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CASE REPORT

Idiopathic lumbosacral plexitis

Yahia Imam[1], Dirk Deleu[1], Khaled Salem[2]

Address for Correspondence:
Yahia Imam
[1]Department of Neurology (Medicine), Hamad Medical Corporation, P. O. Box 3050, Doha, Qatar
[2]Department of Radiology, Hamad Medical Corporation, P. O. Box 3050, Doha, Qatar
Email: yahyazakaria200x@yahoo.com

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ABSTRACT

Idiopathic lumbosacral plexitis (LSP) or non-diabetic lumbosacral radiculoplexus neuropathy (LRPN) is a rare monophasic immune-mediated disorder. The disease is characterized by multiple lumbosacral root and nerve involvement in the absence of trauma, mass lesion or elevated blood sugar. We report a 60-year-old man presenting with a 4-day history of acute-onset sharp left hip pain radiating down his leg associated with numbness over the lateral dorsum of his left foot. MRI of the lumbosacral plexus revealed gadolinium enhancement of mainly L5 on the left. A dramatic response to oral steroids was observed.

Keywords: idiopathic lumbosacral plexitis, diabetic lumbosacral radiculoplexus neuropathy, steroids, magnetic resonance imaging

INTRODUCTION

Idiopathic lumbosacral plexitis (LSP) also known as non-diabetic lumbosacral radiculoplexus neuropathy (LRPN) is an idiopathic disorder characterized by the acute onset of severe leg pain followed by wasting and weakness of the leg muscles. The diagnosis requires clinical and electrophysiological demonstration of lesions affecting multiple nerves and root levels in the absence of other causes of lumbosacral plexopathy e.g. trauma, radiation, diabetes or mass lesions.[1] We report an MRI-documented case of idiopathic LSP, which to our knowledge is the first case to be reported in the Gulf region.

CASE REPORT

A 60-year-old man presented with a 4-day history of acute-onset sharp left hip pain radiating down his leg associated with numbness over the lateral dorsum of his left foot. The pain gradually worsened and prompted admission. He denied any prior episodes of similar pain, preceding febrile illness or immunization. Neurological examination revealed weakness of dorsiflexion of the left big toe and sensory loss to pin prick in dermatome S1 on the lateral aspect of the left foot. The deep

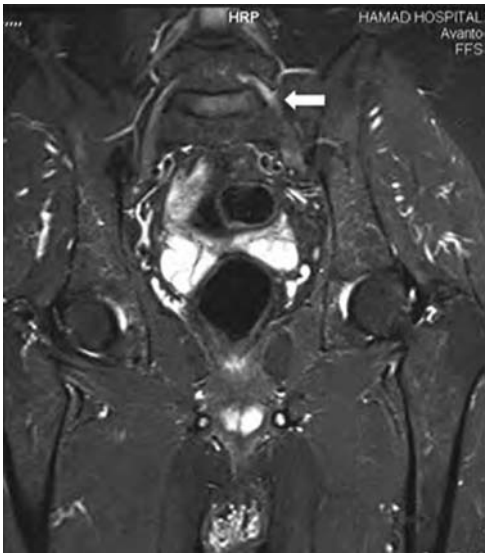

Figure 1. MRI coronal section Short Tau Inversion Recovery (STIR) sequence shows thicker and brighter left L5 nerve root (arrow) as compared with its right counter part.

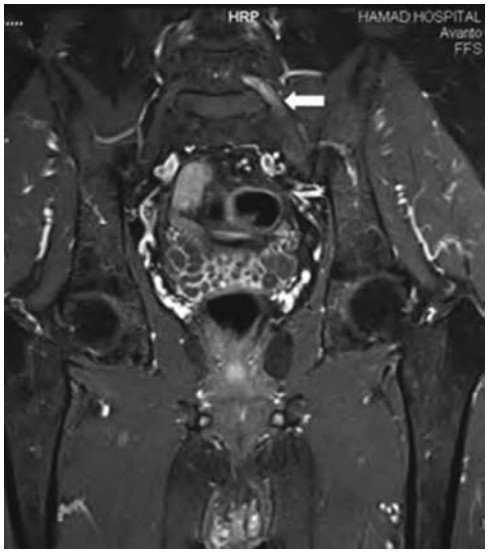

Figure 2. MRI coronal section post gadolinium contrast shows enhancement of the left L5 nerve root (arrow).

tendon reflexes were spared and Lase□gue sign was negative. The patient was bed-bound due to severe pain and required high-doses of morphine, diclofenac and pregabalin to achieve pain control. Complete blood count, erythrocyte sedimentation rate (ESR), chemistries, hemoglobin A1c, fasting blood sugar, a 2-hour glucose tolerance and autoimmune screen were normal. Lumbar puncture was declined. Neurophysiologic testing (day 10) showed absent left sural nerve action potential and mild acute signs of denervation in left L5 and S1 myotomes. Lumbosacral spine Magnetic Resonance Imaging (MRI) was normal. Lumbosacral plexus MRI is shown in Figures 1 – 3. The patient was started on prednisolone 60 mg daily (and gradually tapered) with dramatic reduction in pain and became ambulatory within two days. He was lost to follow up.

