A Rare Disabling Complication of Diabetes Mellitus: Diabetic Lumbosacral Radiculoplexopathy

Diabetes Mellitusun Engelliliği Yol Açan Nadir Bir Komplikasyonu: Diyabetik Lumbosakral Radikülopleksopati

ABSTRACT Diabetes lumbosacral radiculoplexopathy (DLSRP) a rare complication of diabetes mellitus is a peripheral neuropathy presenting with hip and leg pain weakness of involved extremity and wasting of proximal muscles. As it is very rare sometimes it may be difficult to consider and diagnose this entity. Herein we report a 62-year old woman presented with lower extremity weakness and pain that had no known DM until she had admitted to emergency service with these symptoms. Upon clinical suspicion physical examination, laboratory and imaging tests; electromyographic studies revealed DLSRP. After immunomodulatory treatment and physical therapy program, the patient had marked improvement in symptoms and signs. DLSRP must be considered in patients presenting with acute hip and leg pain together with proximal weakness and must be differentiated from other pathologies.

Keywords: Diabetes mellitus; diabetic neuropathies; immunomodulation; rehabilitation

ÖZET Diabetes mellitusun (DM) nadir bir komplikasyonu olan diyabetik lumbosakral radikülopleksopati (DLSRP) kalça ve diz ağrıısı, tutulan ekstremitede güçsüzlük ve proksimal kaslarda zayıflık ile kendini gösteren bir periferik nöropatıdır. Çok nadir olarak görüldüğünden ayırıcı tani açıdan güç önünde bulundurmak ve tanı koymak zor olabilir. Biz burada acil servis altı ekstremitelere güçsüzüğü ve ağrıya başvuran ve o zamana kadar DM tanısı olmayan 62 yaşında kadın hastaya rastladık. Klinik şüpheye sonrası fizik muayene, laboratuvar ve görüntüleme testleri ve elektromiyografik çalışmaları DLSRP tanısı ortaya koydu. İmmünomodülatör tedavi ve fizik tedavi programı sonrası hastanın sempoiz ve bulgularında belirgin iyileşme görülüldü. DLSRP akut kalça veacak ağrıısı ile proksimal kas güçsüzüğü ile başvuran hastalarda akılda tutulmalı ve diğer patolojilerden ayırıcı tanı yapılmalıdır.

Anahtar Kelimeler: Diabetes mellitus; diyabetik nöropatiler; immünomodülasyon; rehabilitasyon

Diabetes Mellitus (DM) is one of the most common metabolic disorders. Worldwide prevalence of DM estimated 8.3% as per the Diabetes Atlas 2012. DM may be asymptomatic for many years and may present with its complications. Diabetic neuropathy, a microvascular complication of DM is defined as peripheral nerve dysfunction diagnosed by excluding other reasons such as genetic, traumatic, nutritional, infectious, toxic and neoplastic agents. Peripheral neuropathy prevalence is 30-50% in course of DM. Diabetic Lumbosacral radiculoplexopathy (DLSRP) is most common form of asymmetric peripheral neuropathies seen in DM and a rare disabling condition. DLSRP or so called diabetic amyotrophy is char-
acterized with acute hip pain, proximal muscle weakness and atrophy. It begins with severe uni-
lateral pain in the back, hip or thigh and can spread
to the other side within weeks to months.3,4 DLSRP
is usually seen among older patients especially over
age 50. The development of this neuropathy is
often not associated with glucose control or the du-
ration of the disease. In this report, a patient diag-
nosed with DLSRP is presented and discussed.

