanced CT, she was referred to an attending gynecologist and was subsequently diagnosed with a huge uterine myoma measuring 20.4 × 23.5 cm.

Based on the diagnosis of DVT caused by a huge uterine myoma, we started continuous intravenous infusion of heparin (15,000 units/day) and used elastic compression stockings for both her lower extremities since the day of admission. Four days after her admission, a total hysterectomy with bilateral salpingo-oophorectomy was performed. To avoid PE during hysterectomy, we placed a temporary filter (Neuhaus Protect SE; Toray Medical, Tokyo, Japan) in the IVC at the suprarenal level via a right jugular vein approach because the infrarenal IVC was completely compressed by the huge uterine myoma (Fig. 2A and B). The huge uterine myoma, measuring 23 × 22 × 8.5 cm and weighing 2060 g, was completely excised without any major complications.

On the day after surgery, the temporary IVC filter was removed and replaced with a new retrievable one (DENALI; Medicon, Osaka, Japan) at the infrarenal level via a right femoral vein approach (Fig. 2C and D).

From postoperative day (POD) 5, oral apixaban 5 mg twice daily was added to the initial heparin regimen; however, the left leg edema did not improve, and the level of D-dimer remained high (20.6 pg/μL). Enhanced CT on POD 11 showed the IVC was widely patent, whereas the proximal segment of the left common iliac vein was totally compressed by the right common iliac artery against the 5th lumbar vertebra, leading to occlusive thrombus formation in the left iliofemoral venous system (Fig. 3).

On POD 17, we performed endovascular treatment for the DVT of her left leg. In doing so, we punctured the left popliteal

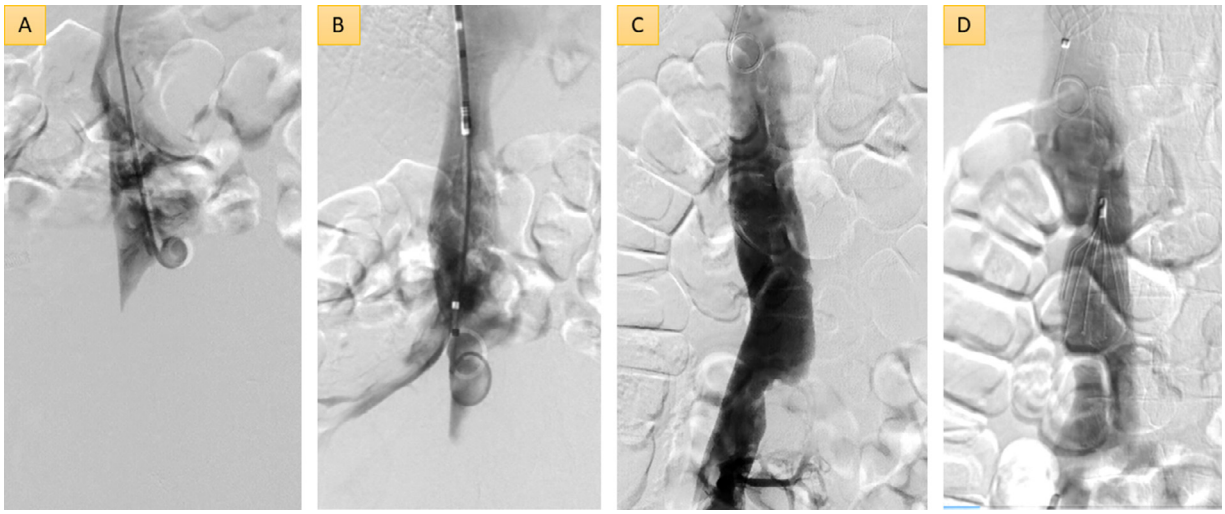

Fig. 2 – Venography of the inferior vena cava (IVC). (A) Before and (B) after insertion of a suprarenal IVC filter via the right jugular vein, (C) after hysterectomy (the IVC appears widely patent), and (D) after placement of an infrarenal IVC filter via the right femoral vein. IVC, inferior vena cava.