DISCUSSION

Lumbosacral plexitis (LSP) also known as non diabetic lumbosacral radiculoplexus neuropathy (LRPN) is less recognized than its diabetic counterpart.It shares the same clinical and pathological features with diabetic lumbosacral radiculoplexus neuropathy (DLRPN).[2] The one distinguishing feature between the two conditions being the absence of elevated blood sugar in the former. Lumbosacral plexitis (LSP) is an uncommon condition; while the incidence has not been studied it is postulated to be considerably less than one per 100,000 people.[1] It has been recognized relatively recently by Evans and others in the early 1980s[3] which probably reflects it rarity. Thereafter reports in the literature were limited to individual case reports and case series involving a relative small number of patient describing natural history and outcome and response to immunosuppressive therapy.[1]

In our case the patient age coincide with the peak age of incidence which 40 – 60 years.

The history is also typical with sudden onset severe unilateral pain followed by weakness and parathesias. The clinical exam demonstrated weakness of left big toe dorsiflexion (L5 root) and sensory loss of S1 dermatome in the foot. This is further collaborated by absence of the sural nerve sensory nerve action potential (SNAP).

Blood sugar and hemoglobin A1c was normal distinguishing it from diabetic lumbosacral radiculoplexus neuropathy.

MRI of the lumbosacral spine was done initially to rule out compressive pathology (i.e. disk herniation), a not uncommon scenario with some patient even undergoing unnecessary laminectomies.[4] Our patient had a normal

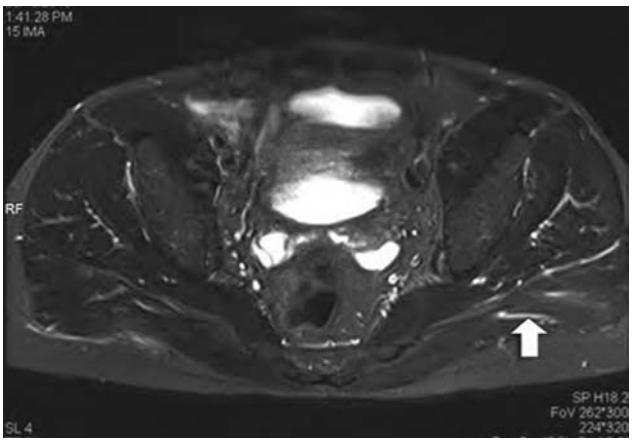

Figure 3. MRI axial STIR sequence shows brighter signal in left gluteal muscles (arrow) as compared with its right counter part indicative of denervation.

ESR in keeping with what Dyke and others have reported.[4] On the other hand, Bradley and others have described a group of patients with LSP and elevated ESR.[5] In this case neuroimaging was helpful in ruling out other diagnosis as well as visualizing enhancement within the lumberosacral plexus and supporting the diagnosis; in keeping with what Ishii et al and Cianfoni et al have described in LSP and DLRPN respectively.[6,7]

Initial reports describe prognosis as good with slow resolution of the condition over weeks or months depending on the degree of involvement of the plexus and weather sensory and motor impairments are both present.[1] However, subsequent papers have reported the possibility of a relapsing, progressive, and disabling course.[1,2,8]

This highlights the importance of early recognition and treatment as it may pose significant morbidity and suffering.

Although it is suggested that LSP and DLRPN share the strikingly similar pathogenesis namely microvasculitis,[2] there are no published randomized controlled trials employing immunotherapy to date.[9] This probably underscores the rarity of the condition.

Small series have reported response to intravenous immunoglobulin and steroids in high doses either alone or in combination, with the addition of azathioprine or cyclophosphamide or without.[1] Tarulli and Rutkove[1] suggested either "intravenous immunoglobulin at a dose of 0.4 g/kg per day for 5 days, repeated at monthly intervals, or 60 mg per day of prednisone for 1 to 2 months followed by a very slow taper" to be of benefit.

Our patient responded to 60 mg of prednisolone within 48 h which in view of his clinical radiological and neurophysiological picture may suggest a mild form of the disease as opposed to a more severe one.

CONCLUSION

LSP remains an uncommon and probably under diagnosed condition. It can cause considerable morbidity and suffering. A high index of suspicion is sometimes required. Neuroimaging is helpful in reaching a diagnosis.

Immunotherapy, particularly steroid might be helpful. Further well constructed studies are need before recommendations on treatment are drawn.

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