CASE REPORT

CLINICAL, LABORATORY AND RADIOLOGIC ASSESSMENT
A 62-year old patient with no known DM history
had suddenly developed lower extremity weakness.
She had admitted to emergency service and then
transferred to intensive care unit (ICU) with dia-
betic ketoacidosis. The patient had been followed
in ICU for six days and insulin and oral antidiabetic
treatment had been started. Her weakness had
spread to right side 2 weeks later. She had aching
and knife-like pain and numbness. She was ambu-
lated with wheelchair for 2 months. She had a his-
tory of essential hypertension for one year.
Physical examination findings were as follows: She
was cooperated and oriented. Blood pressure was
140/100 mm Hg. Pulse rate was 84/min. Cranial
nerves and upper extremity neurologic examina-
tion were within normal limits. Lumbar range of
motion was restricted at the end of range for ex-
tension and lateral flexion. Range of motions of
hips and knees were within normal limits. She had
atrophy in both thighs. According to Medical Re-
search Council (MRC) Scale muscle strengths of
hip flexors, right hip adductors, left hip adductors,
hip extensors, knee flexors and knee extensors
were 2/5, 2/5, 1/5, 4/5, 4/5 and 2/5 consecutively.
Strengths of other muscles were normal. Sensorial
examination was normal. Patella reflexes couldn't
be achieved. Achilles reflexes were normal. No pathologic reflex obtained.

LABORATORY AND IMAGING FINDINGS
Laboratory investigations including complete blood
count, erythrocyte sedimentation rate, C-reactive
protein, muscle enzymes, rheumatoid factor, vita-
min B12, thyroid hormone levels and tumor mark-
ers were within normal limits. Abdominal ultra-
sonography (USG) showed no abnormality except
grade 1 hepatic steatosis. Cranial magnetic reso-
nance imaging (MRI) detected nonspecific is-
chemic gliotic lesions in right thalamus, peri-
ventricular subcortical white matter and left
centrum semiovale. Lumbar MRI showed right
paramedian protrusion and central protrusions at
L2-3, L4-5 and L5-S1 levels respectively. No nerve
compression or spinal stenosis were found. Finally,
lower extremity arterial Doppler USG showed no
meaningful obstruction or stenosis.

ELECTROMYOGRAPHIC FINDINGS
Median, ulnar, radial, sural and superficial sensor-
ial conduction tests showed minimal slowing and
slight decrease in amplitudes. No sensorial action
potential was obtained in bilateral saphenous nerve
conduction test. Peroneal motor conduction tests
showed minimal slowing and slight decrease in am-
plitudes. Tibial nerve motor conduction tests were
normal. F-Wave studies of median, tibial and per-
oneal nerves were normal. Electromyographic
(EMG) investigation of left vastus lateralis and ad-
ductor magnus and right rectus femoris and ad-
ductor longus muscle showed spontaneous
denervation potentials and neurogenic motor unit
potentials. Left tibialis anterior, extensor digitorum
brevis, gastrocnemius medial head, biceps femoris
muscle short head and right vastus lateralis, ad-
ductor longus muscle needle EMG studies were
normal. Paraspinal muscle needle EMG studies
were normal. According to Storh's method com-
pound muscle action potentials for left side was
lower than right side.5 In the light of all these signs
and tests patient had accepted as diabetic amyotro-
phy with distal symmetric axonal sensorimotor
neuropathy and hospitalized into physical medi-
cine and rehabilitation clinic.

MEDICAL TREATMENT AND PROGNOSIS
During her stay in rehabilitation clinic oral antidi-
abetic medications and insulin doses were regu-
lated. 0.4 mg/kg intravenous immunoglobulin
(IVIG) treatment was administered to the patient
for 5 days. For neuropathic pain symptoms pregabalin treatment was started. Femoral nerve blockage was made with 3 cc lidocaine applied under 2.5 cm inguinal ligament and 2 cm lateral to femoral artery and transcutaneous electrical nerve stimulation (TENS) were applied for 30 minutes with conventional TENS mode. Neuromuscular electrical stimulation was applied to quadriceps femoris muscles with 0.3 ms frequency and 20 Hz. Physical rehabilitation program consisted of range of motion and strengthening exercises, balance and coordination exercises and ambulation training with walker was started. After treatment neuropathic pain was decreased from 8 to 4 on visual analog scale. Muscle strength of hip flexors and quadriceps increased to 4/5 according to MRC. She could ambulated with forearm crutches.

