

CASE REPORT : May-Thurner syndrome initially presenting lymphedema without DVT

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Introduction

Unilateral leg edema can be classified into acute or chronic condition. The acute onset of unilateral leg edema is usually due to deep vein thrombosis (DVT), musculoskeletal problem or cellulitis. In this paper, we report a case of acute onset unilateral lower leg edema due to cellulitis in a young female; further work-up revealed leg edema as a clinical manifestation of venolymphatic overloading by May-Thurner syndrome (MTS) without DVT.

CASE REPORT

A 15-year-old woman visited lymphedema clinic of other hospital, presenting sudden heating sensation, swelling, redness, pain in the left thigh. There was no specific history and no drug use. The body temperature was 37.5°C and initial blood test showed a slight increase of white blood cell count and definite increase of high-sensitivity C-reactive protein. No significant stenosis/occlusion of both lower extremity arteries and DVT were observed on femoral computed tomography (CT) angiography. The femoral CT angiography revealed reduced diameter of left common iliac vein (CIV). In previous hospital, she was clinically diagnosed as cellulitis and discharged after 7 days of IV antibiotics (cefazolin + clindamycin). After 1 month of discharge, she visited our lymphedema clinic, suffering from increased left thigh edema and worsening pain of left leg. She felt pain at weight bearing of left leg. The volume of the left lower limb was 8382ml, which was 1582ml (23.26%) more than the right side. We performed duplex scan and pelvic venography. In the duplex scan, DVT was not observed and abnormal backflow from external iliac vein to internal iliac vein was detected. The pelvic venography revealed compression of left CIV by the right common iliac artery (CIA). It was a nearly complete obstruction accompanied with multiple collateral vessels around the CIV. Additionally, a 4 mmHg pressure gradient was observed between the inferior vena cava and the left CIV. Such overall findings were suggestive of MTS. Lymphoscintigraphy showed ilioinguinal lymph nodes on both inguinal area and dermal backflow at the left upper thigh. The patient underwent two weeks of complex decongestive therapy. The volume of left leg was reduced to 8018ml, which indicated volume reduction of 364ml compared to the initial measurement. Pain of left leg gradually faded, as the volume of left leg declined.

Conclusion

The MTS is a compression of extrinsic venous system by arterial system. It leads to focal stenosis at intersection of left CIV and right CIA. This syndrome is often accompanied with

DVT. But we report a case of MTS initially presenting lymphedema and cellulitis without DVT. It is meaningful in that lymphatic overloading caused by CIV obstruction damages lymphatic systems, followed by lymphedema. When treating lymphedema patients, it is necessary to consider phlebolymphe-
dema, swelling due to chronic venous insufficiency and lymphatic insufficiency.

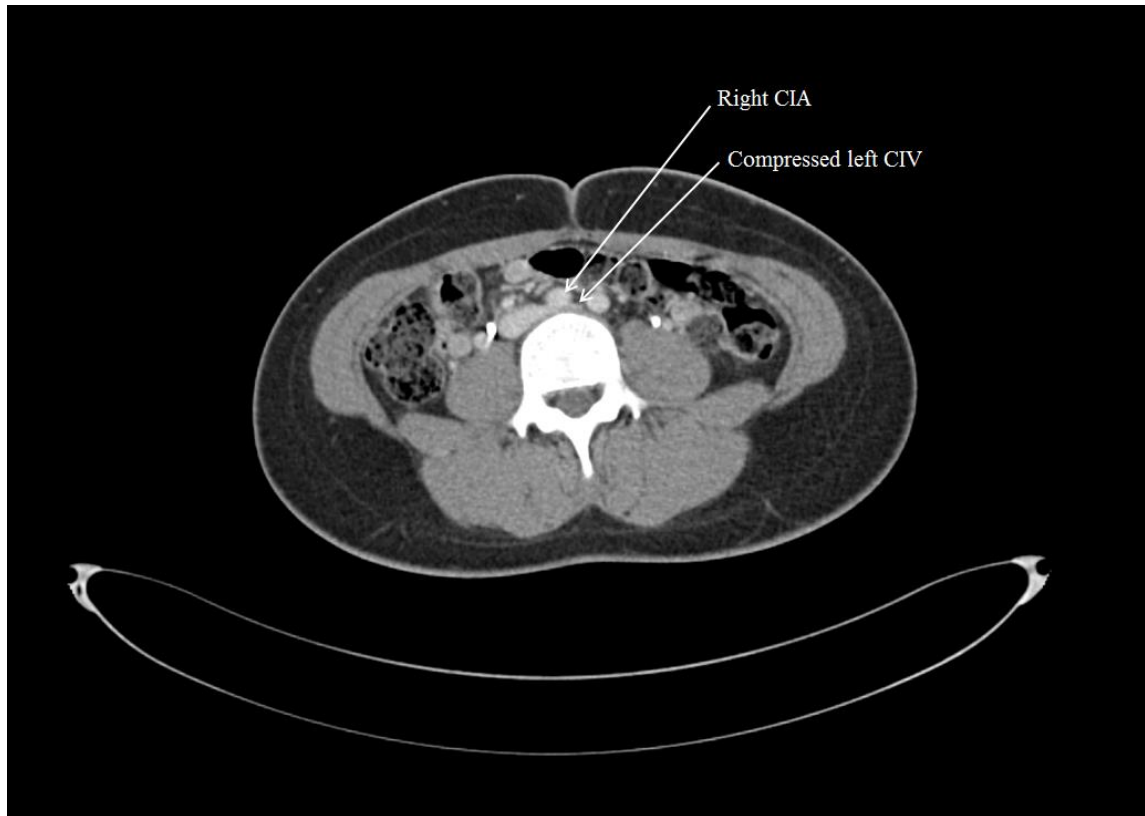
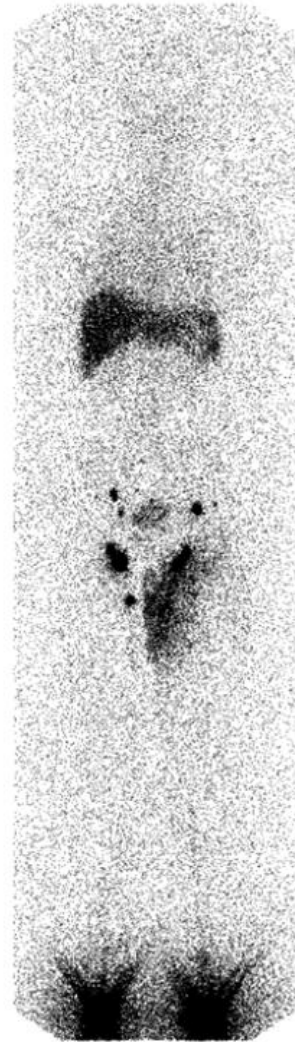


Figure 1. Diameter of left common iliac vein is reduced. Suspected of compressed by right common iliac artery. CIV : common iliac vein, CIA : common iliac artery



ANTERIOR 10MIN



ANTERIOR 60MIN

Figure 2. Dermal backflow was observed in the left upper thigh. ilioinguinal lymph nodes were observed on both sides.



Figure 3. Pelvic venography showed near complete obstruction and multiple collateral vessel development of the left common iliac vein.