OLGU SUNUMU CASE REPORT

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Diabetic Amyotrophic Neuralgia

Diyabetik Amyotrofik Nevralji

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ABSTRACT Amyotrophic neuralgia is an idiopathic and inflammmatory neuropathy characterized by acute-subacute onset of pain and weakness. It can rarely occur in diabetic patients as a form of diabetic neuropathy. In this study, we aimed to present clinical, electromyographic and radiological features of a case with diabetic amyotrophic neuralgia. A 58-years-old female patient was admitted to our department with low back pain. Her medical history revealed that the pain had started one month ago and radiated from the low back to the bilateral hips and left lower extremity significantly. Visual Analogue Scale (VAS) score of low back pain was 100 mm. She also reported that she had the diagnosis of diabetes mellitus for score ten years. On physical examination; the movements of the lower back were decreased and painful. The muscle strength of lower extremities were 1+/5 proximally and 2+/5 distally. Electromyographic assesment was compatible with sensory-motor axonal polineuropathy on lower extremity and diabetic amyotrophic neuralgia. A medical treatment consisting of pregabalin as 150 mg daily was administered. Severity of low back pain decreased to 10 mm after the treatment. Clinicians should take into consideration this pathology in patients with diabetes mellitus, pain and weakness. Electromyography and Magnetic Resonance Imaging (MRI) findings can be useful in differential diagnosis.

Keywords: Amyotrophic neuralgia; diabetes mellitus; pain

ÖZET Amyotrofik nevralji, akut-subakut başlangıçlı ağrı ve güçsüzlük ile karakterize idiyopatik ve inflamatuar bir nöropatidir. Bu durum, diyabetik hastalarda diyabetik nöropatinin nadir bir formu olarak ortaya çıkmaktadır. Bu çalışmada diyabetik nevralji tanısı koyulan bir hastanın klinik, elektromiyografik ve radyolojik özelliklerini sunmayı amaçladık. Elli sekiz yaşında bayan hasta merkezimize bel ağrısı ile başvurdu. Hastanın anamnezi alındığında; ağrının bir ay önce başladığı ve her iki kalça ve sol alt ekstremiteye yayılım gösterdiği öğrenildi. Vizüel Analog Skalaya (VAS) göre bel ağrısının şiddeti 100 mm olarak ölçüldü. Hasta aynı zamanda 10 senedir diabetes mellitus tanısı olduğunu belirtti. Fizik muayenede, bel hareketleri kısıtlı ve ağrılı idi. Alt ekstremitelerin kas gücü proksimalde 1+/5, distalde ise 2+/5 olarak değerlendirildi. Hastanın elektromiyografik değerlendirmesi alt ekstremite sensöri-motor aksonal polinöropati ve diyabetik amyotrofik nevralji olarak raporlandı. Hastaya 150 mg/gün pregabalin tedavisi başlandı. Tedavi sonrası değerlendirmede bel ağrısı şiddeti VAS değeri 10 mm'ye düştü. Klinisyenler diyabet tanılı ağrı ve güçsüzlüğü olan hastalarda bu patolojiyi göz önünde bulundurmalıdır. Ayırıcı tanıda elektromiyografik incelemeler ve Magnetik Rezonans Görüntüleme (MRG) yarar sağlayabilmektedir.

Anahtar Kelimeler: Amyotrofik nevralji; diabetes mellitus; ağrı

Diabetes mellitus is a chronic disease which is associated with the impairment in insulin secretion, insulin resistance, abnormal carbohydrate metabolism and hyperglysemia.[1] The complications affecting the eyes, kidneys, blood vessels and nerves can occur in patients with particularly long term diabetes mellitus.[2] Diabetic neuropathy is a nerve-damaging complication of diabetes mellitus that may develop as a result of diabetic micro-vascular injury in small blood vessels that supply

blood to nerves and macro vascular circumstances.[2] Diabetic neuropathy has many forms including distal symmetric sensori-motor polyneuropathy, autonomic neuropathy, proximal motor neuropathy and focal neuropathies.[2]

Diabetic amyotrophy is a rare, idiopathic and inflammmatory neuropathy which is also known as proximal diabetic neuropathy, Bruns-Garland syndrome or diabetic lumbosacral plexopathy.[2-8] It is characterized by acute or subacute onset of proximal-hip pain, proximal muscle weakness and atrophy.[8,9] This clinical entity is usually diagnosed with physical examination, laboratory tests and electrodiagnostic studies.[1] In this study, we aimed to present clinical, electromyographic and radiological features of a case with diabetic amyotrophic neuralgia.

CASE REPORT

A 58-years-old female patient was admitted to our department with low back pain. Her medical history revealed that the pain had started one month ago and radiated from the low back to the bilateral hips and left lower extremity significantly. Visual Analogue Scale (VAS) score of low back pain was 100 mm. She stated that her mobility was restricted by muscle weakness and pain. She also reported that she had the diagnosis of type II diabetes mellitus for ten years. The treatment of diabetes mellitus was initiated with metformin and insulin was added thereafter. On physical examination; the movements of the lower back were decreased and painful. The muscle strength of lower extremities were 1+/5 proximally and 2+/5 distally. Tendon reflexes were hipoactive and there was hypoesthesia in the left lower extremity. She was using wheelchair for ambulation. Transferring of the patient was partially dependent. There was also limitiation in her self- care activities such as dressing, bathing, toileting and eating. The parameters of labaratory tests were within normal limits. Magnetic resonance imaging (MRI) was performed to evaluate the lumbosacral plexus and exclude the differential diagnosises. No significant nerve pressure was detected and the MRI findings were also reported as both side of the lumbosacral plexus had the same intensity and course. Electromyographic nerve conduction tests revealed that there was no M response at the left peroneal nerve and tibial nerves bilaterally. The low amplitude of compound muscle action potential was also detected at the right peroneal nerve. Needle electromyographic study showed mild neurogenic changes but no acute denervation potential. All of these electromyographic findings were compatible with sensorymotor axonal polineuropathy on lower extremity and diabetic amyotrophic neuralgia. A medical treatment consisting of pregabalin 150 mg daily was administered. Pain severity decreased to 10 mm and the improvement in muscle strength was also detected after the treatment. The muscle strength of lower extremities increased from 1+/5 to 4/5 proximally and increased from 2+/5 to 4+/5 distally. The patient achieved to transfer independently. The limitation in her self care activities was also ameliorated. She could ambulated under observation. The patient signed informed consent form for publication of this work.

