

Late-Diagnosed Arterial Thrombosis Mimicking Complex Regional Pain Syndrome After TKR in a Patient With CREST Syndrome

Tomás L. Nicolino, Mariano Revah, Julián Costantini, Juan Astoul Bonorino, Lisandro Carbó, Matías Costa Paz
 Arthroscopy and Knee Prosthesis Sector, Orthopedics and Traumatology Service, Hospital Italiano de Buenos Aires,
 Autonomous City of Buenos Aires, Argentina

ABSTRACT

Arterial thrombosis after total knee replacement (TKR) is a rare complication; however, it can lead to disastrous consequences. We describe a case of an arterial occlusion after TKR in an 85-year-old female patient. Initially, the clinical presentation made us consider Raynaud (she had a history of CREST) and complex regional pain syndrome, which delayed the diagnosis for 2 weeks. Diagnosis and treatment were accomplished with angiography and the patient achieved a full recovery. The low frequency of this injury may make diagnosis a challenge for the surgeon. In this case, partial occlusion of the arteries avoided more serious complications.

Key words: Total knee replacement; arterial thrombosis; vascular injury; complex regional syndrome pain; CREST syndrome.
Level of Evidence: IV

Trombosis arterial de diagnóstico tardío que simula un síndrome de Südeck luego de un reemplazo total de rodilla

La trombosis arterial luego de una artroplastia total de rodilla es una complicación poco frecuente; sin embargo, puede tener consecuencias devastadoras. Describimos un caso de una oclusión arterial después de una artroplastia total de rodilla en una mujer de 85 años. Inicialmente, la presentación clínica nos hizo pensar en una manifestación de la enfermedad de Raynaud (la paciente tenía antecedente de síndrome de CREST). Entre los diagnósticos diferenciales también se planteó un síndrome de dolor regional complejo, que retrasó el diagnóstico durante dos semanas. Finalmente, el diagnóstico y el tratamiento se realizaron mediante angiografía. La recuperación funcional fue completa. La baja frecuencia de esta lesión puede determinar que el diagnóstico sea un desafío para el cirujano. En este caso, la oclusión parcial de las arterias evitó complicaciones más graves.

Palabras clave: Artroplastia total de rodilla; trombosis arterial; lesión vascular; síndrome de dolor regional complejo; síndrome de CREST.
Nivel de Evidencia: IV

INTRODUCTION

Iatrogenic vascular lesions in a total knee replacement are rare, the published incidence is 0.09-0.17%.^{1,2} While the frequency is low, the consequences can be devastating and include the loss of the affected limb.

The most common types of vascular lesion are arterial thrombosis, arteriovenous fistula, direct vascular damage, and the formation of (pseudo)aneurysms.^{3,4} In the event of the suspicion of arterial thrombosis, one of the diagnostic methods of choice is angiography, which may be useful for diagnosis before revascularization surgery or as a definitive therapeutic option in selected cases. Treatment of arterial thrombosis may include oral anticoagulants or surgical procedures, such as revascularization, thrombectomy, or angioplasty.

Received on June 18th, 2021. Accepted after evaluation on October 21st, 2021 • Dr. MARIANO REVAH • marianorevah@gmail.com  <https://orcid.org/0000-0002-0432-2838>

How to cite this article: Nicolino TL, Revah M, Costantini J, Astoul Bonorino J, Carbó L, Costa Paz M. Late-Diagnosed Arterial Thrombosis Mimicking Complex Regional Pain Syndrome After TKR in a Patient With CREST Syndrome. *Rev Asoc Argent Ortop Traumatol* 2022;87(1):79-84. <https://doi.org/10.15417/issn.1852-7434.2022.87.1.1397>

CREST syndrome (calcinosis, Raynaud's phenomenon, esophageal dysmotility, sclerodactyly, and telangiectasia) is a type of limited cutaneous generalized sclerosis. Although the microangiopathy of this disease is known, whose main presentation is Raynaud's phenomenon, in recent years, the possibility of systemic sclerosis affecting macrovasculature has been studied. It remains a controversial topic, but certain publications support it.⁵⁻⁷

We present the case of an 85-year-old patient with a history of CREST syndrome who underwent a scheduled total knee arthroplasty and suffered a popliteal artery thrombosis diagnosed late—two weeks after surgery—and after treatment achieved a complete functional recovery without sequelae.

