Paraparesis due to ischemic lumbosacral Radiculoplexopathy

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ABSTRACT

Background: Lumbosacral radiculoplexopathy (LSRP) is a rare entity, with several etiologies described in the literature. Vascular lesions to the lower part of the aorta and the common iliac artery can cause LSRP of ischemic etiology.

Clinical Presentation: A 66-year-old man with a previous history of peripheral obstructive arterial disease (POAD) requiring aortobifemoral and femoropopliteal procedures had a prolonged hospital stay due to various complications: stent infection and occlusion, aortoenteric fistula causing hemorrhagic shock, transmetatarsal amputation of the right foot. In this context, he underwent multiple endovascular procedures. At discharge, the patient presented flaccid paraparesis, distal bilateral hypoesthesia, saddle anesthesia and urinary and anal incontinence. Spinal cord infarction was excluded. An electromyographic study confirmed the diagnosis of bilateral LSRP of ischemic etiology.

Conclusion: This case highlights the importance of including ischemic etiology as a differential diagnosis of peripheral nerve injuries, namely in patients with POAD.

Keywords: Paraparesis, lumbosacral radiculoplexopathy, endovascular procedure, peripheral obstructive arterial disease, ischemic radiculoplexopathy, case report.

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Background

Lumbosacral plexopathy is a rare clinical syndrome characterized by acute/sub-acute asymmetric weakness, reduced or absent deep tendon reflexes, dysesthesias and/or paresthesias of lower limbs (LLs) in multiple lumbosacral nerve root distribution [1,2]. It can result from diabetes, neoplasia, infection, trauma, radiation, pregnancy, surgery, retroperitoneal hematoma or abscess, vasculitis, connective tissue disorders, inflammation, infiltration (amyloid), or ischemia [1-3]. Frequently, there is concomitant involvement of the root, plexus, and nerve, leading to a lumbosacral radiculoplexopathy (LSRP). In LSRP, electromyography (EMG) is characterized by the involvement of at least two roots of different levels, two peripheral nerves, and paraspinal muscles' denervation [1].

Hypotension, hypoperfusion, athero-embolization, or temporary/permanent interruption of critical radicular or pelvic arteries during endovascular aortoiliac procedures (bypass or stent placement) can lead to neurological deficits due to diminished blood supply to the spinal cord (SC) or to the lumbosacral plexus (LSP) [4-6,8]. Myelopathy, cauda equina syndrome and LSRP are the most commonly documented neurological pictures related to aortic interventions. The pattern and degree of the neurologic deficit are unpredictable and heterogeneous because of the variable blood supply to the SC [4,7-9].

Ischemic injury to the LSP causing lower limb (LL) paralysis following aortoiliac procedures is a known but rare complication, with recovery rates difficult to predict [4,5,9,10]. LSRP of ischemic origin manifests as sudden unilateral pain at the flank or lumbar region, followed by ipsilateral motor deficits [1,4]. The exact location of the lesion is often difficult to ascertain [4,7,9]. Besides, further investigation such as magnetic resonance imaging (MRI) scans and neurophysiological studies are usually unhelpful [5].

These unpredictable events often lead to permanent deficits and may cause significant morbidity [4-6,9].

Case Presentation

A 66-year-old male with a previous history of smoking and aortobifemoral and femoropopliteal bypass in the left lower limb (LLL) 6 years ago, due to peripheral obstructive arterial disease (POAD), was admitted to the Emergency Department (ED) with a stent's infection, causing important vascular occlusion to the lower body. He presented fever, pain, defense to palpation in the left iliac fossa,

and absence of all LLs arterial pulses, except for the right femoral pulse. Neurological examination to the extremities and perineal areas were normal. Abdominal and pelvic computed tomography angiography (CTA) scans revealed total occlusion of the aortobifemoral stent's left component, a prosthetic-duodenal fistula, as well as a local inflammatory/infectious process (Figures 1 and 2). The left deep femoral artery was repermeabilized by collaterals from the left internal iliac artery (IIA), via lumbar collaterals. On the right side, atherosclerotic stenosis of the superficial femoral artery and occlusion of the right external iliac artery were present.