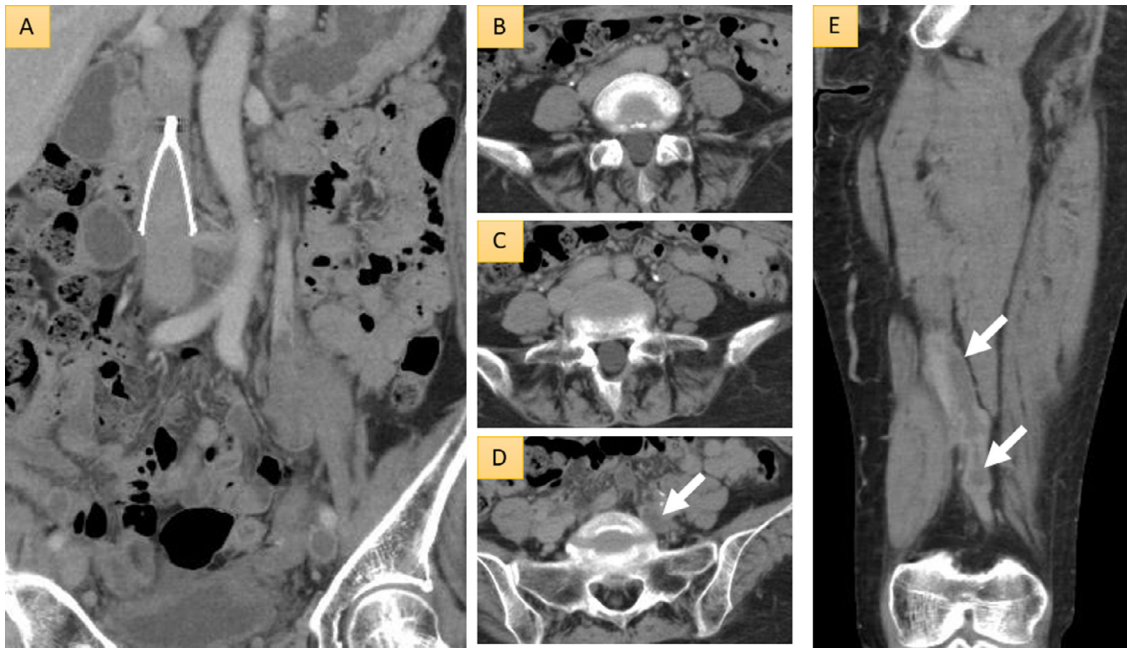

Fig. 3 – Enhanced computed tomography on postoperative day 11. (A) Coronal view of the abdomen, focusing on the abdominal aorta and inferior vena cava, (B-D) axial views, showing the common iliac vein and artery at the level of the fifth lumbar vertebra along with an occlusive thrombosis of the left common iliac vein (white arrow), and (E) coronal view of the left thigh, indicating an occlusive thrombosis of the left superficial femoral vein (white arrow).

vein under ultrasound guidance and inserted a 4 Fr sheath of 25 cm length (Medikit Co, Ltd, Tokyo, Japan) in it. Then, a 0.035-inch guidewire (Radifocus; Terumo Corporation, Tokyo, Japan) supported by a 4 Fr straight catheter 70 cm long (Medikit Co, Ltd, Tokyo, Japan) was carefully advanced into the IVC. After that, we exchanged the guidewire for a 0.014 inch one (Jupiter FC; Boston Scientific Japan, Tokyo, Japan) and dilated the occluded lesion of the left iliofemoral venous system using a 4 × 300 mm balloon catheter (Coyote; Boston Scientific Japan, Tokyo, Japan). Venography after balloon angioplasty revealed blood reflow from the left superficial femoral vein (SFV) into the IVC along with a lot of residual thrombus in the iliofemoral venous system. Therefore, a 4 Fr pulse-spray catheter with an infusion length of 40 cm (Fountain Infusion System; Merit Medical Japan, Tokyo, Japan) extending from the IVC to the left SFV was kept in place to pulse-infuse 60,000 units of