**DISCUSSION**

DLSRP syndrome usually affects type 2 diabetic patients and more frequently males over age 50 years. Although prefers type 2 diabetic patients, type 1 diabetic patients can also be affected.

Pathogenesis of DLSRP remains unclear. Both ischemic and metabolic causes had been accused. Hyperglycemia activates numerous metabolic pathways like polyol pathway, protein kinase C (PKC) pathway advanced glycation end products (AGE) pathway and hexosamine pathway and these pathways acts in mitochondrial reactive oxygen species (ROS) production resulting in neuroinflammation and vascular impairment leading to diabetic neuropathy. Nerve ischemia, inflammatory infiltration, and vasculitis are found in most severe forms of proximal diabetic neuropathy. Overall the distal symmetric polyneuropathy in diabetes is caused mainly by metabolic causes, the asymmetric manifestations are mainly of vascular origin. There is no biologic marker for DLSRP, however in some cases cerebrospinal fluid protein and erythrocyte sedimentation rate may be elevated.

The development of DLSRP is usually not dependent to control of blood glucose or duration of DM. DLSRP may be the first sign of DM like in our report. This rare complication of DM usually begins unilaterally with severe pain in the back, hip or thigh and may spread to other extremity. Patients usually complain about standing up from sitting position or chair due to weakness. Sensorial loss may be seen and distal symmetric sensorimotor neuropathy may accompany.

Sometimes it may be very difficult to diagnose DLSRP especially spinal stenosis and lumbar disc herniation must be ruled out to prevent unnecessary surgery. The most important misdiagnosis is L4 syndrome, which may lead to unnecessary surgery. Co-existence lumbar disc herniation, spinal stenosis and DLSRP have been reported. Dermatomal pain and hypoesthesia are characteristics of lumbar disc diseases but weakness is prominent in DLSRP. Also bilateral weakness is rare in lumbar disc pathologies. In diabetic patients insulin neuritis is another challenging condition leading to reversible acute severe distal limb pain, peripheral nerve fiber damage and autonomic dysfunction, preceded by a period of rapid glycemic control.

Clinical course of DLSRP is variable. Progression of disease varies from 2-18 months. Also recovery or improvement may prolong up to 4 years. DLSRP usually has a good prognosis; but overall symptomatic improvement is rare; residual symptoms such as fatigue, residual weakness, walking difficulties may be seen.

Management of DLSRP is focused on pain and glycemic control. However, the role of glycemic control on prognosis is doubtful. Active physiotherapy and exercises with assistant of orthoses if needed were advised. For pain relief neuropathic pain agents may be used. We achieved a good pain relief with pregabalin and TENS for our patient. There is no effective treatment modality for DLSRP due to lack of clinical trials. The immunomodulatory therapy was first described by Bradley who had used prednisone and cyclophosphamide for five patients and four patients had improved with treatment. Kendel et al, was used immunotherapy with different immunomodulatory drugs in 21 patients with diabetic neuropathy, in all patients in both groups worsening of neuropathy stopped and
improvement started with beginning of the treatment.  

Intravenous immunoglobulin (IVIG) treatment was also used in another case reports and good outcomes were achieved in the literature. We also used IVIG treatment for our patient as 0.4 mg/kg/day for five days and acquired clinical improvement although complete resolution of disease couldn’t be achieved.

For neuropathic pain, narcotic analgesics, tricyclic antidepressants, selective noradrenaline re-uptake inhibitors (SNRI), gabapentin and pregabalin can be used. As a conclusion in patients with acute hip pain and proximal weakness DLSRP must be considered and differential diagnosis must be made. MRI and ENMG may be helpful to rule out lumbar disc diseases and to make diagnosis. Early diagnosis may decrease mortality and may prevent or decrease disability. IVIG and other immunomodulatory may be used for DLSRP. Neuropathic treatment agents may be used for pain relief. Physical therapy and rehabilitation program may improve walking and weakness.

**Conflict of Interest**

Authors declared no conflict of interest or financial support.

**Note:** Informed consent form was obtained from the patient

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**REFERENCES**