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When Recurrent deep vein thrombosis is beyond only a clot- May-Thurner Syndrome

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Abstract

May-Thurner syndrome (MTS), also known as iliac vein compression syndrome which results in luminal narrowing and intimal hyperplasia, which predispose to venous stasis and recurrent deep vein thromboembolism (DVT) We present a 32-year-old woman with MTS who developed recurrent deep venous thromboses despite undergoing thrombectomy and being on recommended anticoagulation.

Case Report

When recurrent deep vein thrombosis is beyond only a clot- May-Thurner Syndrome

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Short Title: Recurrent DVT by a clot.

Key words : Deep vein thrombosis (DVT), May-Thurner syndrome (MTS), anticoagulation, Thrombosis.

Key clinical message:

Unidentified MTS carries a potential risk of recurrent DVT, pulmonary embolism, and chronic venous insufficiency. Early recognition for the high-risk group with typical features is crucial.

Abstract

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May-Thurner syndrome (MTS), also known as iliac vein compression syndrome which results in luminal narrowing and intimal hyperplasia, which predispose to venous stasis and recurrent deep vein thromboembolism (DVT)

We present a 32-year-old woman with MTS who developed recurrent deep venous thromboses despite undergoing thrombectomy and being on recommended anticoagulation.

Introduction

May-Thurner syndrome (MTS), also known as iliac vein compression syndrome, is an

anatomically variable condition in which the right common iliac artery overlies and subsequently compresses the left common iliac vein against the lower lumbar spine, commonly the fifth lumbar vertebra1. This results in luminal narrowing and intimal hyperplasia, which predispose to venous stasis and recurrent deep vein thromboembolisms (DVT). Definitive treatment in most cases is endovascular intervention followed by maintenance anticoagulation, which becomes all the more necessary because of the pro thrombotic effect of the intravascular device. Here we present a case of recurrent DVTs in a patient who was diagnosed with MTS despite undergoing definitive treatment (i.e., endovascular intervention followed by systemic anticoagulation). This case report emphasizes the need for proper guidelines on acute and long-term management of May-Thurner syndrome, especially post-stent thrombotic treatment in patients with other thrombophilic conditions.

Case Report/Case Presentation

A 32 -year old female with past medical history of hypothyroidism, gout, cutaneous polyarteritis nodosa and recurrent miscarriages presented to the emergency department (ED) with 1 week of progressive left lower extremity swelling The swelling was associated with purplish discoloration of her toes, along with a tingling sensation, especially around the calf. She denied any shortness of breath, chest pain or palpitations. Five weeks prior to her presentation, she had a complicated delivery due to placenta accreta where she underwent a Cesarean section. with a significant postpartum hemorrhage requiring 6 units of packed red blood cells and 2 units of fresh frozen plasma transfusions. The bleeding continued, necessitating emergent hysterectomy and bilateral salpingectomy. Due to surgical recovery and post-operative pain, she had minimal ambulation for the month prior to her presentation to the ED. Of note, given her recurrent miscarriages, she had had a prior workup for antiphospholipid syndrome, which was negative.

Assessment:

On exam, she was normotensive, afebrile, with a heart rate of 98. Lower extremities showed left leg warmth, swelling and tenderness up to the thigh with mild purplish discoloration over the toes along with livedo reticularis. Bilateral dorsalis pedis pulses were intact and equal with normal capillary refill. Ultrasound (US) revealed completely occlusive deep vein thrombosis extending from the common formal vein through the tibioperoneal trunk. Computed tomography venography (CTV) of lower extremity showed demonstrated an extensive DVT, extending from tibio-peroneal trunk through the common femoral vein and up to the origin of inferior vena cava (IVC) shown in Figure 1.

Management

Anticoagulation was initiated with intravenous (IV) heparin drip. Vascular surgery and

hematology was consulted, and she was deemed not fit for thrombolytics due to her recent pelvic surgery and significant bleed. Her course was complicated by sub-therapeutic activated partial thromboplastin time (APTT) despite aggressive efforts, necessitating a switch to argatroban. However, due to the unremitting left leg pain, she underwent mechanical thrombectomy and thrombolysis in the left popliteal, femoral and common femoral veins, with prophylactic intravenous vena cava filter placed prior to the procedure.

Notably, her angiogram during the procedure showed high grade stenosis in the distal left common iliac

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vein, indicative of May Thurner Syndrome. Hence, during the procedure, she received left iliac vein stenting to maintain patent venous outflow in the long term shown in Figure 2 . Post procedure she remained on argatroban drip and Tissue plasminogen activator (tPA) was initiated to maximize thrombolysis efforts. Repeat angiogram in less than 48 hours showed the common femoral vein, external iliac vein, common iliac vein to be widely patent. tPA was stopped after 48 hours and the patient was switched to subcutaneous fondaparinux 10mg daily. Three weeks later, she was switched to the oral anticoagulant (OAC) rivaroxaban. Despite being on rivaroxaban, a repeat US of her left leg 10 weeks later showed thrombosis in the previously patent left common femoral vein, extending into external and common left iliac veins with thrombosis of the left iliac vein. Repeat angiogram showed the new complete occlusion of the left common femoral vein as well as the common and external iliac veins. The thrombosis appeared chronic and sclerotic and stent recanalization was attempted but unsuccessful. She had intact pulses and did not have increased lower extremity swelling or any other sign of DVT so remained on anticoagulation with the plan for lifelong rivaroxaban therapy. Repeat US 4 months later showed residual left iliac vein stent thrombosis and nonocclusive thrombus in the left common femoral vein. Another repeat US at 6 months revealed a patent left common iliac vein suggestive of interval clot resolution but persistent left common femoral vein thrombus. IVC filter has remained in place for these past 12 months due to the fluctuating clinical course to ensure

safety.

