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Case Report

Bilateral Ischemic Lumbosacral Plexopathy Presenting as Acute Paraparesia Due to Vascular Graft Occlusion in a Patient With Leriche Syndrome

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Abstract

Acute ischemia of peripheral nerves generally results from occlusion of a main proximal limb artery or from occlusion of many distal arteries. The rareness of such neuropathies can be explained by the relative resistance of peripheral nerves to ischemia. Leriche syndrome is an aortoiliac occlusive disease that is known to cause neurologic complications such as ischemic neuropathy and spinal cord ischemia. Here, we present a man aged 55 years with bilateral lumbosacral plexopathy that presented as acute paraparesia due to thrombotic occlusion of an axillobifemoral graft, which was performed for the treatment of Leriche syndrome.

Keywords: Ischemia, lumbosacral plexopathy, Leriche syndrome

Leriche Sendromlu Bir Olguda Vasküler Greft Oklüzyonu Sonucu Akut Paraparezi Şeklinde Prezente Olan bilateral İskemik Lumbosakral Pleksopati

Özet

Periferik sinirlerin akut iskemisi, genellikle ana proksimal arterin ya da distal arterlerin oklüzyonu sonucu gelişir. Bu nöropatilerin nadir görülmesi, periferik sinirlerin iskemiye dirençli olması ile açıklanabilir. Leriche sendromu, aortailiak oklüzyon sonucu gelişen bir hastalıktır ve iskemik nöropati, spinal kord iskemisi gibi nörolojik komplikasyonlara neden olabilir. Biz bu yazıda, 55 yaşında Leriche sendromu tanısı nedeniyle aksillofemoral greft uygulanan ve greft oklüzyonu sonucu akut paraparezi ile prezente olan bilateral lumbosakral pleksopatili bir olgu sunmaktayız.

Anahtar Kelimeler: İskemi, lumbosakral pleksopati, Leriche sendromu

INTRODUCTION

The lumbosacral plexus, composed of T12-S4 roots, is protected among the deep muscle layers in the retroperitoneal area. As such, lesions of the lumbosacral plexus are rarely seen. Differential diagnostics of lumbosacral plexopathy (LSP) include metabolic, oncologic, inflammatory, ischemic, and autoimmune disorders. The main blood supply of the lumbosacral plexus is from the internal iliac artery and lumbar arteries. Ischemic lumbosacral plexopathy is not common owing to its rich blood supply.

Leriche syndrome is an aortoiliac occlusive disease and its main risk factors are hyperlipidemia, hypertension, diabetes mellitus, and smoking. It is known to cause neurologic complications, as with other peripheral vascular diseases. Akhaddar et al. reported a case of Leriche syndrome that caused acute paraplegia with spinal cord ischemia due to reduced blood flow of

Adamkiewicz's artery. Zankl et al. (1) presented a patient with paresis of both legs (2).

Here, we report a very rare case of bilateral ischemic lumbosacral plexopathy that presented as acute paraparesis due to thrombotic occlusion of a axillobifemoral graft, which was performed in a patient with Leriche syndrome. We think that this is the only case reported in the literature that resulted from vascular graft occlusion.

CASE PRESENTATION

A man aged 55 years presented to our emergency department with persistent painful muscle weakness of the lower extremities that began suddenly 2 days ago. He also described bilateral leg claudication, which he had had for 6 months. He had diabetes, hyperlipidemia and peripheral vascular disease. Axillobifemoral bypass surgery was perfomed 5 years ago because of an aortoiliac occlusion. He was taking oral antidiabetics, insulin, acetylsalicylic acid, and warfarin. On physical examination in the emergency department, left-sided dorsalis pedis, posterior tibial and popliteal arterial pulses were non-palpable and pulses on the right side were weakly palpable. With these findings, a vascular etiology was suspected and computerized tomography angiography revealed occlusion of the axillobifemoral graft. He was taken into surgery urgently and a axillobifemoralthrombectomy was performed. After the operation, ischemic findings on the left side did not regress so he was taken into a second operation on the same day and left femoro-popliteal graft bypass surgery was performed.

After the operations, although vascular insufficiency was treated, weakness was persistent in both lower extremities. The patient was referred to us two days after the operations due to persistantparaparesis. On his neurologic examination, muscle power in his bilateral hip flexors-extensors and knee extensors-flexors were grade 3, grade 2 to 1 in bilateral ankle planter flexors, and grade 1 in bilateral ankle dorsiflexors. There was no weakness in the upper extremities and sphincter muscles of the bladder or bowel. Sensory function tests showed decreased pain sensation below the bilateral inguinal area with glove-like sensory loss in the upper extremities. Deep tendon reflexes were absent in the upper and lower extremities and Babinski's sign was absent.

