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Case Report

Giant subserosal myoma causing deep venous thrombosis in a patient with pre-existing May-Thurner syndrome[☆]

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ARTICLE INFO

Article history:

Received 23 November 2019

Revised 3 February 2020

Accepted 14 February 2020

Keywords:

May-Thurner syndrome
Deep venous Thrombosis
Pelvic mass
ATTRACT Trial
Endovascular

ABSTRACT

One of the rare sequelae of large pelvic masses is direct compression of the inferior vena cava with formation of a deep venous thrombosis (DVT). Although uncommon, multiple cases of thrombosis secondary to pelvic mass compression of the venous system have been reported in the literature. However, our patient showed a disproportionate degree of thrombus and subsequent postthrombotic stricture/stenosis limited to the left iliofemoral system, sparing the right side. These findings make it exceedingly likely that she had some degree of pre-existing May-Thurner syndrome. The superimposed nature of these 2 rare causes of DVT make this presentation remarkably unique. Our case illustrates the advances in endovascular techniques and their application toward DVT treatment for even the most complicated and unique cases.

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Case report

A 37-year-old previously healthy female presented to the emergency department with new-onset shortness of breath and difficulty breathing. Chest x-ray and computed tomography (CT)-pulmonary angiogram were unremarkable for cardiopulmonary pathology; however, the CT revealed a 20-cm abdominal mass extending from the upper pelvis to the upper abdomen, abutting the inferior surface of the liver. A pelvic magnetic resonance imaging demonstrated a 21 × 18 × 12

cm mass, most compatible with a giant subserosal myoma (Fig. 1A and B). The mass was predominantly located in the patient's right hemi-abdomen and pelvis. The patient was scheduled for fibroid resection with the gynecology service. One week prior to her scheduled myomectomy, the patient presented with a tense, swollen, and painful left leg. A lower extremity ultrasound demonstrated left common femoral vein (CFV) and proximal/mid femoral deep venous thrombosis (DVT) with preserved patency of the distal femoral and popliteal veins. Anticoagulation was initiated with enoxaparin.

[☆] Declaration of Competing Interest: All authors have declared that no competing interests exist.

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<https://doi.org/10.1016/j.radcr.2020.02.010>

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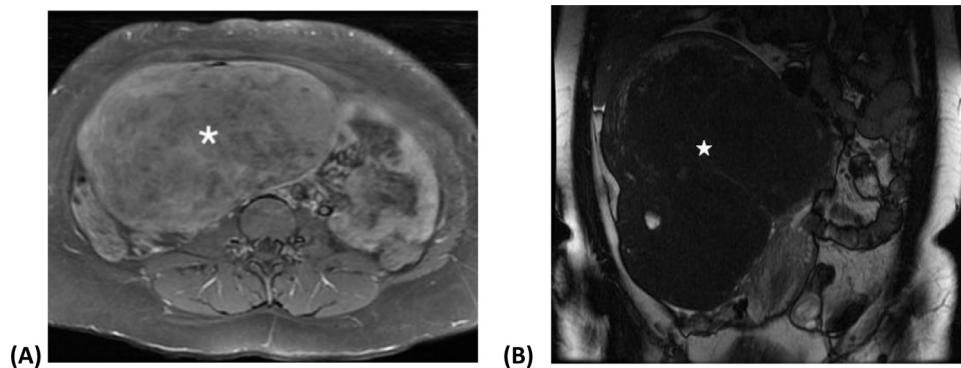


Fig. 1 – Axial and coronal magnetic resonance imaging demonstrating a large pelvic mass measuring 21 x 18 x 12 cm (white asterisk and star).

