A Rare Complication of Diabetes Mellitus: Bruns-Garland Syndrome (Diabetic Amyotrophy)

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ABSTRACT

Background: Diabetic amyotrophy, also known as diabetic proximal neuropathy, is a rare neuropathic complication of diabetes mellitus. It is most commonly characterized by severe pain in the hip and thigh, followed by asymmetric weakness and wasting of the proximal muscles of the lower limbs.

Case Report: This paper presents the case of a 68-year-old man with an 8-month history of poorly controlled Type-2 diabetes mellitus who developed progressive asymmetrical weakness and atrophy in his left thigh. After a physical examination, magnetic resonance imaging, and electrophysiological studies, diabetic amyotrophy was diagnosed.

Conclusion: Physicians should consider the possibility of diabetic amyotrophy in patients with diabetes mellitus who exhibit primary involvement of proximal muscles of the lower limbs, marked by pain, weakness, and atrophy.

Keywords: Diabetes mellitus, diabetic amyotrophy, neuropathy, electrodiagnosis.

INTRODUCTION

Diabetic amyotrophy (DA) is typically observed in male patients over the age of 50 with Type-2 diabetes mellitus (DM); however, it can also manifest in women and patients with Type-1 DM.1 The occurrence rate of DA is 0.3% in Type-1 DM and 1.1% in Type-2 DM.2 This condition is characterized by an acute or subacute onset of asymmetric weakness, severe pain, and muscle atrophy in the proximal region of the lower extremities.3 This article presents a cases of DA development in a patient with poor glycemic control, who was diagnosed with Type-2 DM eight months prior. The case is discussed within the context of existing literature.

CASE REPORT

A 68-year-old male patient, diagnosed with Type-2 DM at another medical center eight months ago, presented at the neurology outpatient clinic two days ago with complaints of severe pain, weakness, and muscle wasting radiating from the left groin area to the anterior thigh. Over the past two months, he experienced difficulty climbing stairs and rising from a seated position. The patient, weighing 90 kg with a height of 190 cm, had lost approximately 11 kg over the previous six months. He had been using a cane for the last month. Neurological examination
revealed extension strength of 4/5, flexion strength of 4/5, and abduction and adduction muscle strength of 4/5 in the proximal left lower extremity, while all other extremities exhibited normal muscle strength. Upper extremities displayed normoactive biceps, triceps, and styloradial reflexes; however, lower extremities showed hypoactive patella and Achilles reflexes bilaterally. Bilateral sole skin reflex was flexor. While sensory and joint position examination yielded normal results, bilateral vibration sense was diminished. The patient's waist range of motion was bilaterally normal and painless. Straight leg raising test (SLRT) and flexion-abduction-external rotation-extension (FABERE) tests were both negative bilaterally. The femoral stretching test also produced negative results. The patient reported intense pain, rating its severity as 10 out of 10. Superficial sensory examination revealed sensations of numbness and tingling in the hands and feet, not corresponding to dermatomes, presenting in a glove-sock distribution. Thigh thickness was measured 10 cm above the patella using a tape measure. Right thigh thickness measured 42 cm, while left thigh thickness measured 38 cm (Fig. 1). Lumbar magnetic resonance imaging (MRI) indicated no disc protrusion (Fig. 2).

In the nerve conduction study, findings supporting sensorimotor polyneuropathy and a reduction in the amplitude of the left femoral motor nerve were observed (Fig. 3).

