

Available online at www.sciencedirect.com



journal homepage: www.elsevier.com/locate/radcr



Case report

Double May-Thurner syndrome causing chronic deep vein thrombosis and natural venous femoro-femoral bypass: a description of rare

case.☆,☆☆

Cecilia Gozzo, MD^{a,*}, Renato Farina, MD^a, Pietro Coppolino, MD^a, Giovanna Cancemi, MD^a, Pietro Valerio Foti, MD^a, Stefano Palmucci, MD^a, Massimo Venturini, MD^b, Antonio Basile, MD^b

^a Department of Medical and Surgical Sciences and Advanced Technologies "GF Ingrassia", Radio diagnostic and Radiotherapy Unit, Catania, Italy

^b Diagnostic and Interventional Radiology Department, Circolo Hospital, Insubria University, Varese, Italy

ARTICLE INFO

Article history: Received 28 March 2021 Revised 7 April 2021 Accepted 9 April 2021 Available online 29 April 2021

Keywords: May-thurner syndrome Doppler ultrasound Vascular compression Computed tomography Deep venous thrombosis

ABSTRACT

May-Thurner syndrome (MTS) belongs to a group of uncommon vascular syndromes. It consists in left common iliac vein (LCIV) compression between the right common iliac artery (RCIA) anteriorly and the lumbar spine posteriorly. A compression of LCIV by the left common iliac artery (LCIA) or by both iliac arteries were described. We present a rare case of "double MTS" which consist in double stenosis of LCIV by both RCIA and LCIA. Double MTS can cause acute or chronic DVT; this latter could be clinical manifest or well compensated.

A 58-year-old woman with chronic mild pelvic pain underwent Doppler Ultrasound (US) of the pelvis and lower extremity vessels which showed thrombosis of both LCIV and ipsilateral common femoral vein caused by the extrinsic compression by both common iliac arteries against the spine. CT angiography confirmed the US data and ruled out other causes of compression. CT scan also showed the development of a natural venous femoro-femoral bypass which allowed to counteract the venous stasis and compensate venous drainage.

Therefore, we decide for a long-term prophylaxis with anticoagulant drugs and doppler US follow-up at 6 months.

In conclusion, doppler US is a non-invasive, low-cost, repeatable and sensitive method which allows to diagnose MTS and associated DVT. It may be considered the first level exam which allows to easily detect pelvic vascular compression syndrome.

© 2021 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

 $^{\,\,^}st$ Competing interests: The authors declare that they have no conflict of interest.

 $^{^{}stlpha}$ Acknowledgment: This study was not supported by any funding.

Corresponding author.
E-mail address: ceciliagozzo91@gmail.com (C. Gozzo).

https://doi.org/10.1016/j.radcr.2021.04.020

^{1930-0433/© 2021} The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Table 1 – Summary of the results obtained by Ultrasound examination.						
Vascular structure	PVV (cm/s)		FR	Distance from vertebral soma (mm)	Diameter (mm)	1
LCIV	Pre-stenotic 6.9	Post-stenotic 18.5	2.6	-	Pre-stenotic 7	Post-stenotic 8
RCIV	26.8		-	-	12.2	
RCIA	-		-	4.4	-	
LCIA	-		-	6.2	-	

FR, flow-ratio; LCIA, left common iliac artery; LCIV, left common iliac vein; PVV, peak vein velocity; RCIA, right common iliac artery; RCIV, right common iliac vein.



Fig. 1 – Drawing illustrating double May-Thurner syndrome. Coronal and sagittal views showing a compression (*) of left common iliac vein in two points respectively by the right common iliac artery (white arrow) and left common iliac artery (grey arrow) against the lumbar spine.

Introduction

May-Thurner Syndrome (MTS), also known as Cockett syndrome, belongs to the group of abdomino-pelvic vascular compression syndromes. MTS was first described in 1957 as a compression of the left common iliac vein (LCIV) between the right common iliac artery (RCIA) anteriorly and lumbar spine posteriorly (Fig. 1), resulting in chronic venous stasis and thrombosis [1–2]. A compression of LCIV by the left common iliac artery (LCIA) or by both iliac arteries were described [3–4]

Several authors suggested the continuous extrinsic pulsations of the RCIA as a pathophysiological mechanism of MTS; this parietal stress may cause endothelial spurs formation in the vein lumen through collagen and elastin deposition [5–7]. If compression of the LCIV is severe it can cause symptoms of deep venous thrombosis (DVT) which can be either acute or chronic [8,9]; in mild cases, it can be asymptomatic [10]. Doppler ultrasound (US) allows to highlight stenosis and obtain an estimate of stenosis degree [11].

Computed Tomography angiography (CTA) allows to detect the compression by overlying iliac artery against the lumbar spine and to exclude other causes of stenosis [12]; in severe cases it is necessary to exclude pulmonary or cerebral embolism. In symptomatic patients with DVT, endovascular thrombolysis with/without endovascular stenting may be indicated and required long-term prophylaxis with antithrombolytic and anticoagulant drugs [13–14].

In asymptomatic MTS a long-term prophylaxis with anticoagulant drugs is recommended.

