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Case Report

Bilateral pan-plexus lesion after substance use: A case report

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ABSTRACT

Peripheral nervous system complications such as acute demyelinating polyradiculopathy and mononeuropathy may rarely develop after substance use. A 27-year-old man used illegal drugs the day before his admission to the emergency service. Initially, he was suspected for rhabdomyolysis, due to elevated blood urea nitrogen, creatinine, aspartate aminotransferase, alanine aminotransferase, myoglobin, and creatine kinase levels. On Day 4, generalized edema and flask paralysis were noted in both upper limbs. The patient was diagnosed with bilateral brachial pan-plexopathy based on electrophysiological study results. He underwent a rehabilitation program. After eight months, repeated electrophysiological study revealed a significant improvement in all bilateral upper limb muscles, except for the right abductor pollicis brevis and abductor digiti minimi muscles. The underlying cause of bilateral brachial pan-plexopathy was rhabdomyolysis secondary to substance use. In conclusion, substance use in patients with non-traumatic plexopathy should always be questioned.

Keywords: Brachial plexus, rhabdomyolysis, substance use.

Brachial plexus lesion is responsible for approximately 14% of all isolated upper limb neurological lesions.[1] Damage of the plexus-forming nerve fibers can be caused by various mechanisms such as laceration, compression, stretching, radiation, and ischemia. The most common cause of brachial plexus injuries is trauma (82% traffic accidents, 6% sports injuries, and 12% causes).[2] However, there are many causes other than trauma such as neoplasm, thoracic outlet syndrome, neuralgic amyotrophy and radiation therapy.[3] In non-traumatic cases, bilateral plexus involvement is rare. Identifying the underlying etiology in a non-traumatic plexus lesion is important for the diagnosis, treatment, and prognosis.

Substance abuse has been increasing in the society worldwide. According to the World Health Organization, 200 million individuals (almost 5%) between the ages of 15 and 64 years annually use one or another of the illegal drugs around the world.[4] Therefore, various complications of substance abuse can be seen in daily practice. Herein, we present a rare case of bilateral brachial pan-plexus lesion resulting from substance use.

CASE REPORT

A 27-year-old male patient with chronic alcohol use was admitted. He was found unconscious at home by his relatives in the morning. This was the first time that he used 3,4-methylenedioxy-N-methylamptamine (MDMA) (ecstasy) and marijuana about 12 hours ago. When he was admitted to the emergency service, his person and place orientation were intact, but the time orientation was impaired. He was somnolent, hyperthermic, and tachycardic. On physical examination, he had no edema or weakness in the upper and lower limbs. Musculoskeletal and other systemic examination findings were normal. In his medical history, there was no related trauma or prior findings of alcohol-induced neuropathy. Laboratory testing revealed elevated blood urea nitrogen (46.6; 6-20 mg/dL), creatinine (3.52; 0.6-1.24 g/dL), aspartate aminotransferase (AST) (1095; 4-40 U/L),

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alanine aminotransferase (ALT) (786; 5-41 U/L), myoglobin (>3000; 28-72 ng/mL), and creatine kinase (CK) (2587; 0-90 U/L) levels. Computed tomography which was performed to rule out brain injury showed no abnormality. The patient was diagnosed with an acute tubular necrosis and toxic hepatitis due to rhabdomyolysis and was followed for three days in the intensive care unit.

