

Bilateral ischemic lumbosacral plexopathy from chronic aortoiliac occlusion presenting with progressive paraplegia

Hyangkyoung Kim, MD, PhD,^a Si Hyun Kang, MD, PhD,^b Don-Kyu Kim, MD, PhD,^b
Kyung Mook Seo, MD, PhD,^b Tha Joo Kim, MD,^b and Joonhwa Hong, MD, PhD,^c *Seoul, Korea*

Spinal cord ischemia is rare but causes significant morbidity and mortality. Spinal cord ischemia has been reported after open and endovascular interventions of the thoracic and abdominal aorta, and, rarely, acute occlusion of aorta from in situ thrombosis or acute embolic occlusion. Acute interruption of the critical blood supply to the spinal cord or root contributes to this devastating neurologic deficit. However, gradually worsening lumbosacral plexopathy and consequent paraplegia related to chronic aortic occlusion is extremely rare. We present a case of a 58-year-old man with progressive lower limb paralysis from atherosclerotic aortoiliac occlusion without history of aortic surgery or evidence of thromboembolism. (*J Vasc Surg* 2014;59:241-3.)

The spinal cord is much less susceptible to atherosclerosis than the brain because of its complex blood supply and tendency toward developing collateral flow.¹ However, the highly variable blood supply of spinal cord may unpredictably lead to ischemic spinal cord injuries.² There is a growing body of literature on ischemic neurologic deficit with the rapid expansion of endovascular aortic intervention. Complex mechanisms are responsible for this devastating complication including underlying arteriosclerosis together with hypotension or interruption of critical feeding arteries.³ Although rare, sudden paraplegia had been reported with acute occlusion of native abdominal aorta from in situ thrombosis or embolic occlusion.⁴ However, progressive lower limb weakness from lumbosacral plexopathy related to chronic abdominal aortic atherosclerotic occlusion is even more uncommon. Here, we report a very rare case of bilateral ischemic lumbosacral plexopathy representing progressive paralysis sparing bladder and bowel function from aortoiliac occlusion without a history of major aortic surgery or other source of thromboembolism. We believe this to be the first reported case of atherosclerotic aortoiliac occlusion with lumbosacral plexopathy presenting as gradual paraplegia.

From the Departments of General Surgery,^a Physical Medicine and Rehabilitation,^b and Thoracic Cardiovascular Surgery,^c Chung-Ang University, College of Medicine.

This study was funded by a Chung-Ang University research grant in 2009 and 2010.

Author conflict of interest: none.

Reprint requests: Si Hyun Kang, MD, PhD, Department of Physical Medicine and Rehabilitation, Chung-Ang University, College of Medicine, Chung-Ang University Hospital, 224-1 Heuksuk-dong, Dongjak-gu, Seoul, Korea 155756 (e-mail: sihyun92@cau.ac.kr).

The editors and reviewers of this article have no relevant financial relationships to disclose per the JVS policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

0741-5214/\$36.00

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<http://dx.doi.org/10.1016/j.jvs.2013.04.008>

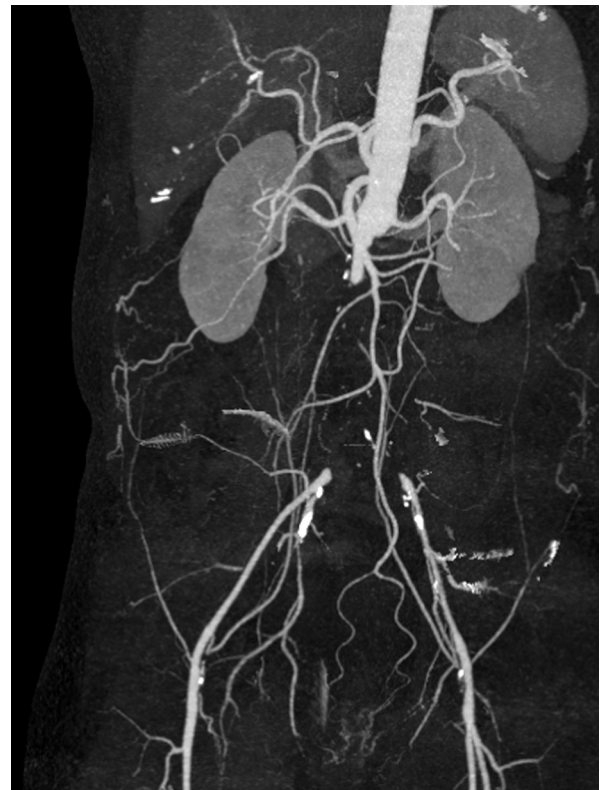


Fig 1. Results of computed tomography angiography. The abdominal aorta was occluded below the both renal arteries to the both common iliac arteries with abundant collaterals.