Magnetic resonance imaging (MRI) of the lumbosacral cord revealed no specific lesions that could have caused the sypmtoms (Figure 1). Motor and sensory nerve conduction studies were performed only in the right lower extremity because of diffuse edema of the left side. On the 7th day postoperatively, the sural nerve was not evoked and decreased compound muscle action potentials of the fibular and posterior tibial nerve was detected with normal nerve conduction velocity. Upper extremity nerve condution studies were normal. Needle electromyography (EMG) revealed abnormal spontaneous activities and severely reduced motor unit potential during volitions in the tibialis anterior and gastrocnemius. No pathologic activity was found and motor unit configuration was normal in the vastus medialis. These results suggested axonal degeneration of the sciatic nerve. To clarify the etiology, MRI of the hip was obtained. On imaging, thickening and edema of the right sciatic nerve was detected (Figure 2) and focal sciatic nerve injury was thought to be present in the right extremity. Follow-up EMG was planned to be performed after 3 weeks because it had not been performed on the left lower extremity.

Follow-up motor and sensory nerve conduction studies showed the same findings in the right lower extremity. On the left side, sural, posterior tibial, and fibular nerves were not evoked. Abnormal spontaneous activities and reduced motor unit potential during volitions were detected using needle EMG in bilateral adductor magnus, adductor longus, gluteus

maxiumus, tibialis anterior, and gastrocnemius. Needle EMG studies of the paraspinal muscles revealed no pathology. Nerve conduction studies of the upper extremities were also normal. In light of these findings, the clinical picture was diagnosed as bilateral lumbosacral plexopathy. Developing in a short time with the patient's vascular pathology, ischemia was considered the main etiologic factor. During clinical follow-up, a full rehabilitation program was conducted but no improvement was observed in the muscle strength.

