

Major orthopedic surgery and its correlation with Cockett syndrome: a case report

Cirurgia ortopédica de grande porte e sua correlação com a síndrome de Cockett: relato de caso

Raffael Marum Bachir¹, Inácio Diogo Asaumi¹, Donato Lo Turco¹, Fábio Luiz Kiyan¹

1. Hospital IFOR, São Bernardo do Campo, SP, Brazil.

ABSTRACT

We report the case of a 42-year-old female patient complaining of pain in the right foot and repetitive sprains for 2 years who underwent surgical repair of pes cavus and ankle ligament reconstruction. Examination images (radiography, nuclear magnetic resonance, and ultrasound), postoperative (PO) period and progression to deep vein thrombosis are presented and discussed, as well as the resolution of this condition by endovascular surgery with angioplasty and stent implantation after suspicion of iliac vein compression syndrome caused by the contralateral iliac artery, known as Cockett syndrome.

Level of Evidence V; Therapeutic Studies; Expert Opinion.

Keywords: May-Thurner syndrome; Orthopedics; Edema; Cyanosis.

RESUMO

Relatamos o caso de uma paciente de 42 anos com queixa de dor no pé direito e entorses de repetição há 2 anos submetida à cirurgia de correção do pé cavo e reconstrução das estruturas ligamentares do tornozelo. São apresentadas e discutidas as imagens dos resultados dos exames (radiografias, ressonância nuclear magnética, ultrassonografia), do pós-operatório (PO) e da evolução para a trombose venosa profunda, bem como a resolução deste quadro pela cirurgia endovascular com angioplastia e implante de *stent*, após ser levantada a hipótese de uma Síndrome de Compressão da Veia Iliaca pela artéria ilíaca contralateral, conhecida como Síndrome de Cockett.

Nível de Evidência V; Estudos Terapêuticos; Opinião do Especialista.

Descritores: Síndrome de May-Thurner; Ortopedia; Edema; Cianose.

How to cite this article: BachirRM, Asaumi ID, Lo Turco D, Kiyan FL. Major orthopedic surgery and its correlation with Cockett syndrome: a case report. *Sci J Foot Ankle*. 2019;13(3):XX-XX.

INTRODUCTION

Cockett syndrome or iliac vein compression syndrome (IVCS) occurs when the right common iliac artery extrinsically compresses the left common iliac vein, resulting in the compression of the latter on the spinal column and obstruction of blood flow to the left lower limb (LLL)⁽¹⁾.

Studies on the laterality of deep vein thrombosis (DVT) date back to the 18th century, becoming a focus of study in 1956 with May and Thurner⁽¹⁾ and in 1965 with Cockett and Thomas⁽²⁾.

In mechanical obstruction mechanisms, veins are compressed between the artery and spine, and cellular obstruc-

Work performed at the Hospital IFOR, São Bernardo do Campo, SP, Brazil.

Correspondence: Raffael Marum Bachir. Rua Braga, 202. São Bernardo do Campo, SP, Brazil. CEP 09725-160. E-mail: raffamarum@hotmail.com

Conflicts of interest: none. **Source of funding:** none.

Date received: March 14, 2019. **Date accepted:** September 20, 2019. **Online:** September 30, 2019



Copyright © 2019 SciJFootAnkle

tion mechanisms, intimal hypertrophy of the vein occurs, resulting from repeated compression from the overlying arterial pulse, which causes shear stress between the anterior and posterior venous walls⁽³⁾.

The prevalence of this anatomical variation is approximately 22-33%^(4,5), but there is a lack of data on its distribution and epidemiological characteristics, hindering the evaluation of its clinical significance.

The clinical picture of IVCS manifests as pain and edema in the LLL and, in more severe cases, as thrombosis of the left iliofemoral vein⁽⁵⁾.

The development of DVT is a multifactorial event resulting from environmental risk factors and patient characteristics. Orthopedic surgery can also predispose patients to DVT⁽⁶⁾, mainly due to the position of the limb during the intervention, localized postoperative (PO) edema and the mobility limitations in the immediate PO period⁽⁷⁾.

Additionally, during surgery, limb manipulation, thermal reaction secondary to the use of cement and other injuries can activate thrombogenic factors that lead to vascular lesions and venous stasis⁽⁸⁾.

Orthopedic surgery can also lead to blood loss, which decreases antithrombin III levels and inhibits the endogenous fibrinolytic system, resulting in thrombus formation and growth⁽⁶⁾.