We describe a rare case of double stenosis of LCIV caused by both RCIA and LCIA, first detected at US and confirmed by CT examinations.

Case presentation

A 58-year-old woman came to our attention complaining chronic and mild pelvic pain, with no other symptoms. Therefore, we decided to perform an abdominal US examination with an Aplio XG (Toshiba) using a convex (3.5 MHz) probe. The B-Mode US showed no alterations of abdominal and pelvic organs. Whereas the color Doppler US examination showed a double compression of the LCIV against the L5 body (Clip 1) by both the LCIA and RCIA with dilation of pre-stenotic tract and subtotal thrombosis of LCIV. The doppler US data were summarized in Table 1.



Fig. 2 – Duplex Doppler US. Measurement of PVV at (A) pre-stenotic and (B) post-stenotic LCIV. LCIV: left common iliac vein; PVV: Peak Vein Velocity.



Fig. 3 – Color-Doppler Ultrasound trasversal scan shows LCIV thrombosis (arrowhead) and measurement of minimum distances between respectively (A) RCIA (short arrow) and L5 body and (B) LCIA (long arrow) and L5 body. LCIV: left common iliac vein; RCIA: right common iliac artery; LCIA: left common iliac artery; L5: fifth lumbar spine vertebrae.

The B-Mode and Doppler US scans of LCIV showed: prestenotic diameter of 8 mm and post-stenotic of 7 mm; flux congestion with increased Peak Vein Velocity (PVV) in poststenotic tract (18.5 cm/s) and reduced PVV in pre-stenotic tract (6.9 cm/s) (Fig. 2A and B).; Flux Ratio (FR) (post-stenotic tract PVV/pre-stenotic tract PVV) of 2.68; the distance between the lumbar spine and the RCIA and LCIA were respectively 4.4 mm and 6.2 mm (Fig. 3). At doppler US examination of lower extremity vessels performed with linear (7.5 MHz) probe, a left common femoral vein (LCFV) thrombosis was depicted (Figs. 4 and 5).

In order to exclude other causes of stenosis, a CTA scan with an Optima 64 slice (GE, Healthcare) device was performed.

CTA confirmed the double compression of LCIV and chronic thrombosis of LCIV and LCFV with typical calcified thrombus. The LCIV thrombosis extended to the femoral bifurcation where large subcutaneous collateral veins from the right common femoral vein arrived as a natural venous femoro-femoral bypass. This latter allowed adequate venous drainage of lower extremity circulation.

The patient was discharged from the hospital after one week and the following drug regimen was prescribed: Warfarin (Coumadin) 4 mg for 6 months, 300 mg aspirin daily for life and 75 mg of Clopidogrel (Plavix, Bristol -Myers Squibb and Sanofi) every day for 4 weeks.

Discussion

In double MTS, the LCIV compression between the RCIA and LCIA anteriorly and the lumbar spine posteriorly may cause acute or chronic DVT [1,8]. This condition may lead to the development of a venous natural femoro-femoral bypass as an alternative venous drainage to counteract the venous stasis, preventing overwhelming venous obstruction [8]. MTS could be asymptomatic or clinical manifest with venous hypertension: swelling of the limbs, pain, claudication and/or thrombophlebitis [15]. Doppler US allows to highlight the stenosis of the LCIV and the consequent vascular alterations: caliber dilation with reduction of PVV in the pre-stenotic tract and increase of PVV in the post-stenotic tract. The PVV values allow to obtain the FR which is the best criterion defining hemodynamically significant venous stenosis [16]. In fact, since a FR value of 2.5 corresponds to a stenosis greater than 50% [17],



Fig. 4 – CT angiography. (A) Axial MIP, (B,C) sagittal MPR reconstructions images of portal venous phase show a double compression of LCIV against the lumbar spine in two points (black arrow and black arrowhead) respectively by RCIA (white arrow) with focal calcification and LCIA (white arrowhead); (D) axial reconstruction shows measurements of minimum distance between vertebral spine and respectively RCIA (orange line) and LCIA (blue line); (E) coronal and (F) sagittal reconstruction show LEIV (white arrowhead) and LFIV with chronic thrombosis and multiple calcification. *LCIV*: left common iliac vein; RCIA: right common iliac artery; LCIA: left common iliac artery; LEIV: left external iliac vein; LCFV: left common femoral vein.



Fig. 5 – CT Angiography. (A) Axial and (B) coronal MIP reconstruction images show subcutaneous collateral veins which connects RCFV (arrow) with LFCV (arrowhead). RCFV: right common femoral vein; LCFV: left common femoral vein.

our patient (FR 2.6) showed a significant venous stenosis well compensated by natural venous femoro-femoral bypass. At CT scan, chronic DVT typically appears as small-caliber vein containing calcified thrombus which can also cause development of venous femoro-femoral bypass [8]. CT angiography is also useful to rule out other causes of venous compression (abdominal or pelvic masses, aneurysms, etc.) [7,15].