On Day 4, he was evaluated by a neurologist due to generalized edema and total paralysis in both upper limbs. Based on neurological examination, the muscle strength of bilateral upper limbs was 0/5, deep tendon reflexes were hypoactive, C5-T1 dermatomes were anesthetic, and Hoffman signs were negative. Neurological examination of the cranial nerves and both lower limbs was normal. It was suggestive of a lesion at the brachial plexus or multiple spinal nerve roots. No abnormality was found in the patient's cervical and cranial magnetic resonance imaging (MRI) which were performed to find the etiology of flask paralysis. After the patient was followed for eight days in the internal medicine clinic, creatinine, AST, ALT, myoglobin, and CK levels decreased and the patient was discharged. He was applied to physical medicine and rehabilitation outpatient clinic for physical therapy. At two months, electrophysiological study including bilateral median, ulnar, lateral/medial antebrachial cutaneous, and radial superficial nerves sensory nerve conduction studies did not reveal any action potentials in bilateral upper limb sensory nerve conduction studies. In addition, no action potentials were obtained in the right median, ulnar and radial nerve motor nerve conduction studies. The amplitude of action potentials was reduced, and nerve conduction velocities decreased in the left median and ulnar nerve motor nerve conduction studies. No action potential was obtained in the bilateral axillary, radial and right suprascapular nerve using the Erb stimulation. The latency of the action potential recorded from the left biceps muscle (musculocutaneous nerve) using the Erb stimulation was prolonged. In the needle electromyography (EMG), bilateral supraspinatus muscles showed a significant reduction in the recruitment pattern, while no motor unit action potential (MUAP) were seen in the other muscles. Also, fibrillation and positive sharp waves were observed in all upper limb muscles. The bilateral cervical paraspinal muscles were normal. Findings suggesting a severe axonal injury with a bilateral brachial plexus lesion with acute denervation findings were observed on EMG.

The patient underwent a rehabilitation program. After eight months from the initial injury, his muscle strength was improved to 3/5 (R/L), 2/5, 0/5 in the shoulder, elbow and hand muscles, respectively. Bilateral C5-T1 dermatomes were hypoesthetic and deep tendon reflexes of bilateral upper limbs were hypoactive. Control electrophysiological study did not reveal any action potentials in the motor and sensory nerve conduction studies for the right upper limb. There was no sensory nerve action potential in the left ulnar and medial antebrachial cutaneous nerve conduction studies. Left median and ulnar nerve motor nerve conduction studies were normal. The latency of the action potentials recorded using the Erb stimulation was prolonged in bilateral axillary, radial, and musculocutaneous nerves. In the needle EMG, there was a significant improvement in all muscles, compared to the initial electrophysiological study, except for the right abductor pollicis brevis and abductor digiti minimi muscles in which no MUAPs were elicited (Tables 1 and 2). A written informed consent was obtained from the patient.

Table 1. Needle electromyography findings in left upper limb

Left	Abductor pollicis brevis	Abductor digiti minimi	Extensor indicis proprius	Deltoid	Triceps brachii	Supraspinatus	Biceps brachii
	Spontaneous activity						
Abnormal spontaneous activity	+	+	++	+	+	+	-
	MUAP analyses						
Amplitude (mV) (0.5-1.5)	0.5-2	0.5-1.5	0.5-1.5	0.5-3	0.5-1.5	0.5-1.5	0.5-1.5
Duration (msec) (5-15)	**5-17**	**5-18**	**5-18**	**5-19**	**5-18**	**5-18**	**5-18**
Polyphasic MUAP ratio	↑	↑	↑↑	↑	↑	↑	↑
Recruitment pattern	↓↓	↓↓	↓↓↓	Normal	↓	Normal	Normal

(-): No abnormal spontaneous activity; (+): Abnormal spontaneous activities, (++++) : Intensive abnormal spontaneous activities; MUAP: Motor unit action potential. Abnormal values are written in bold.

Table 2. Needle electromyography findings in right upper limb

Left	Abductor pollicis brevis	Abductor digiti minimi	Extensor indicis proprius	Deltoid	Triceps brachii	Supraspinatus	Biceps brachii
	Spontaneous activity						
Abnormal spontaneous activity	+	+	++	++	++	+	++
	MUAP analyses						
Amplitude (mV) (0.5-1.5)	*	*	0.5-1.5	**0.5-5**	0.5-1.5	0.5-2	**0.5-5**
Duration (msec) (5-15)			**5-20**	**5-17**	**5-17**	**5-17**	**5-17**
Polyphasic MUAP ratio			↑↑	↑	↑	↑	↑
Recruitment pattern			↓↓↓	↓↓	↓	↓	↓↓

(-): No abnormal spontaneous activity; (+): Abnormal spontaneous activities, (++++): Intensive abnormal spontaneous activities; MUAP: Motor unit action potential; * No MUAPs. Abnormal values are written in bold.