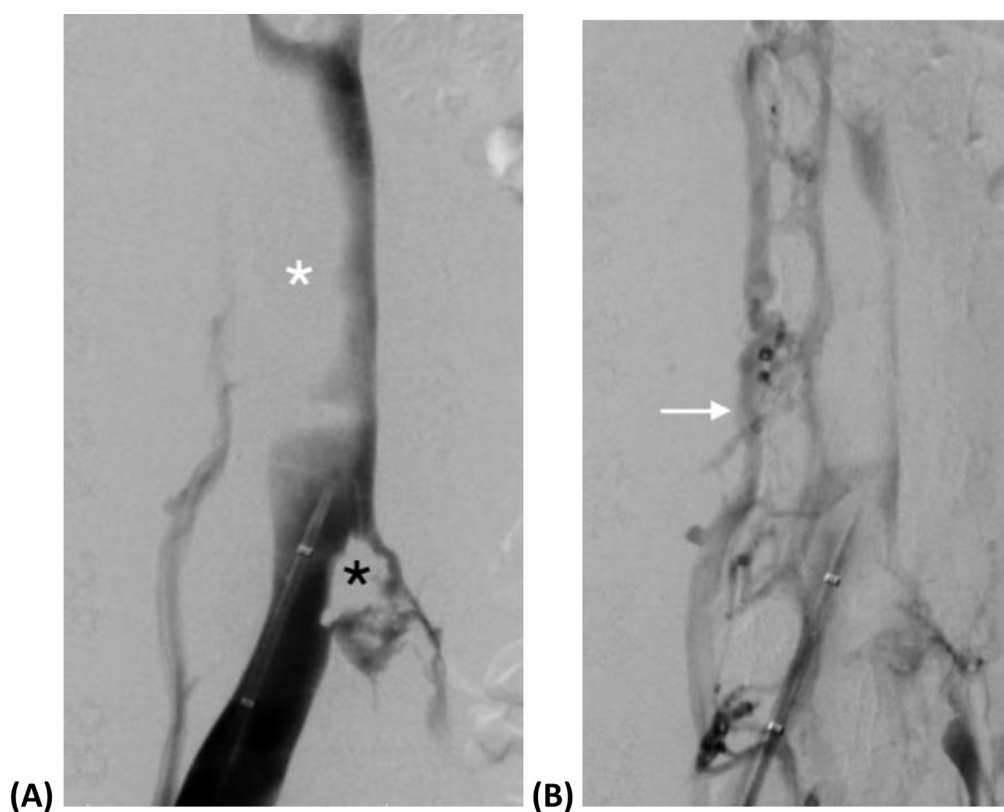


Fig. 2 – Venogram during unsuccessful attempt to place IVC filter via right femoral approach (in supine position). (A) Left CIV filling defects resemble clot (black asterisk). Eccentric IVC filling defect likely from extrinsic antero-posterior compression by the moreso right-sided uterine mass (white asterisk). (B) Collateralization into the ascending lumbar veins noted on later images (white arrow).

The patient did not have any risk factors predisposing her to clot formation and a hypercoagulable workup was negative. Her only physical finding was a palpable abdominal mass in addition to her swollen left lower extremity.

In preparation for the fibroid resection, an attempt to place an inferior vena cava (IVC) filter was made via a right femoral approach but was unsuccessful. The IVC at and below the level of T12 appeared largely compressed by the uterine mass (Fig 2) and/or thrombosed. Subsequently, a retrievable Cook Celect™ Platinum IVC filter (Cook Medical,

Bloomington, IN) was placed in a suprarenal position via a jugular approach.

The following day, the patient underwent pharmacomechanical thrombectomy using the Angiojet™ Zelante DVT System (Boston Scientific, Marlborough, MA) with subsequent balloon venoplasty of the IVC and left common iliac vein (CIV), external iliac vein (EIV) and common/proximal/mid femoral veins through a left popliteal approach using a 10-mm Mustang™ balloon (Boston Scientific, Marlborough, MA). Postthrombectomy venography demonstrated some residual



Fig. 3 – Postthrombectomy/thrombolysis venogram of pelvis (via left popliteal approach in prone position) which was performed prior to fibroid resection. Persistent outflow obstruction of bilateral iliac veins due to persistent IVC compression. Additionally, there appears to be residual stenosis of the proximal left CIV resulting in crosspelvic collaterals draining into the right iliac system and paralumbar collaterals from both sides. Venoplasty of this segment was then performed. (No preintervention images are included as the vein was markedly occluded and gentle venogram did not show any contrast passage.)

clot in the iliac/femoral veins and persistent extrinsic compression of the IVC (Fig. 3). Of note, this procedure was undertaken prior to resection for fear of clot embolization despite filter presence, especially given the IVC could not be definitely cleared of thrombus involvement.

The patient successfully underwent a laparotomy with myomectomy the following day and pathology was consistent with a leiomyoma (weight 1900 g). Anticoagulation was initiated postoperatively.

