Bilateral brachial plexopathy as an initial presentation in a newly-diagnosed, uncontrolled case of diabetes mellitus

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ABSTRACT

A 55-year-old Indian woman with newly-diagnosed diabetes mellitus presented with acute onset right upper limb proximal weakness. This was followed three weeks later by pain, weakness and sensory loss in the left upper limb. Electrodiagnosis showed patchy multiple proximal and distal axonal neuropathies in both upper limbs, consistent with bilateral brachial neuritis. Laboratory investigations, cerebrospinal fluid analysis, and imaging studies were normal except for an anti-nuclear antibody titre of 1:640. Sural nerve and quadriceps biopsy did not show vasculitis. Brachial plexopathy has seldom been associated with diabetes mellitus and could represent a rare subtype of the diabetic neuropathies.

Keywords: bilateral brachial plexopathy, brachial plexus neuropathies, diabetes mellitus, diabetic neuropathies

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INTRODUCTION

Neuropathy is a frequent complication of diabetes mellitus and occurs in approximately 50% of diabetic patients over time.[1] Diabetic neuropathy is not a single disorder, but a series of distinct clinical syndromes. A wide variety of manifestations may develop, and numerous classifications have been attempted. Thomas and Tomlinson advocated a broad subdivision into symmetric polyneuropathies, focal and multifocal neuropathies.[2] This includes sensorimotor polyneuropathy, autonomic neuropathy, cranial neuropathy, trunk and limb mononeuropathies, mononeuropathy multiplex and diabetic lumbosacral radiculoplexus neuropathies. Isolated brachial plexus involvement has been rarely reported as a complication of diabetes mellitus. We present a case of bilateral brachial plexopathy in the setting of newly-diagnosed diabetes mellitus.

CASE REPORT

A 55-year-old Indian woman was admitted with an initial presentation of polydipsia, polyuria and weight loss for a period of three weeks; blood glucose was 381 mg/dL with signs of dehydration. HbA1c at the time was 14.2%. She was hydrated and blood sugars were optimised. She was discharged after one week to continue medication (Metformin) at home.

Fig. 1 Photograph shows the patient's inability to flex the interphalangeal joint of the thumb and the distal interphalangeal joint of the index finger (pinch or OK sign) implying a median nerve lesion proximal to the wrist bilaterally.

Upon reaching home, while taking a shower, she found that she could not lift her right shoulder to wash her hair. Elbow, wrist and hand movements were unaffected. She did not feel any pain or numbness and did not seek medical advice. About three weeks later, she woke up to a severe, sharp pain of her entire left arm that lasted for about four hours. She self-medicated with a non-steroidal anti-inflammatory drug and the pain resolved by the next morning. But she noted numbness over the lateral three fingers of the left hand (palmar aspect). She also noted the left upper extremity to be generally weaker, although she was unclear about the distribution. She has a background history of poliomyelitis at one year of age. This primarily affected her right lower limb and she ambulates independently with a limp. There is family history of diabetes mellitus in her mother and one sibling.

Upon readmission, the manual muscle examination revealed severe weakness of the right deltoid (Medical Research Council [MRC] grade 2/5) and right biceps brachii (MRC 3/5). The left upper extremity showed moderate weakness of all median and ulnar innervated muscles (pronators, thumb and index flexors, and thumb abductors) (Fig. 1). Both lower extremities also showed

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Table I. Results of nerve conduction studies.