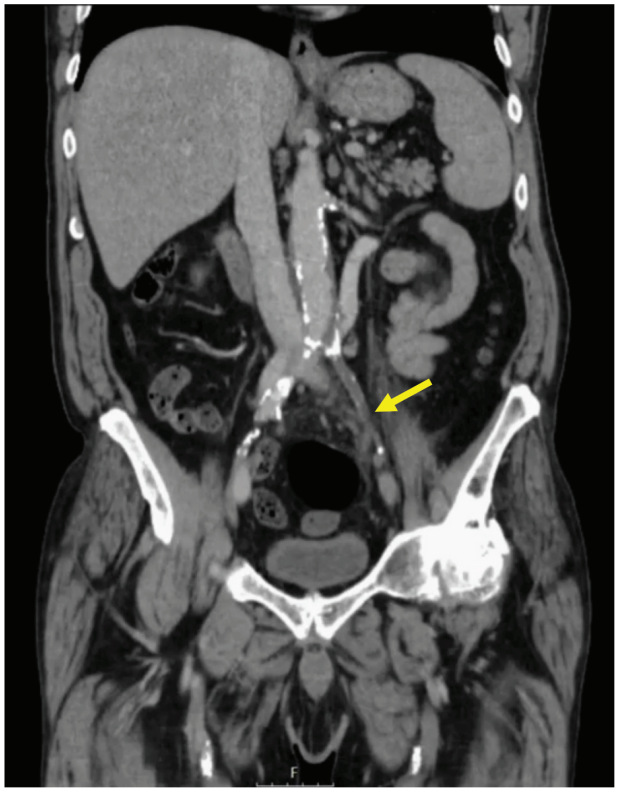


Figure 1. *CTA scan (axial view) showing total occlusion of the aortobifemoral stent's left component (arrow).*

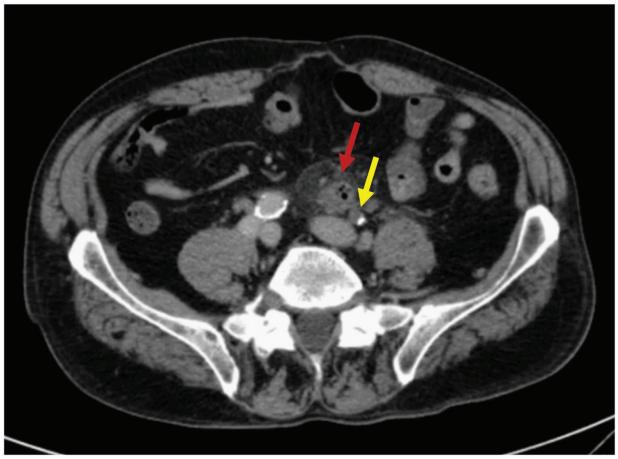


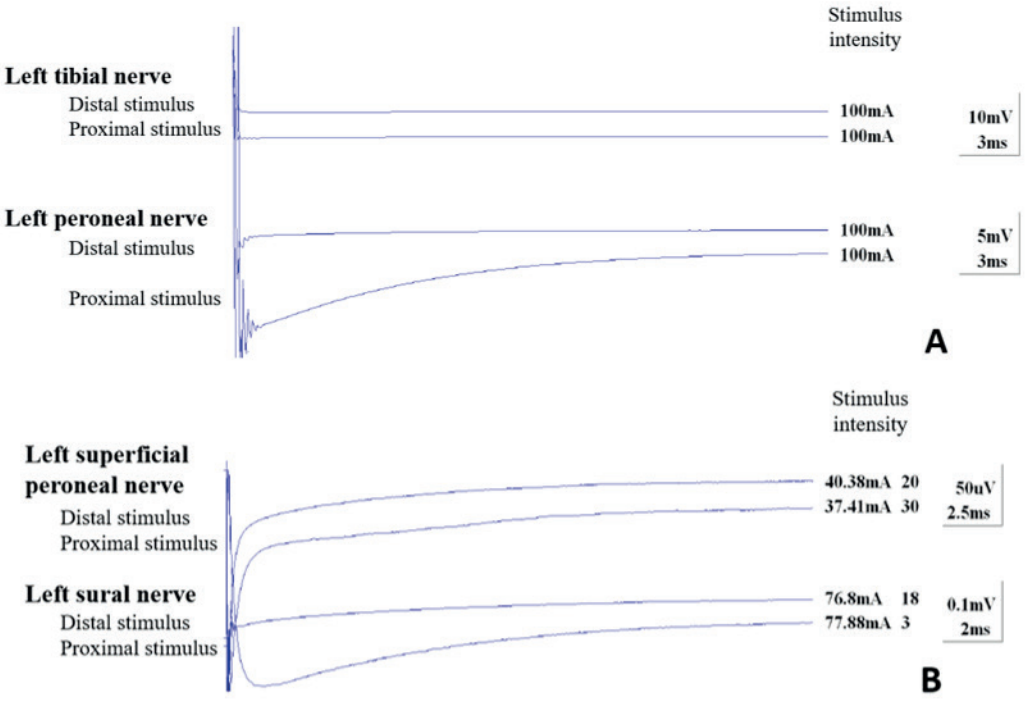
Figure 2. *CTA scan (coronal view) showing total occlusion of the left CIA (yellow arrow), as well as a local inflammatory/infectious process in its length (red arrow).*