CASE REPORT

A 58-year-old man visited the emergency department with gradually worsening bilateral lower extremities weakness and decreased sensation since the initial occurrence 1 month earlier. He had been assessed with lumbosacral radiculopathy at a local clinic. At presentation, he was alert and cooperative, but incapable

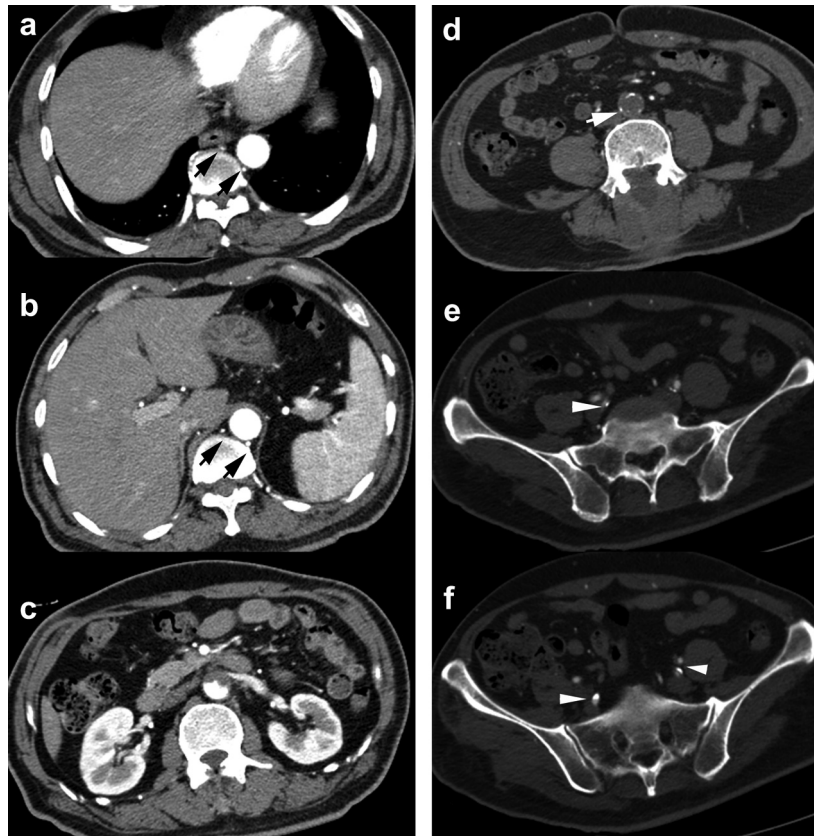


Fig 2. Computed tomography axial image of the aorta (a-d) and internal iliac arteries (e-f). Thoracic (a) and supraceliac aorta (b) and spinal arteries (black arrow) were patent. Infrarenal aorta was calcified (white arrow, d) and occluded. e and f, Distal common iliac arteries were reconstructed by collateral flow. The orifices of the both internal iliac arteries were patent, but tandem stenotic lesion with calcified atherosclerotic plaque (arrow head) was noted along the course.

of unassisted walking. He had no specific medical history including hypertension, diabetes, hyperlipidemia, or other cardiovascular diseases. He was not taking any medication and was not a smoker. Decreased sensation and weakness were noted in both legs, but no pain and tenderness. Otherwise, there was no weakness in the upper extremities, and facial and sphincter muscles of bladder and bowel.

On physical examination, muscle power on bilateral hip flexors and knee extensors were grade 3, and were grade 2 to 1 on bilateral ankle dorsiflexors, big toe extensors, ankle plantar flexors, knee flexors, and hip extensors. Sensory function tests showed decreased sensation below bilateral inguinal area, which was not precisely localized. Deep tendon reflexes were mildly decreased on both extremities and Babinski's sign did not exist.

The dorsalis pedis, posterior tibial, and popliteal arterial pulses on both sides were weakly palpable, and a nonhealing wound on left second toe tip, which became a critical clue to diagnosis, was found. Preoperative ankle-brachial indices were 0.62 and 0.58, respectively. Computed tomography angiography revealed occlusion of the infrarenal aorta to both common iliac arteries with abundant collaterals (Fig 1). Distal common iliac arteries were reconstructed by collateral flow (Fig 2). The orifices of both internal iliac arteries (IIAs) were patent, but tandem stenotic lesion with calcified atherosclerotic plaque was noted along the

