ABSTRACT

Acute atraumatic compartment syndrome is a rare entity; of the peroneal compartment, it is even rarer and bilateral even more so. We present the case of a patient who underwent surgery to rescue a brachiocephalic mega-fistula with three aneurysms associated with a stenosis of the cephalic arch. The technique consisted of aneurysmectomy, removal of the excess vein, transposition, and a new anastomosis distal to the axillary vein. The procedure lasted 220 minutes, and general anesthesia was used. The following day, the patient reported left leg paraesthesia, pain, and inability to evert the left foot or walk. Arterial pulses were normal, as was pulse wave Doppler. MRI confirmed peroneal compartment syndrome, and an emergency fasciotomy was performed. While recovering with full sensation and motility for the next 24 hours on the left side, she presented with pain and cramps in the right peroneal compartment without more severe symptoms, so fasciotomy was not performed; the condition resolved spontaneously within 24 hours. This is the first reported case of atraumatic bilateral peroneal compartment syndrome after dialysis access surgery, with full recovery after prompt treatment.

Keywords: acute compartment syndrome, atraumatic fasciotomy, peroneal compartment, lateral compartment, mega-fistula, dialysis access complication, bilateral compartment syndrome.
INTRODUCTION
Patients with end-stage renal disease are often complex cases for the vascular team. Solving dialysis access failure requires a thorough understanding of the hemodynamic problems and the corresponding techniques and procedures. There is no consensus definition of a mega-fistula, although it can be defined as a generalized aneurysmal dilatation of the entire outflow tract of an arteriovenous fistula (AVF). There is consensus that an aneurysmal segment has twice the diameter of the adjacent normal vein, together with a high intra-access pressure and an access flow greater than 2000 ml/min\(^1\). Outflow stenosis is often present, and previous renal transplantation may also be a risk factor\(^2\). In addition to dilatation of the entire vein, true aneurysms may also occur, which worsen the prognosis of the AVF and require additional procedures for treatment. Treatment options often include segmental graft replacement, angioplasties, covered stents, and, most commonly, ligation of the AFV.

Acute compartment syndrome (ACS) is a severe limb-threatening condition. It is caused by abnormally high tissue pressure in a compartment enclosed by the fascia in the limbs, which contains muscle tissues, nerves, and blood vessels. If the pressure within the compartment exceeds the ischaemic threshold of the tissue, irreversible damage can occur\(^3\). Traumatic ACS is much more common, while spontaneous compartment syndrome presents in the most unexpected ways. Bilateral lateral or peroneal compartment syndrome is rare. It has been described in cases such as methanol intoxication, alcohol, simvastatin-induced, prolonged positioning, spontaneous bleeding due to anticoagulation, and saphenous vein stripping.

CLINICAL CASE
We present the case of a 28-year-old male patient with a brachiocephalic fistula of 9 years of evolution who presented with elevated venous pressure during dialysis. He was on chronic treatment with statins, valsartan, nevibolol, and amlodipine and had a secondary hyperparathyroidism under study. The AVF consisted of a mega-fistula with three aneurysms and a cephalic arch stenosis. Initially, an 8 mm balloon angioplasty was performed with instant recurrence. Due to the problems encountered, surgical treatment was chosen. A salvage technique described by Nezakatgoo et al.\(^4\) was used. Under general anesthesia, the vein was wholly dissected 2 cm from the arterial anastomosis and sectioned as distally as possible; a 24 Fr thoracostomy tube was used to calibrate the AVF and excess vein, and aneurysms were removed (Figure 1). A continuous Prolene\(^\text{TM}\) 6-0 suture line was used over the tube, tunneled with a 90-degree rotation, and reanastomosed to the axillary vein (Figure 1). The procedure lasted 220 minutes with the patient in the supine position, with low-dose noradrenaline requirements, and no complications were observed. The patient woke up with moderate pain in the left calf, full mobility, and sensation; all pulses were palpable. The following day, he evolved with intense pain, lack of sensibility, and no eversion, with a precise diagnosis of peroneal palsy.