Considering the importance of IVCS and orthopedic surgery in the event of DVT and that this condition is rare in foot and ankle surgeries, we consider it relevant to describe the clinical case of a patient affected by both and to discuss her progression.

The objective of this study is to report the clinical case of a patient who underwent major orthopedic surgery and developed DVT from undiagnosed IVCS.

CASE REPORT

This study was approved by the Research Ethics Committee with registration in the Brazil Platform under CAAE number: 09386919.6.0000.5625.

A 42-year-old female patient complained of constant medium intensity pain for 6 months on the side of the right foot and experienced repetitive sprains for 2 years. On physical examination, the patient presented with pes cavus varus and hindfoot with pronounced varus and, on palpation, pain over the anterior fibulotalar (AFT) ligament and tarsal sinus associated with a slight shortening of the posterior chain. She had a 3+/4+ positive drawer test and pain upon external rotation of the limb. Anteroposterior and profile

radiographs of the foot with weight-bearing showed an increased calcaneal pitch and an abnormal Meary's angle, consistent with pes cavus (Figure 1). Nuclear magnetic resonance imaging (MRI) showed AFT ligament thickening, displaced syndesmosis and joint effusion, indicating the need for corrective surgery. In the preoperative examinations, the patient was classified as ASA 1. The surgery began with ankle arthroscopy with noninvasive traction to remove inflammatory tissue. Next, the device was removed, and open surgery was started, with hindfoot and midfoot osteotomies performed for correction of pes equinovarus, FTA ligament reconstruction using the Brostrom-Gould technique and syndesmosis stabilization with the use of flexible fixation (Figure 2). The surgery lasted approximately 3 hours, without the use of crystalloid and blood transfusion. The patient was kept immobilized with a plaster cast and received antibiotic, anti-inflammatory and analgesic medications. According to the hospital protocol, at least 2 of the following criteria are necessary for DVT prophylaxis: total hip arthroplasty, total knee arthroplasty, spinal cord injury, duration of surgery greater than 3 hours and risk factors for venous thromboembolism (VTE). (Figure 3). The patient, however, met only one of the criteria (change in the contraceptive hormone) and therefore was not subjected to prophylaxis. Active physical therapy was started on the first PO day, and the patient was discharged on the second PO day in good general condition. In the first PO week, the patient returned to the outpatient clinic, showing excellent healing and no signs of DVT. At 2 and a half weeks after surgery, she presented with sudden pain in the popliteal region, with irradiation to the left calf and thigh. There was 3+/4+ edema, 4+/4+ hyperemia and skin with a cyanotic appearance (Figure 4). On palpation, the patient presented severe pain in the posterior region of the leg and left calf muscle stiffness, and Homan's, Bandeira's and Bancroft's signs were positive for DVT. Ultrasound with venous and arterial Doppler of the LLL was requested, which detected extensive thrombosis of the iliac vein and its compression by the contralateral



Figure 1. Profile radiograph of the right lower limb showing pes cavus.

Source: Author's personal archive.



Figure 2. Immediate postoperative profile radiograph of the right lower limb showing lateral sliding osteotomy for pes cavus. **Source:** Author’s personal archive.



Figure 4. LLL with cyanotic edema and mild hyperemia, 2 and a half weeks after right pes cavus correction surgery. **Source:** Author’s personal archive.

Check all risk factors for VTE	
<input type="checkbox"/> Stroke	<input type="checkbox"/> Infection
<input type="checkbox"/> Cancer	<input type="checkbox"/> Peripheral renal failure
<input type="checkbox"/> Venous catheters	<input type="checkbox"/> ICU admission
<input type="checkbox"/> Active intestinal inflammatory disease	<input type="checkbox"/> Obesity (BMI ≥ 30kg/m ²)
<input type="checkbox"/> Severe respiratory disease	<input type="checkbox"/> LL paresis or paralysis
<input type="checkbox"/> Active rheumatologic disease	<input type="checkbox"/> Chemo/hormonotherapy
<input type="checkbox"/> Pregnancy and postpartum	<input type="checkbox"/> Hormone replacement/contraceptive use
<input type="checkbox"/> History of VTE	<input type="checkbox"/> Active nephrotic syndrome
<input type="checkbox"/> AMI	<input type="checkbox"/> Thrombophilia
<input type="checkbox"/> CHF class III or IV	<input type="checkbox"/> Varicose veins/venous insufficiency

Figure 3. Risk factors for venous thromboembolism described in a protocol for the hospital where the present study was conducted. **Source:** Author’s personal archive.

iliac artery, indicating IVCS. Some indirect ultrasound signs assisted in the diagnosis: 1) flow volume in the right common iliac vein 40% greater than the flow volume in the left common iliac vein; and 2) index between the peak velocity of the left and right common femoral veins smaller than 0.9. Other possible diagnostic methods include computed tomography and MRI, both with high degrees of accuracy;

however, they were not performed. An evaluation was requested from the vascular team, who chose to initiate anticoagulation and analgesia and perform catheterization with angioplasty and stenting (Figure 5). The patient’s clinical condition improved, and she was discharged from the hospital discharge after 2 days and returned to the orthopedic PO program.