The patient’s fasting blood glucose was 355 mg/dL, HbA1c was 11.4, blood pressure was 110/70, and pulse rate was 74/min. C-reactive protein, creatine kinase, and sedimentation rate were within normal ranges. Tests for Brucella, Human Immunodeficiency Virus (HIV), Epstein-Barr Virus (EBV), Cytomegalovirus (CMV), Venereal Disease Research Laboratory (VDRL), Hepatitis B Virus (HBV), and Hepatitis C Virus (HCV) yielded normal results. The vasculitis-thrombophilia panel was also normal. The patient had no recent history of infection, vaccination, trauma, or exposure to toxins or chemicals. Based on the present findings, we diagnosed the patient with DA. Pulse methylprednisolone (PMTP) treatment was initiated at a dose of 1 g/day; however, it was discontinued when the patient declined due to a worsening of his overall condition resulting from a sudden increase in blood glucose levels. The patient underwent intravenous immunoglobulin (IVIg) treatment at a dosage of 35 g/day for five days. Treatment was supplemented with alpha-lipoic acid 600 mg 1x1, pregabalin 75 mg 2x1, and duloxetine 60 mg 1x1. The patient was referred to the endocrinology department and initiated on insulin aspart 3x10U and insulin glargine 12 units once daily. Additionally, the patient received consultation from the physical therapy and rehabilitation (PTR) department, where isometric and isotonic exercises were prescribed to enhance muscle strength. The dosage of pregabalin was gradually increased to alleviate the patient’s pain complaints. Upon discharge, the patient was provided with dietary and exercise recommendations along with prescriptions for pregabalin 300 mg 2x1, duloxetine 60 mg 1x1, alpha-lipoic acid 600 mg 1x1, and insulin. A one-month follow-up appointment at the neurology outpatient clinic was advised.

Upon returning for a follow-up four months later, the patient underwent 20 days of physical therapy at an external facility. Although the patient no longer reported pain, the muscle atrophy in the left anterior thigh had not improved. Thigh thickness measured 38 cm, 10 cm above the left knee, and
sleep thickness exhibited no change. The patient’s random blood glucose level was 180 mg/dL, with an HbA1c of 8.4.

The second nerve conduction study showed no alterations, and the needle electromyography test was consistent with the prior one. Neuropathic pain treatment with pregabalin and duloxetine was ceased after 18 months of treatment, and the patient’s progress was monitored at irregular intervals. During the subsequent six months, the patient exclusively used insulin and reported no pain complaints; however, muscle atrophy in the left thigh persisted as a sequela.

**DISCUSSION**

The features of DA were initially described as pelvicofemoral weakness and diabetes-related weakness by the German neurologist Ludwig Bruns in 1890. Subsequently, Garland and Taverner expanded upon this definition in 1953, coining the term DA. While upper extremity involvement is rare, these patients can experience atrophy and strength loss in major muscle groups over a few weeks. Differential diagnoses should exclude lumbar disc herniation (LDH), spinal canal stenosis, Guillain-Barré syndrome, chronic demyelinating neuropathy, cauda equina syndrome, and neoplastic lumbosacral plexopathy. In this patient, lumbar spinal stenosis (LSS) and LDH were ruled out based on lumbar MRI findings. Furthermore, LDH and LSS correlate with affected spinal pathways, resulting in pain and paraesthesia. Electrophysiological studies play a pivotal role in DA diagnosis. Nerve conduction studies in DA patients consistently reveal significantly diminished femoral nerve amplitude on the affected side compared to the intact side. Needle electromyography (EMG) detects spontaneous denervation indicators such as positive sharp waves and fibrillation potentials in quadriceps and adductor muscles.

With the notable involvement of immune mechanisms in its pathogenesis, positive outcomes have been achieved through immunotherapy in several patients. In one particular study, five individuals diagnosed with DA underwent IVIg treatment at a dosage of 0.4 g/kg/day for five days. Within ten days, four of these patients demonstrated a significant reduction in visual analogue scale scores, coupled with notable decreases in pain reports and a considerable increase in walking distances within a month. In a documented case report, an individual diagnosed with DA at the age of 57, initially wheelchair-bound, experienced an 80% decrease in visual analogue scale score after five days of IVIg treatment at a dosage of 0.4 g/kg/day, subsequently transitioning to using a cane for walking. Guided by this data, we administered our patient a five-day course of IVIg treatment at a dosage of 35 g/day. We hold the belief that this IVIg regimen played a significant role in rapidly alleviating our patient’s pain complaints.

**CONCLUSION**

In clinical practice, LDH, it is imperative to consider lumbar disc herniation (LDH), a commonly confused condition, in the differential diagnosis of DA, particularly among middle-aged male patients with a diabetes history. Patients presenting pain, muscle atrophy, and weight loss should undergo comprehensive assessment, and electrophysiological studies should be conducted for those suspected of having DA. Employing this approach can effectively avert unnecessary lumbar surgical procedures.
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REFERENCES


