

Man-In-The-Barrel Syndrome: Acute bilateral brachial plexopathy after recurrent microtrauma

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Abstract

The presence of brachial diplegia despite the normal muscular strength of the lower extremities is called the man-in-the-barrel syndrome (MIBS). Although this rare syndrome often occurs due to the bilateral supratentorial brain lesions, it may also rarely occur as a result of infratentorial causes. In this report, we describe a case presenting with MIBS of which etiological underlying cause was bilateral brachial plexopathy developed secondarily to recurrent microtrauma. A 51-year-old male patient presented to our clinic with complaints of pain and weakness on both arms. After electrodiagnostic examination, bilateral brachial plexopathy was identified. The findings of the patient improved following methylprednisolone therapy. It is very important to determine the treatable causes of this syndrome at an early stage.

Keywords: Brachial diplegia, "Man-In-The Barrel" syndrome, Recurrent microtrauma.

Introduction

MIBS is characterized by brachial diplegia. Lower extremities have full muscle strength, or this muscle strength is relatively preserved. The appearance of the patient is like a man being confined in a barrel.¹ This rare syndrome is associated with cerebral hypoperfusion due to border zone infarcts that are generally present between the anterior and middle cerebral artery irrigation areas.² In addition to many cerebral lesions with different natural characteristics that cause the upper limb-related fibres of the pyramidal corticospinal tract to be injured, cervical spinal cord lesions and peripheral nerve diseases may also be present with this disease.³ Herein we presented a patient with MIBS which was due to bilateral brachial plexopathy caused by recurrent microtrauma.

Case Report

A 51-year-old male patient was admitted to our clinic in January 2016 with complaints of pain and numbness

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Figure-1a: Inability to abduct upper limbs.

starting from the back of the neck and spreading to the shoulders, with weakness on both arms, which was more dominant on the right arm. Symptoms progressed gradually within two days. In his neurological examination, the patient was conscious. No meningeal irritation was detected, cranial nerves were intact. In both upper extremities, muscle strength was 1-3/5 in proximal group and 3-4/5 in distal, whereas lower limbs had full muscular strength. Abduction of the upper