course (Fig 2). Magnetic resonance imaging of the lumbosacral spinal cord did not reveal specific lesions, which could have caused the symptoms. On motor and sensory nerve conduction studies, all peripheral nerves on both sides including femoral, common peroneal, tibial, superficial peroneal, sural and saphenous nerves, and H-reflex and F-reflex were not evoked. Needle electromyography revealed abnormal spontaneous activities and severely reduced motor unit potential during volitions on bilateral iliopsoas, vastus medialis, tibialis anterior, peroneus longus, tensor fascia latae, gastrocnemius, biceps femoris short head and long head, gluteus medius, and gluteus maximus. Paraspinalis muscles could not be evaluated by needle electromyography because of poor positioning from weakness in his lower extremities. These results suggest rapidly aggravating peripheral polyneuropathy with severe axonotmesis involving the lower extremities only. At the same time, intravenous immunoglobulin was administered to rule out acute inflammatory demyelinating polyradiculoneuropathy, which was not found effective at all. Putting all of the results together, a clinical diagnosis of chronic aortic occlusion with ischemic lumbosacral plexopathy was made, and we decided to perform surgical revascularization.

On exploration, atherosclerotic occlusion from infrarenal aorta to common iliac arteries was revealed, and an aortobifemoral bypass

was performed using a bifurcated graft. Despite an uneventful recovery, the legs did not show signs of improvement immediately after the surgery. Postoperative ankle brachial index was improved to normal level of 0.99 and 0.98. On follow-up electromyography 5 months later, mild improvement of superficial peroneal and sural sensory nerve conduction and increase in recruitment on volition of several muscles on needle electromyography was observed. We conducted a serial physical examination with a proper rehabilitation program, including muscle strengthening exercise; the patient was able to walk with assistance 1 month after the surgery.

DISCUSSION

Ischemic pathology of the spinal cord secondary to vascular occlusion is not a common phenomenon.^{1,5-7} The pattern and degree of neurologic deficit are unpredictable and heterogeneous because of the variable blood supply to the cord. Myelopathy, lumbosacral radiculopathy, and cauda equina syndrome are the most commonly documented forms, especially related to the aortic interventions.^{3,8-13} Interruption of the artery of Adamkiewicz, lumbar arteries, or the IIAs during the procedure may impede the blood supply to the lower part of the spinal cord with ischemic neuropathy as a result.

Meanwhile, the lumbar plexus is embedded in the posterior part of the psoas major muscle, which receives a very abundant arterial supply.¹⁴ The vasa nervosa to the plexus are derived from the feeding artery of this muscle, in most cases the lumbar arteries. Also, various branches of the IIA, such as inferior gluteal artery via the arteria comitans nervi ischiadici and superior gluteal artery via the lateral sacral artery, feed the lumbosacral plexus.¹⁵ These rich collaterals usually protect the lumbar plexus from ischemic injury even in the event of occlusion of most or all of the lumbar arteries.

Our patient showed a classic manifestation of lumbosacral plexopathy of paraparesis including variable sensory loss, hyporeflexia, and dysesthesias without bladder or bowel symptoms.¹⁵ The electromyographic findings indicated mononeuropathy multiplex and axonotmesis. The etiology includes ischemic, demyelinating, infectious, and compression neuropathy, neoplastic or granulomatous infiltration, and other disorders, such as lumbosacral plexus neuropathy.⁷ Ischemia could be regarded as the most important etiology. However, to rule out acute inflammatory demyelinating polyradiculoneuropathy attributable to relatively acute clinical progression in neurologic perspectives and higher incidence, intravenous immunoglobulin was administered first; this was not effective at all. From the axonotmesis not demyelination, and the paralysis limited to the lower extremities, we could rule out other forms of demyelinating or systemic etiology. Unlike previous reports, there was no sign of acute limb occlusion including pain or pallor or evidence of other cause of aortic occlusion, such as connective tissue disorder, infection, or cancer. Assuming from the calcification around the aortoiliac arteries, degenerative change seemed to be responsible for chronic occlusion. Given that ischemic damage was confined to plexus level, this neurologic deficit was the result of atherosclerotic change, which involved the

end of the feeding arteries of lumbosacral plexus that could not be supplied sufficiently by collaterals. Notwithstanding the disappointing electromyographic results of minimal improvement, revascularization was necessary to discontinue the progression of the axonotmesis, to protect the sphincter, and to heal the toe wound.

CONCLUSIONS

We present a patient with chronic aortic occlusion with progressive paraplegia, a rare event in itself. It seemed noteworthy that ischemic lumbosacral plexopathy might develop in patients without previous history of aortic surgery or acute thromboembolic phenomenon. Small toe wound was the only diagnostic clue to link complex neurologic deficit to vascular occlusive disease without any evidence of critical limb ischemia or intermittent claudication.

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Submitted Mar 6, 2013; accepted Apr 10, 2013.