DISCUSSION

The emergence of any of Virchow’s triad factors (venous stasis, endothelial damage and hypercoagulation)⁽⁷⁾ directly promotes the advent of DVT.

Blood stasis caused by IVCS is a risk factor for the development of thrombosis clinically manifested as progressive LLL edema in individuals without any apparent risk factor for thrombosis and even with the use of anticoagulants. It is considered a chronic event characterized by venous claudication and, sometimes, vague and mild complaints. On physical examination, skin alterations such as varices, lipodermatosclerosis and venous ulcers may be observed⁽⁹⁾.

Orthopedic surgery is considered a risk factor for the development of DVT, mainly due to positioning and manipulating the limb during the intervention; localized PO

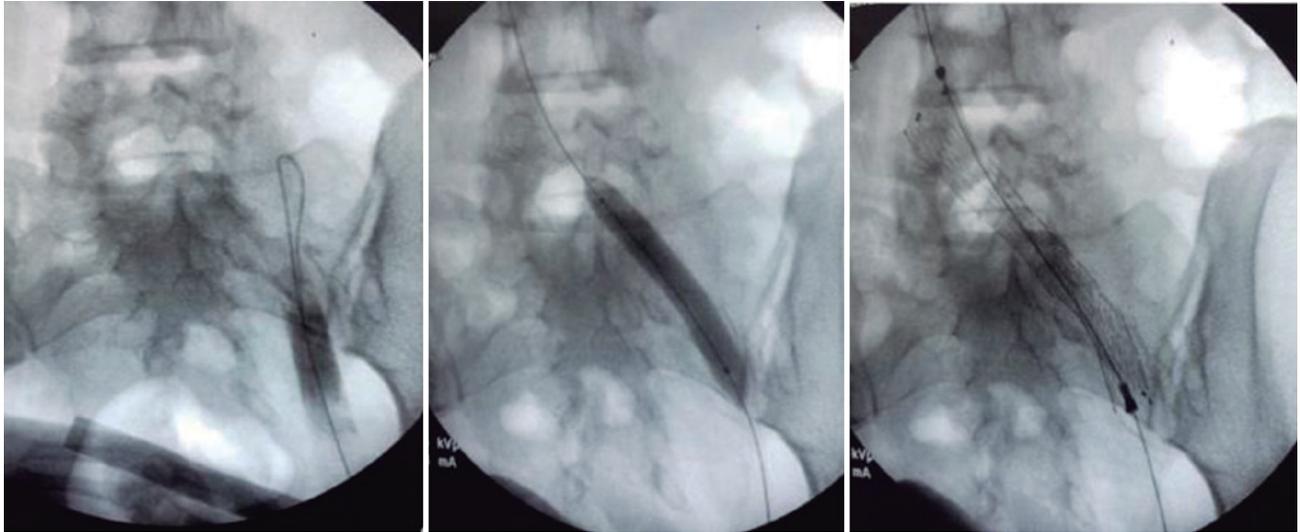


Figure 5. Vascular stenting procedure and outcome of the emergency vascular procedure.

Source: Author's personal archive.

edema; mobility limitations in the PO period; thermal reaction secondary to the use of cement; and blood loss⁽⁶⁻⁸⁾.

Moreover, hospitalization is considered a risk factor because patients are bedridden, immobilized and have mobility limitations. The risk is extremely high, ranging from 10 to 20%, in patients with clinical conditions, and among those hospitalized and subjected to surgery, patients with cancer or undergoing orthopedic surgery are most at risk⁽⁶⁾.

In the reported case, the patient, who was walking, returned to the outpatient clinic on the 17th PO day with complaints of significant sudden pain, significant edema, hyperemia and cyanosis of the LLL. However, because she underwent a major orthopedic surgery, the first option as a cause would be the surgery and the PO period itself. With deeper investigation, it was possible to establish a diagnosis previously unknown to the patient, IVCS.