CLINICAL CASE

An 85-year-old woman was diagnosed with a 19° right genu valgus (Figure 1). As a history, she presented CREST syndrome, hypothyroidism, and arterial hypertension. A total knee arthroplasty (PFC® Sigma, DePuy, Leeds, UK) was performed which lasted 75 minutes, there were no intraoperative complications (Figure 2). No hemostatic cuff was used. At 6 h of the procedure, the patient reported intense pain in the leg and foot, burning sensation with hypersensitivity in the foot, and coloration changes that lasted a few minutes and disappeared with analgesic agents. Initially, Raynaud's disease was suspected due to her underlying condition, but it was ruled out by the clinical presentation. The patient gradually improved during hospitalization and was discharged.



Figure 1. Anteroposterior and lateral panoramic and focalized. Preoperative panoramic, anteroposterior and lateral knee radiographs. Right genu valgum. Tricompartmental osteoarthritis.



Figure 2. Postoperative anteroposterior and lateral knee radiographs. Total knee arthroplasty.

On day 10 of the surgery, she consulted again for intense pain in the leg, with marked hypersensitivity and no fever or pain when moving the knee joint. Slight edema and coloration changes were observed on the anterior side of the leg. No evident joint effusion, increase in temperature, or alteration of the surgical wound were detected (Figure 3).

The patient was evaluated at the clinical emergency center and hospitalized for a suspected complex regional pain syndrome. Among the differential diagnoses, a deep vein thrombosis was also proposed and ruled out with a negative venous Doppler ultrasound. The results of the laboratory analyses were: erythrocyte sedimentation rate 98 mm/h and C-reactive protein of 115.

The patient continued with a regular evolution, without an accurate diagnosis. On day 14, another Doppler ultrasound was performed, this time arterial and venous, and partial occlusion of the popliteal artery and the tibiofibular trunk was diagnosed. An angiogram was requested to complete the evaluation and indicate the possible treatment.

As shown in Figure 4, the popliteal artery had a partial occlusion of its middle and distal third with recanalization in the tibiofibular trunk that presented a diffuse severe stenosis. The anterior tibial artery was permeable up to the distal third where it was occluded, the fibular and posterior tibial arteries were permeable.



Figure 3. Clinical evolution 10 days after surgery. The discoloration is observed in the anterior side of the leg and in the distal region of the surgical wound. Mild edema in the lower limbs, without an increase in temperature or joint effusion.