Discussion/Conclusion

May-Thurner syndrome is a condition where patients develop compression of the iliocaval venous system by the arterial system on a bony structure causing disruption of venous blood flow. Many variants exist; the most commonly seen is compression of the left common iliac vein by the right common iliac artery against the lumbar vertebrae. This develops through intimal hypertrophy of the iliac vein wall secondary to mechanical compression and arterial pulsation. It creates potential stasis and subsequent thrombosis. As early as 1851, Virchow noted that left leg DVT is more common than right by 5 times2. Afterwards, in 1908, the anatomical variation was described and was linked to Virchow's finding3. However, MTS was not recognized until 1957 when May and Thurner described the compression of left iliac vein by the right iliac artery against lumbar spine causing thrombosis4. The exact incidence and prevalence of MTS remains largely unknown. Multiple reports suggested prevalence by 20-30%1,2,5. In general, it is a less reported cause of DVT. MTS can be challenging to recognize and requires a high index of suspicion, identifiable risk factors, and more invasive

testing to diagnose6. Interestingly, the mere presence of this anatomical finding might not predispose patients to DVT, unless combined with either the presence of hypercoagulability, endothelial injury and more stasis in conjunction (Virchow's triad) which would lead to thrombus formation2. Patient have various clinical presentations, ranging from asymptomatic to venous claudication and skin damage7. Current observational studies suggest MTS is more commonly seen in young females (60%) 8. Patients who develop DVT in this gender and age group are mostly on oral contraceptives or may have had recent pregnancy, or less commonly, prolonged immobilization1. These factors predispose the asymptomatic MTS patient to become symptomatic. Other risk factors include: scoliosis, which may predispose to MTS through compression from the lower lumbar vertebra; dehydration; and radiation exposure9,10,11. As such, signs of left sided DVT in high risk patients i.e., young and female, particularly with risk factors, should prompt extensive evaluation including a doppler US of the affected extremity. A retrospective analysis of 50 abdominal computed tomography (CT) scans of patients with abdominal pain but without lower extremity symptoms showed that 25% of the individuals had hemodynamically significant lesions causing at least 50% stenosis in the left common iliac vein while two-thirds had at least 25% compression2. Thus, this finding may represent a normal anatomic variant rather than a pathological condition2. Treatment of May-Thurner syndrome involves thrombolysis of the formed clot, stent placement to alleviate the anatomical compression stenosis, and long-term anticoagulation to prevent further thrombosis. In certain cases, thrombectomy could be also considered, especially in cases that thrombolysis is at great risk. Vena cava filters are almost always deployed to prevent pulmonary embolism from showering emboli during the procedure 2,12. The question that arises

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is the choice of anticoagulation (AC) and their duration. Recently a systematic review and meta-analysis concluded stent thrombosis or occlusion occurred in 10%-20% of post-stent placement patients at median of 12 months regardless of choice of antithrombotic management13. Thus, it was not able to suggest a standard type, dose or duration due to inconsistencies in the included studies and no availability of any prospective study comparing treatments/therapeutic approaches. Endo., *et al* suggest that after illiocaval venous stenting, stent patency is best predicted by combined antiplatelet and anticoagulation rather than antiplatelets alone14. There is a lack of data on postinterventional

medical therapy in endovascular therapy15. Although antiplatelet therapy is

frequently used, the duration is variable given the absence of practice guidelines15. The optimal approach with anticoagulation with or without antiplatelets still remains unknown and warrants further randomized control trials. Most DVTs that are associated with MTS can resolve with or without stent placement while the patient is on anticoagulation. Nevertheless, some patients develop extreme complications such as post-thrombotic syndrome, venous claudication, varicose vein formation, venous stasis ulceration and recurrent DVT16. For example, 36.8% of the female patients that were involved in one study developed post-phlebitis syndrome17. In these cases, like ours, complications occur even after

stent placement and in such cases venous bypass surgery remains as the last resort.

CONCLUSION:

May-Thurner Syndrome is an underdiagnosed but well-known risk factor for thromboembolic

events. Unidentified MTS carries a potential risk of recurrent DVT, pulmonary embolism and

chronic venous insufficiency. Early recognition for the high-risk group with typical features is

crucial. Endovascular intervention remains the mainstay of treatment. However, evidence on the best anticoagulation regimen to be used is still lacking. More randomized control trials on the guidance of anticoagulation versus antiplatelets are warranted.

Statements

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Statement of Ethics:

This article was approved by Medical Research Center in Qatar before submission to the journal.

Also Written informed consent was obtained from the patient for publication of this case report.

Conflict of Interest:

The authors declare that there is no conflict of interest.

Authors Contribution:

Hussam Alhasson took the lead in writing the manuscript, literature review as well as created the legends, wrote the manuscript and performed literature review. Pratyusha Tirumanisetty, Hussam Alhasson, Mohammad Abu-Tineh and Mustafa A. Al-Tikrity revised manuscript critically for important intellectual content.

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Hussam Alhasson and Pratyusha Tirumanisetty and all other co-authors took care of the patient, as well as contributed to and approved the final version of the manuscript.

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