The patient did not receive prophylaxis for DVT because she presented only one of the criteria (contraception change). Notably, the surgery lasted approximately 3 hours and thus was not considered a criterion for prophylaxis (>3 hours). Although lower limb surgery was performed, thrombotic events in foot and ankle surgeries are rare, in contrast to hip and knee surgeries, which have a thrombotic event incidence of approximately 60%. In addition, despite being immobilized, the patient had performed active physical therapy since the first PO day. At this stage, the diagnosis of IVCS was not yet known; however, if there had been prior knowledge of this condition, together with

knowledge regarding the change in contraceptive, DVT could have been avoided through prophylactic treatment.

We chose endovascular surgery with angioplasty and stenting. Currently, there are 2 options established in the literature for IVCS correction: venous decompression by open or endovascular surgery, which has the main objective of decreasing venous blood stasis in the LLL and preventing future thromboembolic events.

Endovascular techniques combined with balloon angioplasty with stenting have high rates of clinical success, providing recanalization of the iliac venous system and minimizing surgical trauma. Sandri (2011)⁽⁹⁾ evaluated 54 patients with IVCS who were treated percutaneously and found almost complete improvement of symptoms in 92.5% of patients.

The current literature reports that the endovascular technique combined with balloon angioplasty with stenting provides excellent results in the recanalization of the iliac venous system, subjecting patients to minimal surgical trauma with little chance of complications⁽¹⁰⁾.

CONCLUSION

IVCS and orthopedic surgery are important risk factors for the development of DVT and, in the present study, caused this pathological condition.

Endovascular surgery with angioplasty and stenting is the main mode of treatment, presenting excellent clinical results.

Authors' contributions: Each author contributed individually and significantly to the development of this article: RMB *(<https://orcid.org/0000-0002-5462-0905>) conceived and planned the activities that led to the study, interpreted the results of the study, wrote the article, participated in the review process; IDA *(<https://orcid.org/0000-0002-4074-0412>) conceived and planned the activities that led to the study, interpreted the results of the study, participated in the review process, approved the final version; DLT *(<https://orcid.org/0000-0001-9024-2553>) conceived and planned the activities that led to the study, interpreted the results of the study, approved the final version; FLK *(<https://orcid.org/0000-0002-6450-9768>) conceived and planned the activities that led to the study, interpreted the results of the study, approved the final version. *ORCID (Open Researcher and Contributor ID).

REFERENCES

1. May R, Thurner J. Elin gefabspom in der vena iliaca communis sinistra als ursache de rlinksseitigen beckenve-nentrombosen. *Z Kreislaufforsch.* 1956;45(23-24):912-22.
2. Cockett FB, Thomas ML. The iliac compression syndrome. *Br J Surg.* 1965;52(10):816-21.
3. Kalu S, Shah P, Natarajan A, Nwankwo N, Mustafa U, Hussain N. May-thurner syndrome: a case report and review of the literature. *Case Rep Vasc Med.* 2013;2013:740182.
4. Cockett FB, Thomas ML, Negus D. Iliac vein compression: its relation to iliofemoral thrombosis and the post-thrombotic syndrome. *Br Med J.* 1967;2(5543):14-9.
5. May R, Thurner J. The cause of the predominantly sinistral occurrence of thrombosis of the pelvic veins. *Angiology.* 1957;8(5):419-27.
6. Eriksson BI, Eriksson E, Gyzander E, Teger-Nilsson AC, Risberg B. Thrombosis after hip replacement. Relationship to the fibrinolytic system. *Acta Orthop Scand.* 1989;60(2):159-63.
7. Merli GJ. Pathophysiology of venous thrombosis, thrombophilia, and the diagnosis of deep vein thrombosis-pulmonary embolism in the elderly. *Clin Geriatr Med.* 2006;22(1):75-92.
8. Bredbacka S, Andreen M, Blombäck M, Wykman A. Activation of cascade systems by hip arthroplasty: no difference between fixation with and without cement. *Acta Orthop Scand.* 1987;58(3):231-5.
9. Sandri GA. Tratamento endovascular das obstruções venosas crônicas do segmento iliocaval. *J Vasc Bras.* 2011;10(2):137-44.
10. Sandri JL. Síndrome de May-Thurner: tratamento endovascular. In: Brito CJ, organizador. *Cirurgia vascular.* 2ed. Rio de Janeiro: Revinter; 2007. p. 1653-64.