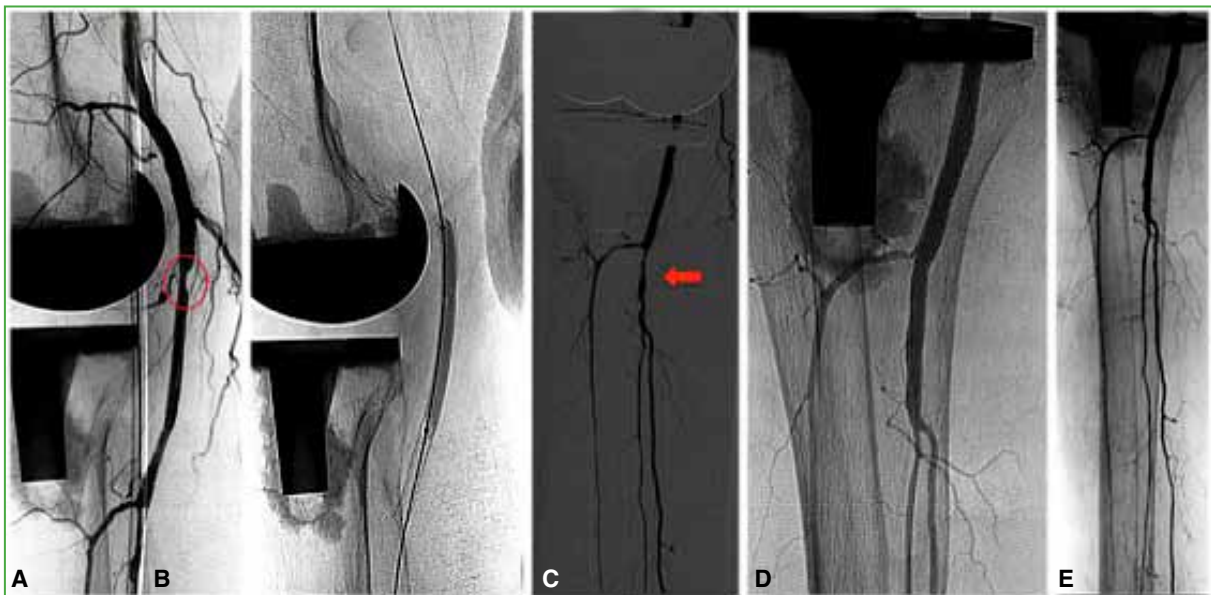


Figure 4. A sequence of digital angiography images for diagnosis and treatment. **A.** Occlusive lesion of the popliteal artery. **B.** Placement of the popliteal stent. **C.** Permeability in the popliteal artery with obstruction in the tibiofibular trunk (arrow). **D.** Placement of the stent in the tibiofibular trunk. **E.** Posterior control showing distal permeability.

A Complete® SE stent (Medtronic Inc., Santa Rosa, CA, USA) of 5 x 80 mm was placed with which the occlusive lesion of the popliteal artery was covered to later perform an angioplasty for the chronic residual lesion. In the posterior control, we observed a permeable popliteal artery without areas of significant residual stenosis and persistence of the trunk lesion that was treated with a 3.5 x 30 mm Resolute Integrity® stent (Medtronic Inc., Santa Rosa, CA, USA); a good final angiographic outcome was achieved, with no evidence of residual stenosis.

The patient progressed favorably and the pain improved. In the annual postoperative control, knee range of motion was 0-115°. The pain score, according to the visual analog scale, was 2/10 and the *Knee Society Score* was 85/90. There were no complications from the vascular lesion. A control arterial Doppler ultrasound was performed which revealed that the two stents placed (popliteal and tibiofibular trunk) were permeable, with biphasic flow at normal velocity.

DISCUSSION

Popliteal artery thrombosis after total knee arthroplasty is a very rare but severe complication. It can manifest with loss of sensation, pain, paleness or mottling of the skin, and pulse loss. It is important to administer treatment as soon as possible to decrease the risk of sequelae and loss of the limb. While direct arterial damage with heavy bleeding is usually diagnosed immediately during surgery, the diagnosis of the rest of the conditions, including thrombosis, may be delayed.

In our case, the patient presented clinical signs that led to the suspicion of other conditions, mainly complex regional pain syndrome, once the possibility of infectious complications or deep vein thrombosis was ruled out.

Complex regional pain syndrome, formerly known as Südeck syndrome, manifests itself as a painful condition that is not proportional to the event that triggers it, associated with different clinical manifestations, such as edema, changes in sensitivity, color and temperature of the limb. Our patient came with severe and burning pain, hypersensitivity, changes in skin color and temperature. These signs and symptoms, with no other diagnosis to justify them, led us to suspect a complex regional pain syndrome. Differential diagnoses of this condition include infections, neuropathic pain, Raynaud's disease, arterial insufficiency. Other diseases that could have explained the condition are a microcirculatory disorder due to CREST syndrome or cholesterol embolism from a broken popliteal plaque. The complementary studies to rule out these diseases allowed us to reach the diagnosis of thrombosis of the popliteal artery. The accurate diagnosis was delayed, two weeks after surgery, an average time similar to that reported in other publications (Table).⁸

Table. Average time until diagnosis according to the literature.

Injury	Time until diagnosis
Hemorrhage	Intraoperative
Thrombosis	12 days (from 0 h to 6 weeks)
Pseudoaneurysm	72.3 days (8 days to 12 months)
Arteriovenous fistula	24 days (6 days to 6 weeks)

In this case, the patient had a partial occlusion of the popliteal artery (30%), which allowed some distal perfusion in the limb and avoided more serious consequences. Arterial thrombosis is due to the rupture of an atheromatous plaque. Risk factors for plaque rupture are peripheral vascular disease, use of hemostatic cuff in a patient with femoral or popliteal artery calcifications, and indirect trauma from manipulation of the joint. While the patient had no risk factors before surgery, it is important to note the importance of these factors in selecting patients for surgery and exposing them to such risks.