FIGURE 1. Mega-fistula with three aneurysms (top) and fistula calibration with thoracostomy tube (bottom).
The creatine phosphokinase (CPK) value was 13,624 IU/L. Arterial Doppler reported normal pulse waves with arterial calcification. MRI showed significant edema only in the left lateral compartment (Figure 2). An emergency fasciotomy was performed, where the muscles (pale at the time) showed rapid recovery of perfusion within a couple of minutes. The skin was sutured and closed without tension. The patient regained total motility and sensation within 24 hours. He started with pain and cramping in the contralateral peroneal compartment, with no nerve palsy or other symptoms. Another MRI was performed and again showed edema in the lateral compartment (Figure 3). No sensation and motility decreased in this limb, so no decompression was performed. CPK decreased to 4118 IU/L, 2588 IU/L, and 1295 IU/L in the following days. On the seventh day, already asymptomatic, he was discharged from the hospital. The patient has total motility and sensibility three years later, with a functional native AVF.

**FIGURE 2.** MRI of the left leg. Edema is seen in the peroneal compartment.

**FIGURE 3.** MRI of the right leg. Non-tension edema is seen in the peroneal compartment.

**DISCUSSION**

Compartment syndrome is expected in the context of acute trauma and vascular pathology, mainly in situations of acute ischemia. Younger male patients are at increased risk of developing compartment syndrome due to the relative hypertonicity of the muscles, which does not leave adequate space for localized inflammation. Acute atraumatic compartment syndrome (ACS) is a rare situation, demanding suspect and requiring immediate treatment. Several comorbidities have been considered for the development, such as hypertension, dyslipidemia, obesity, diabetes, renal disease, cancer, and cardiac disease; multiple medications such as antihypertensives, statins, anticoagulants, antidiabetics, opioids, paracetamol, vasoactive drugs, and diuretics; and several situations such as trauma, acute ischemia, exercise, postural and some surgeries (cardiac and orthopedic). Both anticoagulants and statins have been independently associated with an increased risk of developing ACS. In 2020, the first case of spontaneous gene-related ACS in a young male with sequential four-limb ACS was published. The association to a genetic mutation in exon 3 of the GYG1 gene, related to muscle metabolism, is likely.

Long-term surgeries in lithotomy position are recognized as a risk factor for ACS. ACS has been reported in patients with cardiovascular pathology after cardiac surgery, usually on the same limb from which the saphenous vein was removed. Diagnosis is not accessible after this type of surgery, as the patient is usually on assisted ventilation, and no information on functional status can be obtained. ACS has also been described after prolonged aortic endovascular procedures, most likely due to prolonged ischaemia. In our case, position, hypotension due to anesthesia, and the need for vasoactive drugs were risk factors influencing the outcome.
Spontaneous or atraumatic bilateral compartment syndrome is a rare condition. It has been reported most frequently after exercise, in both upper and lower limbs, and after prolonged standing11,12. Bilateral peroneal ACS has been described after horseback riding, usually with a very late diagnosis13.

Fasciotomies should be performed promptly once ACS is established. Fasciotomies performed within 6 hours achieved complete functional limb recovery in a study by Rorabeck and Macnab14. When performed within 12 hours of acute onset, 68% achieved normal limb function, but only 8% did so when fasciotomy was performed after that period15. Hyperbaric oxygen therapy has been reported as another treatment for ACS, but fasciotomy is still the standard treatment16. In our case, muscle perfusion recovered a few minutes after fasciotomy and peroneal nerve palsy recovered within 24 hours despite having a 32-hour interval from symptom onset and treatment. The pain was the only symptom in the right leg, and the MRI showed mild edema, so it was decided to maintain expectant management. As the patient was already on dialysis, elevated CPK levels and renal failure were not a concern.

CONCLUSIONS

We present the first case of acute compartment syndrome after dialysis access surgery; the fact that it is of the peroneal compartment and bilateral makes it even rarer. A high index of suspicion, early diagnosis, and a multidisciplinary approach are the best ways to deal with this type of situation, as they maximize the chances of recovery and avoid permanent disability.

Declarations
The authors declare no conflict of interest.

Consent
Written informed consent was obtained from the patient to publish this case report.