In symptomatic patients with extended thrombosis of left iliac vein, the endovascular treatment is successful in 91% of

patients [7]. It consists in thrombolysis with or without inferior vena cava filter placement, followed by left iliac vein endovascular stenting [7].

In our case the patient had no symptoms of venous congestion in left lower limb probably due to the venous drainage compensation by the existing natural bypass between the LCFV and RCFV.

In asymptomatic MTS, as in our patient, a long-term prophylaxis with anticoagulant drugs is recommended.

Therefore, we decided to prescribe a long-term conservative treatment with anticoagulants and doppler US follow-up at 6 months.

Conclusion

A double compression of LCIV by both common iliac arteries is a rare condition which may cause acute or chronic DVT. This latter could be associated with the development of a venous natural femoro-femoral bypass as an alternative venous drainage.

US is a rapid, non-invasive, low-cost and repeatable method that allows to highlight all the vascular alterations typical of MTS and DVT. It may be considered the first level exam which allows to easily detect pelvic vascular compression syndrome.

Authors contribution

CG: Drafting the article, collecting and interpretation of data; RF, PC, GC: Data acquisition; PVF, SP, MV, AB: All authors read and approved the final manuscript.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Patient consent

The consent was obtained from the patient for the publication of this case report and accompanying images.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2021.04.020.

REFERENCES

 May R, Thurner J. The cause of the predominantly sinistral occurrence of thrombosis of the pelvic veins. Angiology 1957;8(5):419–27.

- [2] Moudgill N, Hager E, Gonsalves C, Larson R, Lombardi J, DiMuzio P. May-Thurner syndrome: case report and review of the literature involving modern endovascular therapy. Vascular 2009;17(6):330–5.
- [3] Caggiati A. The left common iliac artery also compresses the left common iliac vein. J Vasc Surg 2011;54(6 Suppl) 56S–61S.
- [4] Sharafi S, Farsad K. Variant May-Thurner syndrome: Compression of the left common iliac vein by the ipsilateral internal iliac artery. Radiol Case Rep. 2018;13(2):419–23.
- [5] Al-Nouri O, Milner R. May-Thurner syndrome. Vasc Dis Manage 2011;2018:8.
- [6] Molloy S, Jacob S, Buckenham T, Khaw KT, Taylor RS. Arterial compression of the right common iliac vein; an unusual anatomical variant. Cardiovasc Surg Jun 2002;10(3):291–2.
- [7] Mousa AY, AbuRahma AF. May-Thurner syndrome: update and review. Ann Vasc Surg. 2013;27:984–95.
- [8] Wu WL, Tzeng WS, Wu RH, Tsai WL, Chen MC, Lin PC, et al. Comprehensive MDCT evaluation of patients with suspected May-Thurner syndrome. AJR Am J Roentgenol 2012;199(5):W638–45.
- [9] Dhillon RK, Stead LG. Acute deep vein thrombus due to May-Thurner syndrome. Am J Emerg Med 2010;28(2) 254.e3–4PMID: 20159406. doi:10.1016/j.ajem.2009.05.016.
- [10] Kibbe MR, Ujiki M, Goodwin AL, Eskandari M, Yao J, Matsumura J. Iliac vein compression in an asymptomatic patient population. J Vasc Surg 2004;39(5):937–43.
- [11] Larkin TA, Hovav O, Dwight K, Villalba L. Common iliac vein obstruction in a symptomatic population is associated with previous deep venous thrombosis, and with chronic pelvic pain in females. J Vasc Surg Venous Lymphat Disord. 2020 S2213-333X(20)30110-4.
- [12] Lugo-Fagundo C, Nance JW, Johnson PT, Fishman EK. May-Thurner syndrome: MDCT findings and clinical correlates. Abdom Radiol (NY) 2016;41(10):2026–30 PMID: 27271358. doi:10.1007/s00261-016-0793-9.
- [13] Sebastian T, Spirk D, Engelberger RP, Dopheide JF, Baumann FA, Barco S, et al. Incidence of Stent Thrombosis after Endovascular Treatment of Iliofemoral or Caval Veins in Patients with the Post-thrombotic Syndrome. Thromb Haemost Dec 2019;119(12):2064–73 Epub 2019 Oct 28. PMID: 31659739. doi:10.1055/s-0039-1697955.
- Bondarev S, Keller EJ, Han T, Young VA, Gupta S, Vogelzang RL, et al. Predictors of disease recurrence after venoplasty and stent placement for May-Thurner syndrome. J Vasc Interv Radiol 2019;30(10):1549–54.
- [15] Gozzo C, Giambelluca D, Cannella R, Caruana G, Jukna A, Picone D, et al. CT imaging findings of abdominopelvic vascular compression syndromes: what the radiologist needs to know. Insights into imaging 2020;11(1):48.
- [16] Labropoulos N, Borge M, Pierce K, Pappas PJ. Criteria for defining significant central vein stenosis with duplex ultrasound. J Vasc Surg 2007;46(1):101–7.
- [17] Farina R, Foti PV, Iannace FA, Fanzone L, Pennisi I, Conti A, et al. May Thurner syndrome: description of a case with unusual clinical onset. J Ultrasound 2020;Online ahead of print.