The case was satisfactorily resolved by angiographic interventionism, which allowed diagnosis and treatment through the implantation of two stents.

Treatment of these vascular complications has advanced in recent years. Although it depends on the type of injury produced, open surgery has been confined to specific cases when direct repair or revascularization is required. Digital angiography is a tool of great diagnostic and therapeutic utility. Various repair techniques have been described either by embolization, cauterization, and placement of stents that will depend on the type of injury that has occurred. This is a safe method with a low rate of complications and a lower risk of infection than in an open procedure, it is performed under local anesthesia, which allows a quick return to the patient's rehabilitation program.^{9,10}

Conflict of interests: The authors declare they do not have any conflict of interests.

T. L. Nicolino ORCID ID: <https://orcid.org/0000-0002-9550-3713>

J. Costantini ORCID ID: <https://orcid.org/0000-0001-8900-6254>

J. Astoul Bonorino ORCID ID: <https://orcid.org/0000-0001-6798-8242>

L. Carbó ORCID ID: <https://orcid.org/0000-0002-8053-0890>

M. Costa Paz ORCID ID: <https://orcid.org/0000-0002-8217-1086>

REFERENCES

1. Abularrage CJ, Weiswasser JM, Dezee KJ, Slidell MB, Henderson WG, Sidawy AN. Predictors of lower extremity arterial injury after total knee or total hip arthroplasty. *J Vasc Surg* 2008;47(4):803-7. <https://doi.org/10.1016/j.jvs.2007.11.067>
2. Calligaro KD, Dougherty MJ, Ryan S, Booth RE. Acute arterial complications associated with total hip and knee arthroplasty. *J Vasc Surg* 2003;38(6):1170-7. [https://doi.org/10.1016/s0741-5214\(03\)00918-2](https://doi.org/10.1016/s0741-5214(03)00918-2)
3. Sharma H, Singh GK, Cavanagh SP, Kay D. Pseudoaneurysm of the inferior medial geniculate artery following primary total knee arthroplasty: delayed presentation with recurrent haemorrhagic episodes. *Knee Surg Sports Traumatol Arthrosc* 2006;14(2):153-5. <https://doi.org/10.1007/s00167-005-0639-4>
4. Dennis DA, Neumann RD, Toma P, Rosenberg G, Mallory TH. Arteriovenous fistula with false aneurysm of the inferior medial geniculate artery. *Clin Orthop Relat Res* 1987(222):255-60. PMID: 3621729
5. Youssef P, Englert H, Bertouch J. Large vessel occlusive disease associated with CREST syndrome and scleroderma. *Ann Rheum Dis* 1993;52(6):464-6. <https://doi.org/10.1136/ard.52.6.464>
6. Dorevitch MI, Clemens LE, Webb JB. Lower limb amputation secondary to large vessel involvement in scleroderma. *Br J Rheumatol* 1988;27(5):403-6. <https://doi.org/10.1093/rheumatology/27.5.403>
7. Man A, Zhu Y, Zhang Y, Dubreuil M, Rho YH, Pelo-quin C, et al. The risk of cardiovascular disease in systemic sclerosis: a population-based cohort study. *Ann Rheum Dis* 2013;72(7):1188-93. <https://doi.org/10.1136/annrheumdis-2012-202007>
8. Pal A, Clarke JMF, Cameron AEP. Case series and literature review: Popliteal artery injury following total knee replacement. *Int J Surg* 2010;8(6):430-5. <https://doi.org/10.1016/j.ijvs.2010.04.008>
9. Bagla S, Rholl KS, van Breda A, Sterling KM, van Breda A. Geniculate artery embolization in the management of spontaneous recurrent hemarthrosis of the knee: case series. *J Vasc Interv Radiol* 2013;24(3):439-42. <https://doi.org/10.1016/j.jvir.2012.11.011>
10. Sadat U, Naik J, Verma P, See TC, Cousins C, Chitnavis JP, et al. Endovascular management of pseudoaneurysms following lower limb orthopedic surgery. *Am J Orthop (Belle Mead NJ)* 2008;37(5):E99-E102. PMID: 18587511