Nerve	Recording site	Variable	Right	Left	Normal
Motor studies					
Median	Abductor pollicis brevis	Amplitude-D/P (mV)	4.9/4.2	3.5/2.1	> 6.0
		Distal latency (ms)	7.1	4.2	< 4.0
		Conduction velocity (m/s)	50.0	48.0	> 50.0
		F-wave minimal latency (ms)	29.9	29.3	< 28.8
Ulnar	Abductor digiti minimi	Amplitude-D/P (mV)	8.4/7.5	5.9/5.7	> 7.0
		Distal latency (ms)	2.8	2.9	< 3.1
		Conduction velocity (m/s)	74.0	63.0	> 50.0
		F-wave minimal latency (ms)	27.3	ND	< 29.8
Radial	Extensor indicis propius	Amplitude-D/P (mV)	6.3/5.8	2.2/2.4	> 5.0
		Distal latency (ms)	2.0	2.1	< 3.1
		Conduction velocity (m/s)	53.0	51.0	> 50.0
Axillary	Deltoid	Amplitude (mV)	2.6	4.9	> 4.0
		Distal latency (ms)	3.2	3.2	< 4.8
Musculocutaneous	Biceps brachii	Amplitude (mV)	1.9	4.1	> 4.0
		Distal latency (ms)	5.4	5.0	< 3.5
Peroneal	Extensor digitorum brevis	Amplitude-D/P (mV)	ND	NR	> 2.5
		Distal latency (ms)			< 5.0
		Conduction velocity (m/s)			> 40.0
Peroneal	Tibialis anterior	Amplitude-D/P (mV)	ND	2.3	> 3.0
		Distal latency (ms)		4.5	< 4.5
		Conduction velocity (m/s)		59.0	> 40.0
Tibial	Abductor hallucis longus	Amplitude-D/P (mV)	ND	1.3	> 2.5
		Distal latency (ms)		5.7	< 5.0
		Conduction velocity (m/s)		42.0	> 40.0
		F-wave minimal latency		50.9	< 52.7
Sensory studies					
Median	Digit 2	Amplitude (μV)	14.0	NR	> 15.0
		Distal latency (ms)	3.2		< 3.6
Ulnar	Digit 5	Amplitude (μV)	29.3	19.2	> 15.0
		Distal latency (ms)	2.5	3.0	< 3.1
Radial	Anatomical snuffbox	Amplitude (μV)	21.0	NR	> 14.0
		Distal latency (ms)	4.2		< 2.7
Lateral antebrachial	Forearm	Amplitude (μV)	NR	9.6	
		Distal latency (ms)		2.0	
Medial antebrachial	Forearm	Amplitude (μV)	NR	NR	
		Distal latency (ms)			
Sural	Ankle	Amplitude (μV)	ND	21.7	> 4.0
		Distal latency (ms)		3.1	< 4.6

mV: millivolts; ms: milliseconds; m/s: metres per second; μV: microvolts; NR: no response; ND: not done

weakness, more severe in the distal muscles and worse on the right lower limb. This was attributed to the previous poliomyelitis. The deep tendon reflexes were absent except for the left triceps jerk. The sensory examination shows mild hypaesthesia over the lateral aspect of the right arm and was reduced severely over the palmar aspect of the lateral three digits of the left hand. Vibration and proprioception sense were intact.

The patient's blood glucose was apparently well controlled. Both the fasting blood sugar and HbA1c showed normal results. The following tests were normal, including the renal, liver and thyroid function tests; protein electrophoresis; antineutrophil cytoplasmic antibodies and anticardiolipin antibodies; rheumatoid arthritis factor; anti-dsDNA; magnetic resonance imaging of the cervical spine and brachial plexus. Cerebrospinal fluid (CSF) analysis showed normal results. The erythrocyte sedimentation rate was slight elevated at 22 mm/hr and the anti-nuclear antibody was positive at 1:640. The left sural nerve and quadriceps muscle biopsies were normal with no evidence of vasculitis. A course of intravenous immunoglobulin was administered at one g/kg for two days with minimal improvement.

The electrodiagnostic examination was performed about eight weeks after the onset of the initial symptoms. In the right upper extremity, there was evidence of severe axon-loss axillary and musculocutaneous neuropathy (Table I). The biceps brachii showed spontaneous discharges on electromyography (EMG) while the following muscles showed high amplitude motor unit

Table II. Results of electromyography.

Muscle	Side	Spontaneous activity		Motor unit potentials		Recruitment
		Fibs	PSW	Amp	Duration	
Deltoid	Right	Nil	Nil	Inc	Long	Discrete
Biceps brachii	Right	2+	2+	Inc	Long	Discrete
Triceps	Right	Nil	Nil	Inc	Long	Discrete
Extensor digitorum communis	Right	Nil	Nil	NL	NL	Complete
Pronator teres	Right	Nil	Nil	Inc	Long	Discrete
First dorsal interossei	Right	Nil	Nil	NL	NL	Moderate
Deltoid	Left	Nil	Nil	NL	NL	Moderate
Biceps brachii	Left	Nil	Nil	NL	NL	Complete
Triceps	Left	Nil	Nil	NL	NL	Moderate
Extensor digitorum communis	Left	Nil	Nil	NL	NL	Moderate
Pronator teres	Left	3+	3+	No MUPs		–
Extensor indicis	Left	1+	Nil	Inc	Long	Discrete
Flexor pollicis longus	Left	1+	1+	No MUPs		–
Flexor carpi ulnaris	Left	2+	2+	Inc	Long	Moderate
First dorsal interossei	Left	Nil	Nil	NL	NL	Moderate
Cervical paraspinals	Left	Nil	Nil			
Tibialis anterior	Left	Nil	Nil	Inc	Long	Discrete
Gastrocnemius, medial	Left	Nil	Nil	Inc	Long	Discrete