DISCUSSION

3,4-Methylenedioxymethamphetamine (MDMA; ecstasy) is a ring-substituted amphetamine derivative which is also structurally related to the hallucinogenic compound mescaline.[5] It is rapidly absorbed with a half-life of seven hours and metabolized to methylenedioxyamphetamine (MDA).[6] Severe side effects of MDMA abuse include hyperthermia, seizures, hypertensive crisis, arrhythmia, metabolic disorders, disseminated intravascular coagulation, rhabdomyolysis, acute renal failure, hepatotoxicity, cerebrovascular events (cerebral hemorrhage, cerebral infarction), psychiatric disorders, serotonin syndrome, and sudden death.[5,7] Some of these findings such as metabolic disorders, rhabdomyolysis, acute tubular necrosis, and hepatotoxicity were also present in our patient.

Russell et al.[8] reported a 20-year-old female patient who developed rhabdomyolysis, disseminated intravascular coagulopathy, and multi-organ failure induced by MDMA within 24 to 48 hours. Jeon et al.[9] also reported a 57-year-old male case of rhabdomyolysis complicated by lumbosacral plexopathy who was known to be a chronic alcoholic. In this case, nerve conduction studies of both lower limbs and electromyographic studies were found to be normal. However, on Day 4, pelvic MRI showed multiple irregular nodular and geographic lesions in both gluteus minimus and medius muscles, which were slightly hypointense in T1- and hyperintense in T2-weighted images, and showed heterogeneous contrast enhancement after intravenous administration of gadolinium. It was diagnosed as lumbosacral plexopathy, associated with rhabdomyolysis. Unlike Jeon et al.,[9] we did not use upper limb or brachial plexus MRI, although we used cervical and cranial MRI in our case. Similar to our case, the laboratory parameters began to improve on Day 7 and a significant neurological improvement was observed at six months.

Polyneuropathy, brachial, and lumbosacral plexopathy, probably immunological in origin, were already described in heroin users.[10] Dabby et al.[11] described six patients who developed acute peripheral nervous system injury following intravenous or intranasal heroin self-administration. Two of these patients had lumbosacral plexus lesion, two had symmetric distal axonal sensorimotor neuropathy, and the other two had brachial plexus lesion. Similar to our case, five of these six patients had elevated CK levels and rhabdomyolysis and were suspected for a toxic mechanism which could be responsible for acute neuropathy following heroin use.

Unlike the other cases developed following the use of intravenous heroin, rhabdomyolysis and brachial pan-plexus lesion occurred after oral (ecstasy) and intranasal (marijuana) substances in our patient. In the literature, there were cases of plexopathy which developed secondary to heroin or rhabdomyolysis. However, no acute plexus lesions were presented related to the use of MDMA previously. In our patient, plexus involvement was bilateral and very severe which affected all plexus parts. Furthermore, symmetric and complete involvement of the neural structures excluded the possibility of a trauma. It is unlikely that an acute effect of chronic alcohol use may have a role in our patient, since the patient did not have alcohol-related neuropathy findings and the plexus lesion developed acutely with the substance use. However, it is unclear whether the etiology of plexus lesion was the substances or rhabdomyolysis, since both upper limb strength and sensory loss could be recognized after about four to five days following the substance use and

hospitalization. Therefore, bilateral brachial plexus lesion might be directly related to the use of MDMA or marijuana. However, in the literature, we could find no case with a plexus lesion due to marijuana or MDMA use. Similar to the literature, a toxic etiology for peripheral nerve injuries and rhabdomyolysis might be responsible in the development of plexus lesions.

The mechanism and prognosis of plexus lesions due to rhabdomyolysis and substance use have not been clearly understood, yet. In our patient, both clinical and electrophysiological follow-up studies showed an improvement in the upper and middle trunk muscles at eight months after a rehabilitation program. We believe that rehabilitation program can be effective in the recovery process.

In conclusion, substance abuse has been increasing in the society with an increased rate of complications. Even if it is a rare complication, we should keep substance abuse in our mind as the etiology of non-traumatic plexus lesions.

Declaration of conflicting interests

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