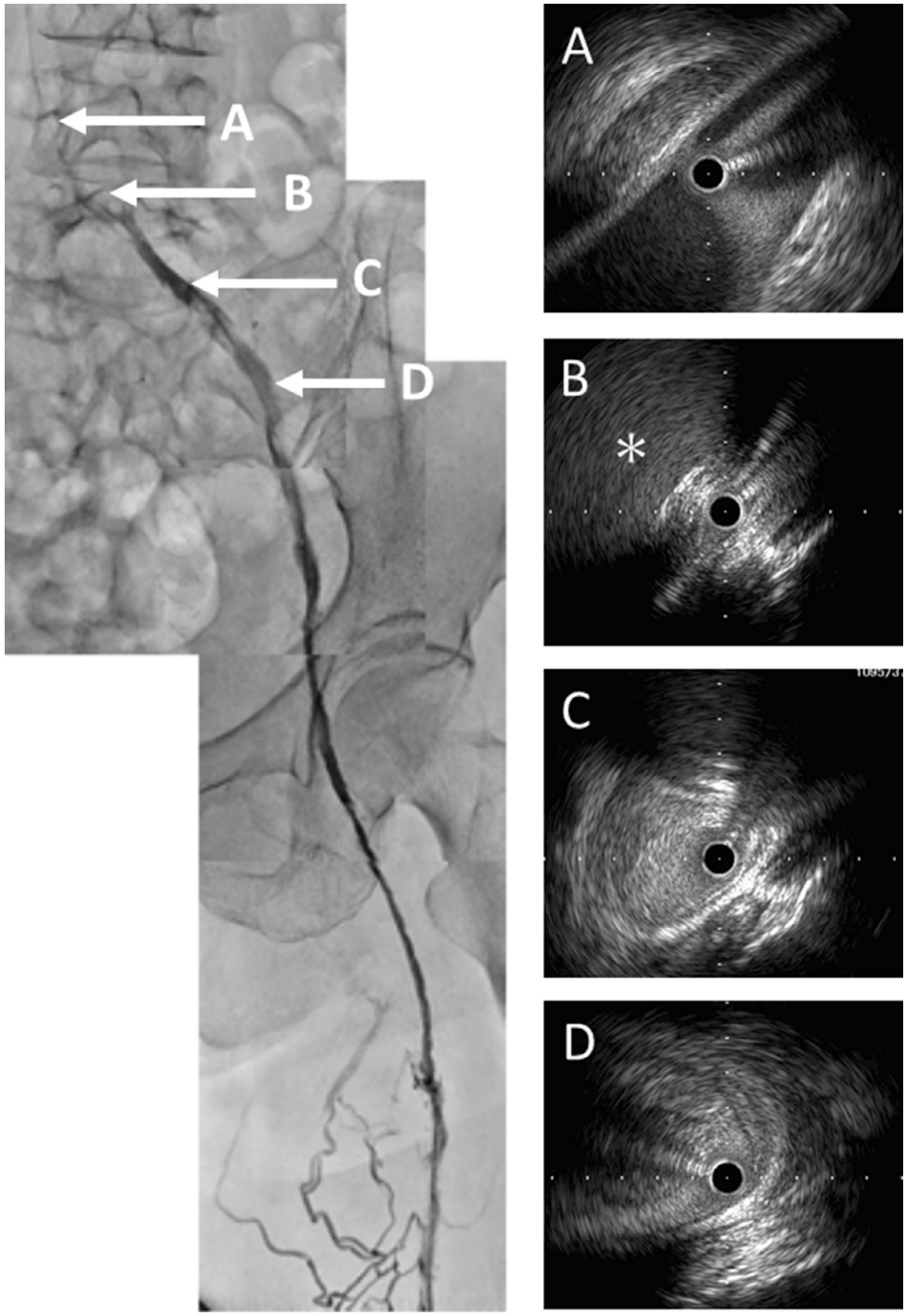

Fig. 4 – Venography and IVUS findings in the second session of endovascular treatment. IVUS images were indicates the right common iliac artery compressing the left common iliac vein. IVUS, intravascular ultrasonography.

urokinase every 3 hours for 2 days until reaching a total of 960,000 units.