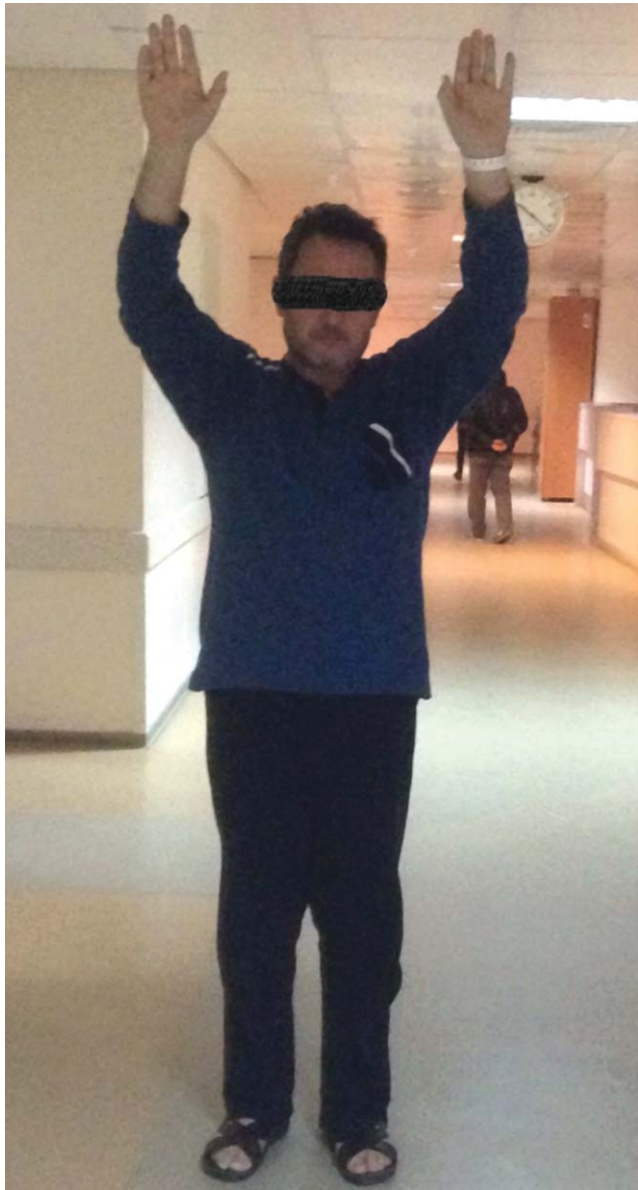


Figure-1b: Normal muscle strength after treatment.

limbs was not possible (Figure-1a). Deep tendon reflexes were hypoactive in the upper extremities, whereas they were normoactive in the lower extremities. The plantar response was bilateral flexor. The sensory examination revealed dysesthesia and allodynia on both hands. The patient had no history of chronic illness and medication. After retiring from his desk job at the municipality, he started to work in a glass factory since three months. He had been placing glass plates of different sizes from one machine to another, and both shoulders were stretched in abduction and external rotation during this process. Complaints began at rest after eight hours of work.

There was no major trauma history. Cranial and cervical magnetic resonance imaging (MRI) was normal. In the laboratory examination, biochemistry, haemogram, sedimentation, thyroid function tests, HIV serology, tumour markers and vasculitis panel were normal. The biochemical analysis and microscopic examination of the cerebrospinal fluid were also normal. In the electroneuromyographic examination (ENMG) of the patient, the speed of the motor and sensory nerve transmission, compound muscle action potential (CMAP) amplitudes and sensory nerve action potential (SNAP) amplitudes in the bilateral lower extremities were normal. SNAP amplitudes and transmission rates of both upper extremities were normal. Bilateral median nerve CMAP amplitudes and transmission rates were normal, bilateral ulnar nerve CMAP amplitudes were small. Bilateral radial nerve and axillary nerves could not be stimulated at the Erb point. The distal latency of the musculocutaneous nerve was significantly longer on the right and amplitude was found to be small bilaterally. In the needle electromyographic study of bilateral C5-6-7-8 cervical myotomes, spontaneous denervation findings were absent, neurogenic changes in motor unit potentials and a decrease in the number of motor units ignited were observed. Bilateral brachial plexopathy was suspected for the patient. Muscle strength reached 5/5 on both upper extremities by following methylprednisolone therapy 1000 mg/day for five days (Figure-1b). Control EMG revealed that CMAP amplitudes of radial and axillary nerves were started to be recorded with the stimulation from the Erb point.

Discussion

MIBS is a very rare syndrome. Bilateral anterior watershed infarction, systemic hypoperfusion or hypovolaemic shock,² pontine and extrapontine myelinolysis,⁴ closed head trauma,⁵ central nervous system metastases,⁶ bilateral intratumoural haemorrhage,⁷ cervical cord contusion,⁸ cervical region metastases,⁹ spinal cord infarcts,¹⁰ cervical myelopathy,¹¹ hereditary and sporadic ALS, Sjögren and HIV-associated motor neuron disease,¹² bilateral brachial plexopathy^{13,14} and cyclosporin-dependent neurotoxicity¹⁵ are some of the etiologic factors presented in the literature for MIBS.

Conclusion

Brachial plexopathy-associated MIBS has been previously presented as radiation plexopathy¹³ and postoperatively developed plexopathy.¹⁴ To our knowledge, MIBS has not yet been reported as an acute manifestation of brachial plexopathy due to recurrent microtrauma. In accordance with the etiological reasons mentioned above, it is clear that this syndrome is not always with a good prognosis.

Therefore, it is very important to determine the treatable causes quickly.

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Informed and written consent was taken from the patient.

Conflict of Interest: The authors confirm that this article content has no conflicts of interest.

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