On postoperative day 4, an abdominopelvic CT with venogram of the lower extremities showed residual thrombus within the left iliac and femoral veins; the IVC appeared to be patent. Postoperatively her left leg remained slightly

larger than her right; however, it was nontender and no additional signs of postthrombotic syndrome (PTS) were evident. She was continued on enoxaparin 100 mg daily and bilateral compression stockings remained in place. Bilateral lower extremity ultrasound 4 weeks after surgery revealed no residual thrombus in the left lower extremity; but, showed a dampened waveform suggesting downstream obstruction.

Three months later, through a left CFV puncture an iliofemoral venogram and inferior vena cavogram were performed. Occlusion of the left CIV and EIV, tortuosity of the femoral veins, as well as extensive crosspelvic and paralumbar collaterals were noted (Fig. 4). The IVC and right iliac system remained patent. All the lesions were crossed and venoplasty of the left CIV and EIV and proximal CFV were performed using a 10-mm Mustang™ balloon (Boston Scientific, Marlborough, MA). A venogram demonstrated improved patency of the iliofemoral system, yet stenosis at the proximal CIV and prominent collaterals remained (Fig. 5A). A 16 mm × 90 mm Wallstent™ (Boston Scientific, Marlborough, MA) was deployed in the left CIV and EIV. Given incomplete response with persistent collaterals, the distal EIV and CFV were also stented using two 10 mm × 40 mm Epic™ Vascular stents (Boston Scientific, Marlborough, MA). Venography demonstrated markedly improved flow throughout the left iliofemoral venous system without flow into the collaterals (Fig. 5B). A follow-up bilateral lower extremity duplex scan was performed 4 weeks later showing no evidence of DVT within the bilateral femoral-popliteal system. The IVC filter was subsequently removed.

The patient was seen most recently at a follow-up visit 18 months after the index event; she continues to do well with no recurrence of symptoms or signs of PTS.

Discussion

DVT formation from direct compression of the venous system is a rare complication of a pelvic mass. Although uncommon, multiple cases of thrombosis secondary to pelvic mass compression of the venous system have been reported in the literature. However, after conducting a literature search, we found no reported cases of a pelvic mass with underlying May-Thurner syndrome (MTS) causing thrombosis.

One of the most worrisome complications of an untreated DVT is PTS, which can be a significant source of morbidity, decreasing the patient's quality of life and increasing the economic burden of treatment substantially (cost to healthcare system is an estimated \$7000 annually per patient) [1,2]. The main symptoms of PTS are chronic pain, edema, swelling, skin changes (including venous ulcerations), and a perceived heaviness of the affected limb [3]. These symptoms arise secondary to a prior DVT event, which damages the valves and causes obstruction to venous outflow. The incidence of DVT is thought to be 1 in 1000 [1,4] people per year, and studies have shown that 20% to 50% of those patients will go on to develop symptoms of PTS [3,4]. In our case, the primary goals were prompt removal of the compressive pelvic mass and treatment of the thrombus in order to prevent the dreaded complications of PTS.