After antibiotic treatment and surgical correction, the patient developed critical ischemia of the right lower limb (RLL), needing deep femoral artery thrombectomy and a bypass between the common iliac artery (CIA) and the right common femoral artery. One month later, in February, the patient was discharged. He was stable, with no pain, but presented an overall symmetrical weakness of all limbs (Medical Research Council [MRC] scale grade 3), with reduced tendon reflexes, in the context of Intensive Care Unit-Acquired Weakness. The remaining neurological examination was normal.

In April, he was once again admitted to the ED, this time in hemorrhagic shock due to abundant hematochezia secondary to an aorto-enteric fistula. He complained of pain and LLs weakness upon admission. After emergency surgical correction, an axillobifemoral bypass was performed. The next day, acute ischemia (grade IIb, according to Fontaine's Classification) of both LLs was noted, demanding urgent thrombectomy of the left branch of the axillobifemoral bypass. An infragenicular femoropopliteal bypass was performed in both LLs. Despite early revascularization, the patient developed severe ischemia of the right forefoot, leading to a transmetatarsal amputation. At this point, the patient presented motor and sensory deficits in the LLs and a few urinary incontinence episodes - difficult to characterize during the postoperative postoperative period. Upper limb examination was normal. Despite rehabilitation treatment during the hospital stay, his deficits worsened over time. At discharge, in mid-August, he presented asymmetric flaccid paraparesis (MRC scale grade 0 globally in the RLL, left knee and left foot; grade 2 in the left hip), absence of tendon reflexes, distal bilateral hypoesthesia in the LLs (sensory level L4/L5), saddle anesthesia and urinary and anal incontinence. Neurological examination of upper limbs and respiratory function were normal. These findings were suggestive of SC injury. Nevertheless, lumbosacral MRI (T2 sequences with fat saturation, STIR sequences, and T1-weighted sequences with intravenous contrast) excluded this hypothesis, as well as lumbar and sacral roots compression. EMG of the LLs (Figure 3) revealed the absence of sensory and motor nerve action potentials in the LLL (it was not possible to study nerve conduction in the RLL due to the aforementioned amputation and bandaging of the right foot), as well as signs of denervation in all muscles examined (myotomes L2 to S2) in both LLs, suggestive of bilateral lumbosacral plexopathy. Concomitant presence of left paravertebral L4 muscles denervation signs also evoked a possible root lesion, leading to the diagnostic hypothesis of radiculoplexopathy. The diagnosis of bilateral LSRP of ischemic etiology was then assumed.