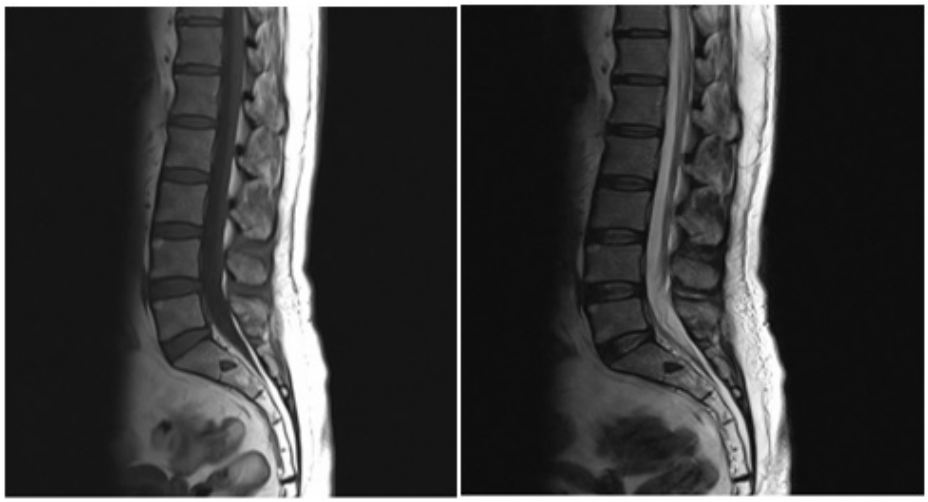

Figure 1: *There is no pathology on T2 weight lumbosacral MRI*

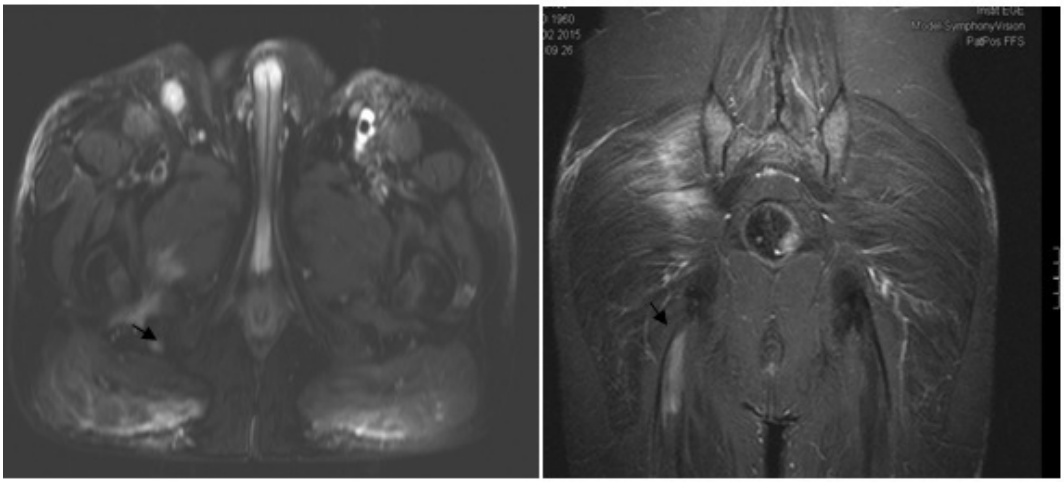

Figure 2: *Thickening and edema of the right sciatic nerve seen as hyperintense on hip MRI (black arrow).*

DISCUSSION

Lower limb weakness can be a presenting feature of various vascular syndromes that affect the spinal cord, lumbosacral roots or plexus. The clinical manifestations of these syndromes depend on the anatomic site and extent of vascular injury. Anatomically, the lumbosacral plexus is supplied by five lumbar arteries, which originate from the abdominal aorta, the deep circumflex iliac artery, a branch of the external iliac artery, and the iliolumbar and gluteal branches of the internal iliac artery (3). Acute ischemia of peripheral nerves generally results from occlusion of a main proximal limb artery or from occlusion of many distal arteries. The rareness of such neuropathies can be explained by the relative resistance of peripheral nerves to ischemia. It results from their slight metabolic needs (oxygen requirement of a human nerve = 0.3 mL/100 g) and from their rich vascularization, which provides a generous blood flow (43 mL/100g/min) (3). We think that the occlusive disease of the aorta and iliac artery, which are main sources of blood flow for the lumbosacral plexus, put our patient at risk in terms of ischemia, and occlusion of the graft triggered the ischemia. Although diabetes causes chronic neuropathy by affecting the small arteries of nerves, it could be another facilitating factor for ischemia in our patient because he had been diabetic for 12 years.

Acute bilateral lumbosacral plexopathy(LSP) secondary to aortoiliac occlusive disease is a rare presentation and can mimic an acute spinal cord or cauda equina syndrome. In our patient, this clinical entity was not the direct result of aortoiliac occlusion. It was observed due to thrombotic occlusion of the graft. MRI must be performed to eliminate a spinal pathology, and in our patient, MRI findings did not support a spinal injury. EMG is another diagnostic tool, and at the beginning, EMG findings were compatible with a focal right-sided sciatic injury. We also detected focal sciatic edema on the hip MRI. The left lower extremity was not evaluated initally with EMG because of leg edema and we could not explain the paraparesis by a right-sided sciatic injury. Therefore, we repeated EMG and nerve conduction studies to detect nerve injury on the other side. In acute nerve injury, follow-up needle EMG performed 2-3 weeks after onset can show spontaneous denervation potantials, we thought that this could give an idea about the extent of the injury. In the second EMG, nerve conduction studies and needle EMG findings showed bilateral obtural, femoral, and sciatic nerve injury, thus, with the clinical findings we diagnosed bilateral lumbosacral plexopathy. In the literature, there is only one case of Leriche syndrome that presented as focal sciatic neuropathy (4). Our EMG findings at the beginning also suggested sciatic neuropathy and MRI supported the sciatic nerve injury. We thought that this finding was related to nerve ischemia.

A number of structural and non-structural pathologies can present with LSP, the common ones include diabetic and idiopathic lumbosacral radiculoplexus neuropathy, which usually manifests with a subacute, asymmetric, progressive lower limb syndrome associated with weight loss. Other causes of LSP include neoplasia, infection, trauma, radiation, pregnancy, surgery, retroperitoneal hematoma, aneurysms of the abdominal aorta and its distal branches, vasculitis, and connective tissue disorders (5). The acute onset of bilateral lower limb weakness in our case with multiple vascular risk factors argue in favor of a vascular etiology.

In conclusion, we presented a patient with acute paraparesia who had aortoiliac occlusive disease that had been treated surgically previously. It seemed noteworthy that ischemic lumbosacral

plexopathy might develop in patients with previous vascular surgery when vascular findings re-emerged as a result of thrombotic graft occlusion. Therefore, this complication should also be considered if paresia persists after effective surgical treatment.

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