DISCUSSION

Diabetic amyotrophy typically develops in male patients with type 2 diabetes mellitus in the middle or advanced ages.[1,2,10] On the other hand it can rarely occur in in females and type 1 diabetes mellitus.[2] This clinical entity affects especially the components of lumbosacral plexus.[10] The common symptoms of the disease consist of proksimal lower extremity and hip pain involving iliopsoas, quadriceps, hip abductors and adductor muscles followed by proximal lower extremity muscle weakness, autonomic failure, weight loss.[1,9] The clinical findings can occur symmetrically or asymmetrically and sensory loss may also develop in some of the patients. By the progression of the disease, nearly half of the patients need wheelchairs for ambulation and the progression may continue up to 18 months.[2,10,11] Even though the recovery process which has become slowly and constantly incomplete, generally develops spontaneously; the relapses usually occur ipsilaterally or contralaterally.[10]

In a study with 33 patients of diabetic amyotrophy by Dick et al., the ages of the patients ranged from 35 to 80 and the majority of the patients had type II diabetes mellitus. The most severe symptoms were pain and weakness with the percentages of 82% and 18% respectively at the beginning of the disease. The distribution of symptoms was compatible with proximal (hip, thigh, buttock, or back) region in 64% of the patients and the ratio of distal part involvement (foot or leg) was 36%. Additionally the onset of the disease occured unilaterally in 88% of the patients. Most patients had proximal and distal sensory loss and nearly 50% of the patients developed new autonomic symptoms such as orthostatic hypotension, urinary dysfunction, constipation, diarrhea, tachycardia, sexual dysfunction. Weight loss of >10 pounds was also detected in the ratio of %85.[12,13] In our case; the most severe symptoms were pain and weakness consistently with literature. On the other hand the distribution of the symptoms occured symmetrically and no autonomic symptoms or weight loss occured in our patient.

The pathogenesis of diabetic amyotrophy hasn't been explained clearly but ischemic and metabolic conditions are thought to be related with the pathology.[8] It was also suggested that the occlusion of vasa nervosum and infarction may occur during the pathogenesis due to the neurological deficit and anatomical distribution of the disease.[2]

In a study by Kelkar et al., pathogenesis of diabetic amyotrophy was investigated in 15 patients with diabetic amyotrophy and 2 control patients with diabetes mellitus.[11] According to the results of the study; it was detected the typical findings of polymorphonuclear small-vessel vasculitis affecting epineurial vessels with transmural infiltration of postcapillary venules with polymorphonuclear leukocytes in four patients. Immune globulin M (IgM) deposits were also found along the endothelium and intramurally in affected vessels. Activated complement deposition was determined along the endothelium of small vessels.[11]

Laboratory tests including blood count, sedimentation rate, fasting blood glucose, hemoglobin A1c, coagulation profile can be useful for the evaluation of the patients with diabetic amyotrophy.[1] The level of cerebrospinal fluid protein is usually increased as to be between 60-100 mg/dl, but it can generally rise to 400 mg/dl; erythrocyte sedimentation rate may also be elevated (<50 mm/hr).[14] The regulation of diabetes mellitus and the disease duration, mostly don't effect the development of diabetic amyotrophy; in addition to that diabetic amyotrophy can be the first indiation of diabetes mellitus.[8,14]

Neuroimaging such as MRI is not able to show the lesions of diabetic amyotrophy directly but it can be useful to exclude the other neurologial conditions such as structural injuries, disc herniations or nerve pressures.[1]

Electrodiagnostic studies can be used for the confirmation of the diagnosis. Nerve conduction studies may detect reduced amplitudes of the compound muscle action potentials and sensory nerve action potentials; on the other hand a slight deceleration can be exposed in conduction velocities.[13,15] High amplitude motor unit action potentials, fibrillation potentials, decreased motor unit recruitment may be identified in needle electromyography study.[16,17]

There is still no proven treatment for diabetic amyotrophy. It was suggested that immune supression therapies including cyclophosphamide, oral prednisone, intravenous immune globulin, intravenous methylprednisolone and plasma exchange can be helpful for treatment.[18-21] In a study by Chan et al. in 2017; it was revealed that there is a lack of evidence from randomised trials to support a positive or negative effect of any immunotherapy in the treatment in diabetic amyotrophy.[22] On the other hand; medical treatments such as pregabaline, gabapentine, trycyclic antidepresants, venlafaxine , duloxetine can be used for the treatment of neuropathic pain symptomatically.[1] Physical-occupational therapy programmes and assistive devices for walking should be also organised for the patients according to the recuirements.[1]

CONCLUSION

Diabetic amyotrophic neuralgia is a rare complication of diabetes mellitus. It can be difficult to distinguish this clinical entity from other pathologies. Clinicians should be aware of this pathology in this patient group. The diagnosis should be confirmed by clinical, radiological and electrodiagnostic examinations.

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