

# Diabetic Lumbosacral Plexopathy- Bruns Garland syndrome: A Case Report

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## Abstract

Brun's Garland syndrome commonly known as diabetic amyotrophy is a rare complication of diabetes mellitus. It is one of the many forms of diabetic neuropathy. It has been extensively described in the past as a distinct entity with many isolated case reports and case series. Various treatment options have been tried over the years yielding confounding results. Here we describe a case of Diabetic amyotrophy treated with Pulse Methylprednisolone and IvIg resulting in clinical improvement and recovery.

**Keywords:** Diabetes Mellitus, Neuropathy, Plexopathy, Amyotrophy, Bruns-Garland syndrome

## Introduction

Diabetic lumbosacral radiculoplexus neuropathy (DLSRPN) is a rare manifestation of Diabetes Mellitus. It is one of many neurological complications of Diabetes. (Ng et al., 2019) Ludwig Bruns had described three such patients in the year 1890 who presented with severe pain in the hip and thigh followed by weakness and wasting of leg muscles. These cases were overlooked until this condition was identified by Hugh Garland and Deryck Taverner in the year 1950. (Greenbaum, 1962) He went on to notice that these patients showed remarkable recovery in their weakness, areflexia and extensor planters, but had a significant residual proximal muscle wasting. Many terminologies have been used over the years for this entity, like diabetic myelopathy, diabetic amyotrophy, Bruns-Garland syndrome, diabetic mononeuritis multiplex, diabetic lumbosacral plexopathy, diabetic polyradiculopathy and multifocal diabetic neuropathy. The currently accepted nomenclature are Diabetic amyotrophy, DLSRPN (diabetic lumbosacral radicular plexopathy) and Bruns-Garland syndrome. Many case series have been described for this condition over the years with varying treatment options. This case describes an elderly individual with freshly detected type 2 diabetes mellitus who presented with asymmetrical weakness of both lower limbs along with sensory symptoms. He was managed initially with IV pulse methyl pred and later with IvIg. He showed improvement in his symptoms in the form of regaining power and alleviation of pain.

## Case report

This 61-year-old, male, was detected to have Type 2 Diabetes Mellitus 8 months back when he presented with unintentional and unquantified loss of weight and fatigue along with osmotic symptoms. He was initiated on oral antidiabetic agents (OADs) (Tab Metformin 500mg BD and Tab Glimiperide 1mg OD). A month after starting on OADs he started having pain in left thigh which sharp and radicular in nature, radiating till his left leg. It was acute in onset, gradually progressive and continuous. 15 days later it was followed by similar involvement in the right thigh. He also had progressive weakness of both lower limbs since last 4 months which was insidious in onset, progressive in nature. The weakness was more proximal

in the left and distal in the left lower limb. There was no weakness of upper limb/trunk/neck/ facial weakness. There was no bladder bowel involvement or band of tightness on the trunk. He also complained of decreased sensation to clothes in both lower limb and no decrease in hot or cold sensations. His symptoms progressed to the point that he was bed bound and dependent for his activities of daily living when he reported to this hospital.

The patient was evaluated for painful, asymmetrical lower motor neuron type weakness of both lower limbs in the background of recent detection of Type 2 Diabetes Mellitus and initiation of oral glucose lowering drugs. On clinical examination, the patient had motor weakness involving both the proximal and distal muscles of bilateral lower limbs with diminished sensation to touch and vibration. He had absent knee and ankle reflexes on both sides with normal biceps, triceps and supinator reflex. His planter reflexes were equivocal. He had normal levels of Vitamin B12, folate levels, negative tumour markers, no M-spike on serum electrophoresis and no abdominal organomegaly. His ACE levels were normal. MRI spine did not show any significant abnormality. CSF studies were essentially normal with no albumino cytological dissociation. His nerve conduction showed bilateral lower limb sensorimotor neuropathy with radiculopathy L5/S1 on left side. Viral markers (Hepatitis B, Hepatitis C and HIV) were negative. During his admission patient was noticed to have persistent tachycardia, evaluation revealed normal thyroid function tests and serum cortisol levels. Ultrasound abdomen and 2D echocardiography were essentially normal. Bed side autonomic function tests revealed orthostatic hypotension and inappropriate response to Valsalva. There was no relevant past history and his family history was not significant.

He was exhibited pulse therapy of methyl prednisolone 1000 mg x 3 days along with supporting active and passive physiotherapy. Patient had well controlled blood sugar levels on admission (Fasting-88mg/dl, Post-prandial-136mg/dl), however he was shifted to Insulin (Inj Glargine 15 U SC HS) during his hospital stay in view of elevated blood sugar levels post methyl prednisolone therapy. He had marginal improvement of his lower motor functions and was exhibited IVIg 150 g over five days. Over the next 15 days patient had gradual but significant improvement in his muscle power. He was able to walk with the help of support and had relief from the pain in his lower limbs. He was discharged with significant functional improvement and was relieved of his pain.