Discussion

LSP arises from the L1-S4 nerve root, with a variable contribution from the T12 nerve root, and is responsible



Muscle (inervation)	Interpretation	Fib	PSW	Voluntary activity			
				Amp	Dur	Pol	IP
Left paravertebral (L4)	Fib, 0 activ	3/10	4/10				
Left medialis gastrocnemius (tibial, S1 S2)	Complete denerv	7/10	7/10				---
Right medialis gastrocnemius (tibial, S1 S2)	Complete denerv, 0 activ	5/10	5/10				
Left tibialis anterior (deep peroneal, L4 L5)	Complete denerv	7/10	7/10				---
Right tibialis anterior (deep peroneal, L4 L5)	0 activ	7/10	7/10				
Left bíceps femoris (tibial, L5 S1 S2)	Complete denerv	6/10	6/10				
Left vastus lateralis (femoral, L2 L3 L4)	Partial acute denerv	5/10	5/10	N	N	+	---
Right vastus medialis (femoral, L2 L3 L4)	Complete denerv	10/10	10/10				

C

Figure 3. *EMG showing absence of sensory nerve action potentials and motor unit action potentials in the LLL and abundant signs of denervation in all muscles examined (myotomes L2 to S2) in both LLs, as well as scarce signs of denervation in the left paravertebral L4 muscle. (A) Motor nerve study: left tibial and left peroneal nerves potentials were not obtained (even with potential uptake in the left tibialis anterior muscle). (B) Sensory nerve study: no potentials were obtained from the left superficial peroneal and left sural nerves. (C) Needle EMG: abundant fibrillations and positive waves were present in all evaluated muscles at rest (except in the left paravertebral L4 muscle, where its presence was scarce); in activity, it was only possible to activate the left vastus lateralis muscle, where polyphasic potentials were evident, with a marked decreased recruitment. Fib = fibrillation potentials; PSW = positive sharp waves; Amp = amplitude; Dur = duration; Pol = polyphasicity; IP = interference pattern; activ = activation; denerv = denervation.*

for sensory and motor functions to the ipsilateral LL and pelvic girdle. It is located in the posterior part of the psoas major muscle. The vasa nervosa to the plexus are derived from the blood supply to this muscle, which includes five lumbar arteries on each side of the abdominal aorta, the deep circumflex iliac artery, a branch of the external iliac, and the iliolumbar and gluteal branches of the IIA [4-6,8,9,11]. Blood flow through these arteries may be impaired due to aortic lesion, or it may be sacrificed during surgery involving vascular clamping, leading to ischemic LSP lesions [4-6,8,9,11]. Although branches of the IIA anastomose widely in the pelvic area, in patients with advanced POAD iliac artery occlusion can culminate in pelvic ischemia, even in the presence of an apparently patent contralateral IIA [7,9-12].

The overall neurological risk of endovascular and open abdominal aortic surgery is low, ranging between 0% and 1%. The risk is higher for emergency cases (1.4%-2%)

comparing to elective cases (0.1%-0.2%) and may result in SC, lumbosacral, and peripheral nerve lesions [4,8,9,12]. Several possible causes have been reported, such as interference with pelvic blood supply, prolonged aortic or suprarenal cross clamping, intraoperative hypotension, thromboembolic phenomena or a combination of these factors [6,7]. LSP lesions have been rarely reported as a complication of aortoiliac vascular procedures or disease [9-12].

According to the literature, SC, cauda equina, and LSP involvement are often difficult to distinguish [9,10]. Lumbosacral plexopathy can mimic an "incomplete" SC infarction, presenting with some of the following deficits: flaccid paraplegia, often progressing to an upper motor neuron picture; loss of pain and temperature sense with preserved deep sensation and proprioception; bowel and bladder dysfunction; variable electromyographic findings, depending on whether the lesion is complete or not [10]. EMG typically shows an asymmetrical reduction of the amplitudes of both compound action potentials and sensory nerve action potentials caudally to the lesion (including paraspinal muscles) [10]. Conduction velocities are usually normal or only mildly reduced. A prolonged late response (F wave) may be present on the affected side. Fibrillation potentials and neurogenic motor unit potentials in muscles involved by multiple lumbosacral levels, including paraspinal muscles, are also present, with normal or only mildly reduced conduction velocities [1]. Etiology and accurate location of the lesion require neuroimaging and electrophysiologic studies [1,3].

Based on the distal SC and lumbosacral nerve roots neurovascular anatomy and the clinical presentation, Gloviczki et al. [6] identified six different types of neurologic ischemic injury with prognostic value. Differentiating them can be difficult and requires careful neurologic evaluation followed by MRI (or myelography) and EMG. Type I lesions involve complete infarction of the distal SC and conus. Type II lesions correspond to an anterior spinal artery syndrome. Type III lesions are due to bilateral root ischemia with or without patchy distal cord and conus ischemia. Type IV injury results from unilateral plexus ischemia caused by infarction of the lumbar or sacral plexus. Both types III and IV have a favorable prognosis. Type V injury shows segmental SC infarction. Type VI injury occurs due to posterior spinal artery infarction [2,5,6,9].