As shown in Figure 4, venography and intravascular ultrasonography (IVUS) after continuous direct thrombolysis revealed resolution of most of the thrombus in the iliofemoral venous system, whereas the proximal left common iliac vein turned out to be still under firm compression by the right common iliac artery. Hence, May–Thurner syndrome was identified by both intravascular ultrasonography and CT findings as the underlying mechanism for her DVT. Subsequently, we deployed an 8 × 40 mm balloon catheter (Senri; Terumo Medical Corporation, Tokyo, Japan) to dilate the stenotic segment of the left common iliac vein, ending up with catheter recoil (Fig. 5A). Therefore, a 12 × 60 mm self-expanding nitinol stent (Epic; Boston Scientific Japan, Tokyo, Japan) was implanted in the left common iliac vein, with its tip extending slightly to the

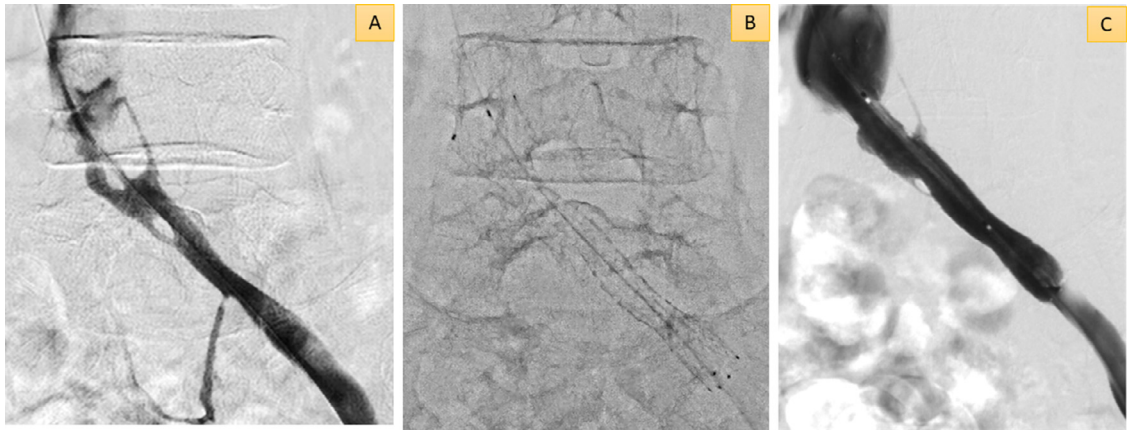

Fig. 5 – Stent implantation in the stenotic segment of the left common iliac vein after continuous direct thrombolysis. Digital subtraction venograms (A) before and (C) after implanting an EPIC stent (12 × 60 mm) in the left common iliac vein (B).

IVC (Fig. 5B). Final venography confirmed complete patency of the left common iliac vein and good blood reflow from the left SFV into the IVC (Fig. 5C).

Clopidogrel sulfate 75 mg/day was added to the treatment regimen after stent implantation. Her left leg edema rapidly improved, and D-dimer level reduced to 2 pg/μL on the day following stent placement. Finally, she was discharged without any complications on POD 26, and the IVC filter was removed on POD 56. Although AT with clopidogrel sulfate 75 mg/day was discontinued 2 months after discharge, she has been maintained on apixaban 5 mg/day up to date. At 2 years of follow-up, the left common iliac vein stent was patent with no evidence of recurrent DVT.

Discussion

The incidence of DVT after gynecological surgery is relatively high, ranging reportedly between 17% and 40% [1]. However, preoperative DVT occurs at a rather low rate of around 3%, especially secondary to malignant tumors [2]. Shiota M. et al have reported that the prevalence of preoperative DVT is 3.8-fold higher in patients with a uterine weight of over 1000 g and that the size of uterine myomas is a good indicator of preoperative DVT [3]. The case presented here had a huge uterine myoma of 2060 g that was found to firmly compress the IVC, causing poor venous blood flow and subsequent blood stasis followed by DVT.

There are no explicit guidelines for the treatment of huge uterine myomas complicated by DVTs, but it is generally recommended that hysterectomy be performed prior to aggressive treatment for DVT. To prevent perioperative PE, prophylactic AT and compression stockings are routinely used, with IVC filters opted for on a case-by-case basis. In a literature search of studies on huge uterine myomas complicated by DVTs, Satii et al reported that IVC filters were used in 15 of 27 cases undergoing hysterectomy, including their own cases [4]. In the current case, the compression of the IVC and left common iliac vein was released following excision of the huge uterine myoma, exposing the patient to the risk of developing PE as a result of the extensive thrombosis involving the left iliofemoral venous system. In addition, AT had to be discontinued 24 hours before and after surgery to prevent surgery-related bleeding. Therefore, we decided to implant an IVC filter. Nevertheless, an IVC filter could not be placed below the renal vein level since the IVC lumen was totally obstructed by pressure from the huge uterine myoma. Accordingly, a temporary IVC filter was placed at the suprarenal level just before surgery and was subsequently replaced with a retrievable one at the infrarenal level on the day following surgery.