Fibs: fibrillation potentials; PSW: positive sharp waves; Amp: amplitude; Inc: increased amplitude; MUPs: motor unit potentials; NL: normal

potentials (MUPs) and a reduced recruitment pattern: deltoid, triceps, pronator teres. In the left upper extremity, there was evidence of lower trunk involvement with axonal loss seen in median, ulnar and radial distribution in the nerve conduction studies (NCS). The following muscles showed spontaneous discharges on EMG: pronator teres, extensor indicis, flexor pollicis longus and flexor carpi ulnaris. The MUPs were of increased duration and the recruitment pattern was reduced. The bilateral first dorsal interossei, bilateral extensor digitorum communis, left biceps brachii, left triceps, left deltoid, and left cervical paraspinals showed normal findings (Table II). In the lower extremities, the peroneal NCS showed low to absent compound muscle action potentials (CMAPs) when recording from extensor digitorum brevis and abductor hallucis muscles for the peroneal and tibial nerves, respectively. The sural sensory response was normal. The needle examination showed chronic high amplitude MUPs with a discrete recruitment pattern. These findings were consistent with the previous poliomyelitis with involvement of both lower limbs, with a greater severity on the right side. The normal sural studies ruled out a concomitant polyneuropathy in this patient.

In summary, multiple nerves of both upper extremities were involved in an asymmetric manner. In the right upper extremity, there was evidence of axon-loss axillary, musculocutaneous and median neuropathies. On the left upper extremity, there was evidence of axon-loss median, radial and ulnar neuropathies with overall pattern being consistent with brachial neuritis. The lower extremities showed evidence of previous poliomyelitis. The normal sural sensory potentials make a concomitant polyneuropathy less likely in this patient.

DISCUSSION

Isolated brachial plexopathy has rarely been reported in the setting of diabetes mellitus. One report described the occurrence of brachial plexopathy in association with diabetic ketoacidosis while the hyperglycaemia and acidosis were being corrected.[3] Another report described a case of bibrachial plexopathy in a patient with well-controlled type 2 diabetes.[4] It is interesting to note that in our case, the patient's brachial plexopathy did not occur during the period when she was grossly hyperglycaemic. Her problems only occurred at the time she was sent home with the blood sugar presumably stabilised. This leads one to speculate whether the rapid correction of the metabolic imbalance is somehow connected with the plexopathy.

The differential diagnosis would include chronic inflammatory demyelinating polyradiculopathy (CIDP), diabetic amyotrophy, and mononeuropathy multiplex. CIDP usually presents with insidious onset of weakness, which can affect both proximal and distal muscles. Patients with diabetes mellitus have a predisposition to developing CIDP. However, the electrodiagnostic studies did not show evidence of demyelination or conduction block. The patient's lumbar puncture also showed normal results with no elevation of CSF protein.

Diabetic radiculoplexus neuropathy or diabetic amyotrophy is a well-recognised subacute, painful, asymmetric lower-limb neuropathy that is associated with weight loss and diabetes mellitus. A review found that 10% of patients with lumbosacral radiculoplexus neuropathy had a more diffuse disorder consistent with cervical radiculoplexus neuropathy.[5] Nevertheless, all reported cases of upper limb involvement have concomitant lower-limb involvement, which is not present in this patient.

Diabetic mononeuropathy multiplex presents as peripheral nerve deficits that appear abruptly, usually sequentially, and at irregular intervals, affecting two or more peripheral nerves, and often on different limbs.[6] Since this patient does present with multiple neuropathies, it is not impossible that a mononeuropathy multiplex is the cause of her problems. However, it would be more parsimonious to attribute it to a single lesion affecting the brachial plexus. In addition, it would be unlikely that a mononeuropathy multiplex would affect multiple nerves of the upper extremity only and spare the nerves of the lower extremities.

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