LL weakness can be a presenting feature of different vascular syndromes affecting the SC and lumbosacral roots or plexus [10]. The clinical manifestations of these syndromes depend on the anatomical site and extent of vascular injury [2]. Progressive LL weakness related to lumbosacral plexopathy due to chronic abdominal aortic atherosclerotic occlusion is uncommon [8]. In our patient, the subacute onset of his deficits after a major endovascular procedure, along with his previous history of POAD requiring many vascular interventions, pointed to a vascular etiology. Facing a clinical picture of flaccid paraparesis alongside with sphincter incontinence, an SC infarction appeared to be the most likely diagnosis, but lumbosacral MRI failed to identify any sign suggestive of SC injury. Disruption of blood flow to the LSP (consistent with type III injury) due to major endovascular procedure, in a patient with advanced POAD, seems to be the most likely cause. The aortoiliac occlusive disease has to be taken into consideration as a possible contributor to this lesion. Once this unexpected severe complication occurs, careful neurologic evaluation should be done to localize the lesion and aid the prognosis [6].

In this case, one shall also consider Critical Illness Polymyoneuropathy (CIP) in the differential diagnosis list, as the patient had been hospitalized for several months, due to multiple surgical procedures and some medical complications. However, as the patient's upper limbs and respiratory muscles were not affected, and the flaccid paraparesis was asymmetrical, CIP seems to be an unlikely etiology. Also, CIP wouldn't explain sphincters incontinence [13]. Despite that, this entity was probably one of the secondary diagnoses, and it might have contributed to the deficits worsening.

The management of LSRP varies according to the etiology, but pain management and intensive rehabilitation program remain the mainstay in most cases [1,6].

Conclusion

Ischemic LSRP remains a rare, unpredictable, and devastating complication of aortoiliac procedures. Our case highlights the need to consider an ischemic etiology in the differential diagnosis of peripheral nerve injuries, especially if previous POAD is reported. As critical LL ischemia can be detected in an early stage, increasing clinicians' awareness of this entity can prevent the establishment of nerve damage due to a potentially reversible cause.

What is new?

Ischemic LSRP remains a rare, unpredictable, and devastating complication of aortoiliac procedures, which may cause paraparesis. Our case highlights the need to consider an ischemic etiology in the differential diagnosis of peripheral nerve injuries. As critical LL ischemia can be detected in an early stage, increasing clinicians' awareness of this entity can prevent the establishment of nerve damage due to a potentially reversible cause.

List of Abbreviations

CIA	Common iliac artery
CIP	Critical Illness Polymyoneuropathy
ED	Emergency Department
EMG	Electromyography
IIA	Internal iliac artery
LL	Lower limb
LLL	Left lower limb

LLs	Lower limbs
LSP	Lumbosacral plexus
LSRP	Lumbosacral radiculoplexopathy
MRC	Medical Research Council
MRI	Magnetic resonance imaging
POAD	Peripheral obstructive arterial disease
RLL	Right lower limb
SC	Spinal Cord

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Conflict of interests
The authors declare that there is no conflict of interest regarding the publication of this article.

Consent for publication
A written informed consent to publish this case was obtained from the patient and any accompanying images.

Ethical approval
Ethical approval is not required at our institution for publishing an anonymous case report.

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Summary of the case

1	**Patient (gender, age)**	Male, 66 years-old
2	**Final diagnosis**	Bilateral ischemic LSRP
3	**Symptoms**	Asymmetric flaccid paraparesis, absence of tendon reflexes and distal bilateral hypoesthesia in the LLs, saddle anesthesia, urinary and anal incontinence
4	**Medications**	Antibiotherapy
5	**Clinical procedure**	• Diagnostic: abdominal and pelvic CTA scans, lumbosacral MRI, EMG of the LLs • Treatment: multiple endovascular procedures to the LLs, transmetatarsal amputation to the right foot
6	**Specialty**	Neurology, vascular surgery