## Discussion

Brunns Garland syndrome has been described in various case series over the years. It has been reported in both new onset as well as long standing cases of Diabetes Mellitus. It typically starts with asymmetrical pain in the lower extremities (proximal more than distal). (Albers et al., 2020) This classically progresses over the course of time to weakness and atrophy of the involved limbs. The median age of onset is 65 years and the median time to diagnosis after onset of diabetes is around 4 years according to various case series. This patient is a newly diagnosed case of diabetes who after institution of therapy developed these symptoms. Nerve conduction studies show decreased amplitudes of lower limb motor and sensory action potentials. The aetiology of this condition has been long debated upon and is controversial. Both ischaemic and metabolic hypotheses have been proposed in the past. According to latest evidence a vasculitic aetiology of ischaemia followed by axonal degeneration and demyelination is the widely accepted theory. The main clinical features of diabetic amyotrophy are weakness, wasting and pain. (Flatow & Michelsen, 1985) This is mostly found in the muscles of lower limbs namely the quadriceps muscle. Though the weakness starts on one side, it almost always spreads to the other side in an asymmetrical manner. Sensory symptoms like pain, dysaesthesia and paraesthesia are also commonly found in these

patients. On examination, there is weakness in the involved muscles along with sensory symptoms involving the decreased proprioception, touch, pain and temperature. Lower limbs tendon jerks, especially the patellar, are classically found to be absent. In some patients planter reflexes may show extensor responses. In patients who undergo muscle biopsy classical findings of denervation and microvascular hyalinization are also found. Pipestem micrangiopathy is a distinctive pathologic reaction pattern consisting of necrotizing myopathy, minimal cellular infiltration, and a microangiopathy with thick "pipestem" vessels and microvascular deposits of complement membrane attack complex.(Tracy et al., 2009) It is classically found in autoimmune diseases and carcinomas. Electromyography shows evidence of denervation in affected muscles.

While contemplating on treatment options only one randomised control trial is found in the database that too was unpublished and not peer reviewed. In this study participants received 1g IV methylprednisone thrice weekly and continued for 12 weeks in tapering doses. The (Neuropathy Impairment Scale (NIS) or NIS lower limbs (NIS (LL))) was used to evaluate the patients. There NIS was improved by 4 points in the study group. Other case series or case reports also employ different treatment options, namely IV methylprednisolone, IvIg and even IV cyclophosphamide with varied success.

IV methyprenisne and IvIg remain the modalities of choice till date for this entity. The course of this disease is variable among different individuals. Many patients show improvement in their symptoms, both motor and sensory. The disease is known to recur with episodes occurring on opposite sides of limbs classically. The cornerstone of long term therapy remains good glycemic control, physiotherapy and occasional pain relief.(Tracy & Dyck, 2014)

**Review of Literature**

	<b>Intervention</b>	<b>Outcome</b>
Said 1994	3 people received IV methylprednisolone	All 3 people had rapid relief of pain within days after treatment. 1 had no weakness. The strength in the other 2 recovered over several months.
Krendel 1995	8 people received IVIg. 3 people received prednisolone. 2 people received a combination of IVIg and prednisolone. 1 people received combination of IVIg, prednisolone and IV cyclophosphamide. 1 people received combination of IVIg and IV cyclophosphamide.	In all of those treated, worsening stopped when treatment began and there was a subsequent improvement in strength. Re-recovery was complete or nearly complete in 5.
Pascoe 1997	3 people received prednisolone. 5 people received Plasma exchange 3 people received IVIg.	All improved, except 1 who received prednisolone, 1 who received plasma

	1 people received Plasma exchange and IVIg.	exchange and the 1 who had both plasma exchange and IVIg
Jaradeh 1999	9 people received PE. 6 people received IVIg.	All had no further deterioration and had dramatic subjective improvement, in particular rapid resolution of their pain.
Kilfoyle 2003	10 episodes of Diabetic amyotrophy in 9 people were treated with oral or intravenous MP.	In 6 episodes, there was a marked improvement in pain within days of starting treatment. Pain resolved in 7 episodes within 3 months of symptoms' onset.
Zochodne 2003	2 people received IVIg.	Progression of pain and weakness despite treatment.
Tamburin 2009	5 people received IVIg. 2 of those treated had repeat IVIg treatment due to pain recurrence after 7 to 18 months.	4 had reduction of severe pain starting 5 to 10 days after IVIg infusion. The 2 with pain recurrence also improved with further IVIg treatment.
<b>Dyck 2006</b>	Randomised, double-blind, placebo-controlled study. 75 participants (44 men and 31 women) with a mean age of 65.3 years (range 36 to 81) received either Intravenous methylprednisolone or placebo.	Time to improve NIS (LL) by four points (primary endpoint). Study findings were presented in abstract form but unpublished.

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