Initially, we thought that her left leg DVT would improve once the compression of the IVC and left common iliac vein was relieved by hysterectomy, but in fact, it did not despite augmented AT. CT on POD 11 showed that the left common iliac vein was still compressed by the right common iliac artery even after performing uterine myomectomy, which led us to consider the presence of May–Thurner syndrome.

May–Thurner syndrome was first described by May and Thurner as a phenomenon in which chronic pulsations of the overlying right common iliac artery lead to spur formation along the wall of the left common iliac vein [5]. The exact prevalence of May–Thurner syndrome is unknown, but it is not an uncommon condition, reported in 14%-22% of autopsy cases. Moreover, one-third of patients with hemodynamically significant vein compression due to May–Thurner syndrome have been reported to be asymptomatic [6,7]. In this case, the mechanism responsible for DVT was thought to involve venous stasis induced by the huge uterine myoma in addition to May–Thurner syndrome, given the fact that there was no DVT in her right leg.

The treatment of iliofemoral thrombotic lesions secondary to May–Thurner syndrome has recently been facilitated by the development of endovascular treatment options including continuous direct thrombolysis, balloon angioplasty, mechanical thrombectomy, and stent placement [8]. Compared to AT alone, the combination of endovascular treatment and AT

has been reported to decrease the risk of recurrent thrombosis and/or severe post-thrombotic syndrome (PTS).

In a study by Xue et al, continuous direct thrombolysis followed by stent placement achieved good patency rates and enhanced PTS [9]. Yet, continuous direct thrombolysis raises the risk of bleeding and thus should be undertaken cautiously, especially in post-surgical patients such as the one presented here. On POD 17, continuous direct thrombolysis was initiated after the attending gynecologist determined that the risk of surgical site bleeding was low for urokinase, which was injected at a dose of 60,000 units every 3 hours through a pulse-spray catheter so that its administration could be stopped in the event of bleeding; fortunately, however, no vaginal or other surgery-related bleeding was observed during thrombolysis.

Although continuous direct thrombolysis for 2 days reduced the iliofemoral thrombus to less than 50% and restored blood flow through occluded veins into the IVC, the proximal left common iliac vein remained severely stenotic and was consequently stented. It should be noted that there is a lack of consensus on stenting for May–Thurner syndrome. Furthermore, left iliac vein compression is not necessarily pathological and has been reported to be present in 25% of the adult population, rarely leading to thrombosis. Yet, it is worth mentioning that although stent implantation simply because the left common iliac vein is stenotic should be avoided, Che et al demonstrated that stenting contributed to prolonged patency when the iliac venous pressure gradient across the area of compression was greater than 2 mmHg after continuous direct thrombolysis [10]. In this case, even after dilatation with an 8 × 40 mm balloon catheter, the pressure gradient remained 16 mm Hg.

Post-stenting AT for May–Thurner syndrome has not been established yet, but warfarin therapy with an international normalized ratio of 2-2.5 for at least 6 months—and even for a lifetime in some cases—has often been reported. With respect to antiplatelet agents, despite scant reports of 12 months of dual antiplatelet therapy, not many studies have directed considerable attention to them. One-year stent patency rate has been reported to range from 60% to 96% with a variety of ATs. In the present case, lifelong apixaban therapy was combined with a 2-month regimen of clopidogrel sulfate. Both the left iliofemoral venous system and common iliac vein stent have remained patent so far.

To the best of our knowledge, there is only 1 study—conducted by Barnaby J et al—reporting a case of a DVT complicated by a huge uterine myoma and May–Thurner syndrome. We believe since May–Thurner syndrome is not necessarily an uncommon condition, it might be masked or latently present in patients with left leg DVTs caused by huge uterine myomas [11].

Conclusion

We reported an extensive left leg DVT in a patient with a huge uterine myoma and May–Thurner syndrome, who received successful gynecological surgery and staged endovascular treatment.

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