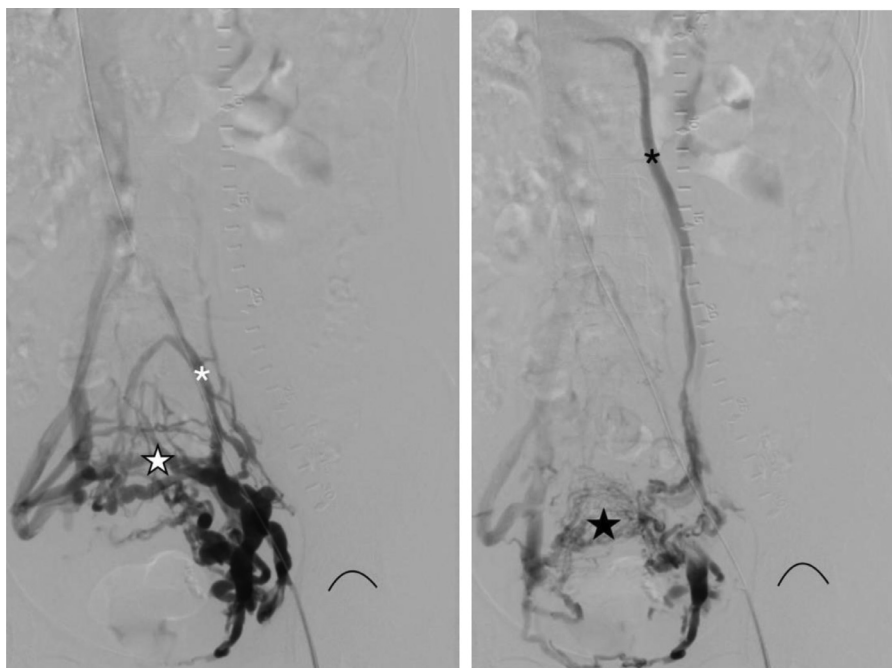


Fig. 4 - Venogram via left femoral approach three months postthrombectomy/venoplasty. There is interval worsened stricture of the left CIV and EIV, which are not seen (delineated by wire silhouette) with markedly hypertrophied collaterals. (presacral = white star; paralumbar = white asterisk, parametrial = black star; gonadal = black asterisk; femoral head = black curvilinear line).

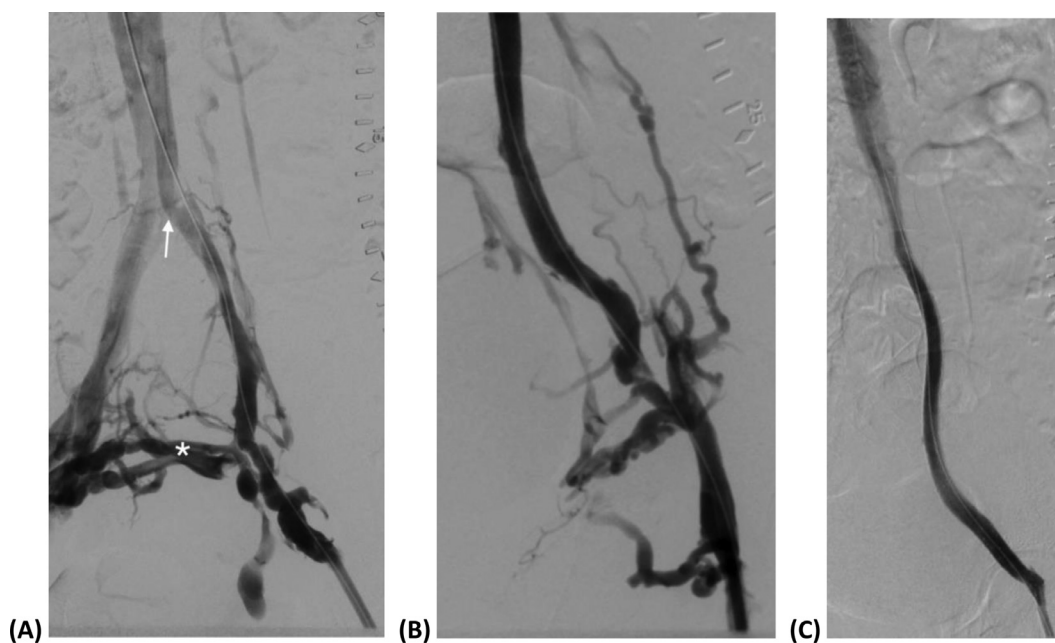


Fig. 5 - Postangioplasty image (A) shows improved patency through the EIV/CIV with some outflow into the IVC; however, stenotic areas remain, particularly at the proximal CIV (white arrow), with persistent collaterals (white asterisk) implying a significant outflow obstruction. Poststenting images (B and C) show improved outflow with persistent, albeit decreased, collateralization following the 16-mm CIV/proximal EIV stent and elimination of all collaterals following additional deployment of 10-mm distal EIV/CFV stents.

Uterine fibroids, benign tumors of the myometrium, have been known to cause DVT as one of their rare sequela. DVT under these conditions are thought to occur secondary to the leiomyoma growing large enough to exert direct pressure on the deep venous system (predominantly the IVC) leading to venous stasis. A study by Shiota et al [5] showed that DVT rates were significantly higher in patients with uterine weights of 1000 g or more (11.5%) compared to uterine weights less than 1000 g (3.0%). The surgical specimen in our case weighed 1900 g.

The vast majority of the reported cases of pelvic mass causing DVT were treated with hysterectomy and long-term anticoagulation. In our case, the pelvic mass was removed using myomectomy and the DVT was treated with pharmacomechanical thrombectomy, angioplasty, stenting, and long-term anticoagulation. Our main point of discussion would be whether the iliac vein should have been stented at the time of the pharmacomechanical thrombectomy. However, the presence of the uterine mass obstructing and/or compressing the IVC precluded this option.

MTS is classically defined as compression of the left CIV between the right common iliac artery and the fifth lumbar vertebrae, leading to focal intimal thickenings and the formation of venous spurs. It is thought that the pulsatile nature of the right iliac artery passing anterior to the left iliac vein leads to chronic injury with subsequent intimal fibrosis. May and Thurner [6] noted these spurs were present in 15% to 22% of cadavers on postmortem surveillance. They also noted that spurs were absent in fetal cadavers, showing this to be an acquired rather than a congenital condition.

As seen in Fig. 3, our patient had recalcitrant stenosis of the left iliac system. The high likelihood of pre-existing MTS also explains why the entire DVT was confined to the left iliofemoral system, with the right side remaining unaffected, even though the mass preferentially deviated rightward. If the thrombosis were only due to IVC compression, we would expect some thrombosis would extend into the right iliac system as well. Ideally, we should have performed intravascular ultrasound to prove the presence of venous spur [7]; however, it was unavailable at our institution. The classic situation of this stenosis leads us to strongly believe that this patient had some degree of pre-existing MTS.

Over the past 30 years, the scope of treatment for acute DVT has markedly expanded. The mainstay of treatment used to be anticoagulation and systemic thrombolysis and then progressed to surgical techniques to extract the thrombus. Most recently, minimally invasive endovascular techniques (catheter-directed thrombolysis, pharmacomechanical thrombectomy, angioplasty, and stenting) have become common practice. Endovascular techniques have been shown to be particularly effective in patients with underlying MTS or other causes of altered anatomy. Pharmacomechanical catheter-directed thrombolytic/thrombectomy (PCDT) rapidly removes thrombus and is hypothesized to reduce the risk of PTS; this has been called the “open-vein hypothesis [8].” Berger et al [9] were the first to describe the use of catheter-directed thrombolysis, angioplasty, and stenting for the treatment of proven MTS. Years later, Patel et al [10] demonstrated 100% initial success rate in 10 patients who had

proven MTS treated with catheter-directed thrombolysis and iliac vein stent placement; however, 2 patients went on to develop in-stent thrombosis. A study by Hager et al [11] showed that stenting for MTS was a safe and effective treatment with patency rates of 91% at 36 months. Other studies treating left-sided DVT with catheter-directed thrombolysis and stent placement have shown good early results with a mean technical success of 95% and patency rates of up to 96% at 1 year [12].

The landmark ATTRACT trial [13] (Acute Venous Thrombosis: Thrombus Removal with Adjunctive Catheter-Directed Thrombolysis) has reopened the debate on what is the best approach to treating patients with MTS in order to avoid developing PTS. The study followed 692 patients with proximal DVT, assigning them to one of 2 groups, the first receiving anticoagulation alone (control), the second receiving anticoagulation with the addition of pharmacomechanical thrombolysis. The goal of the trial was to assess the risk of developing PTS between 6 and 24 months. The trial showed that between 6 and 24 months there was no significant difference ($P = .56$) between the two groups. However, there was an increased risk of major bleeding events in the thrombolysis group (1.7% vs 0.3%, $P = .049$).

The ATTRACT trial has fallen under harsh criticism since its release as it failed to meet its primary endpoint. According to Marston [14], for the trial to be completed in reasonable time the design included cases of thrombosis in the femoropopliteal segments alone and those with more proximal iliac obstructions. There is known to be a higher risk of PTS in iliofemoral thrombosis; if the trial were to use these cases exclusively it may have had a greater chance of meeting the primary endpoint; however, as he noted it would have risked failing to meet goals set by the funding agency and the study's budget. The trial was also limited by a substantial number of missing assessments for PTS, patients lost to follow-up [13], and a short follow-up period of only 2 years.

In fact, the subgroup analysis of the ATTRACT trial did suggest that the PCDT group may show benefit for proximal thrombosis of the iliac vein and those patients presenting with more severe symptoms, allowing them to experience more rapid resolution.[13] The subgroup data also suggest that patients over 65 years of age are less likely to benefit from PCDT, compared to younger patients. However, the study does not have the power to draw definite conclusions from these subgroup analyses; leaving us with only one definite conclusion—more research is needed. This leaves us with a challenging and ever-evolving dilemma of whether to treat our acute DVT patients with PCDT or anticoagulation alone.

Our case predates the publication of the ATTRACT trial; nevertheless, we would not have changed our management of this patient.

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