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Case Report

Man-In-The-Barrel Syndrome as a Presenting Symptom of Lead Poisoning: Report of Two Cases

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ABSTRACT

Man-In-The-Barrel Syndrome (MIBS), bilateral proximal upper limbs weakness, has a wide range of causes distributed over central and peripheral nervous systems. Rarely, it was reported to be a presentation feature of lead toxicity. Here, we report two cases with MIBS due to lead toxicity that responded well with chelation therapy.

Keywords: Chelation therapy; Man-In-The-Barrel Syndrome; Peripheral nervous systems; Lead toxicity

INTRODUCTION

Man-In-The-Barrel Syndrome (MIBS) is defined as a bilateral brachial muscular weakness, predominantly proximal parts, with near-normal strength of lower extremities [1]. This syndrome may originate from central lesions, including lesions of cerebral or spinal cord, or from peripheral lesions. The typical cause of MIBS is cerebral insults with involvement of bilateral frontal lobes that may have different etiologies including hypoxic-ischemic damage (typically border zone infarction between anterior cerebral and middle cerebral arteries), closed head trauma, brain metastasis, multiple sclerosis, and pontine/extrapontine myelinolysis. In addition, the lesions of cervical spinal cord in the form of infarction, contusion, abscess, neoplasms, and spondylosis may lead to MIBS. Furthermore, some causes involving peripheral nervous system may present with MIBS. These etiologies consist of myopathies, bilateral brachial plexopathy, Amyotrophic Lateral Sclerosis (ALS), HIV-associated motor neuron disease, Multifocal motor neuropathy with conduction block, cervical radiculopathy and myasthenia gravis [2,3].

Lead is a toxic substance and affects multiple organs, especially nervous system. Lead poisoning may have some nonspecific and systemic manifestations such as anorexia, nausea, abdominal pain, constipation and/or anemia. The nervous system, both Central Nervous System (CNS) and Peripheral Nervous System (PNS), may involve specifically. The neurological symptoms include headache, seizure, encephalopathy, short term memory impairment and myalgia. The characteristic features of lead poisoning are motor neuropathy predominantly involving radial and peroneal nerves, polyneuropathy resembles Guillain-Barre Syndrome (GBS), and motor neuron disease [4,5]. Rarely, it was reported that Man-In-The-Barrel Syndrome may be a presentation feature of lead toxicity. Here, we report two cases of sub-acute onset MIBS due to lead toxicity that were significantly responsive to chelation treatment.

CASE PRESENTATION

Patient 1

A 46 years old man presented with weakness at proximal of upper limbs since 2 months before admission in hospital. The weakness had begun insidiously and progressed slowly. In addition, he complained of numbness in distal extremities. No sphincter or bulbar involvement was mentioned. Furthermore, he suffered from abdominal pain and constipation that undergone laparotomy with the diagnosis of appendicitis last month of this admission, which pathology was normal. On the neurologic examination, the muscle force was weak in proximal of upper limbs (Medical Research Council's scale (MRC)=3 out of 5 and distal of lower limbs (MRC=4). Deep Tendon Reflexes (DTR) was diminished and mild hypoesthesia was found at distal extremities.

Laboratory data revealed anemia (hemoglobin=8 g/dl; Mean Corpuscular volume (MCV)=85 fL) and basophilic stippling and teardrop red cells on peripheral blood smear. The findings made the possible diagnosis of lead toxicity and the lead level was requested. The results showed high level of serum lead (45

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mcg/dL; with normal upper limit of 11). All other investigations were normal including for porphyria, vasculitis, other heavy metal toxicities, and paraneoplastic processes. Electrodiagnostic studies revealed subacute axonal sensory-motor polyneuropathy (Table 1). The treatment protocol included beginning with intravenous Ethylene-Diamine-Tetraacetic Acid (EDTA) andconsequent treatment with penicillamine. The follow-up visit showed marked improvement in clinical features within two months.

Table 1: Electrodiagnostic test details.

	Nerve	Motor study			Sensory study	F wave		
		Distal latency (ms)	Amplitude (mV)	Conduction velocity (m/s)	Peak latency (ms)	Amplitude (μV)	Conduction velocity (m/s)	F minimum latency (ms)
Case 1	Median, Rt	3.33	2.2	46	3.3	12.6	47	37
	Median, Lt	3.18	3.4	50	3.21	15.3	49	35
	Ulnar, Rt	2.4	5.6	53	3.05	15.2	48	34
	Ulnar, Lt	2.5	5.7	51	3	9.5	46	33
	Tibial, Rt	3.65	6.9	50	-	-	-	53
	Tibial, Lt	3.91	8.6	54	-	-	-	51
	Peroneal, Rt	4.79	2.1	45	4.1	6.8	42	-
	Peroneal, Lt	3.75	2	39	3.9	6.4	40	-
	Sural, Rt	-	-	-	4	9.6	39	
	Sural, Lt	-	-	-	4.2	9.3	43	
Case 2	Median, Rt	3.91	3.75	42	3.04	38.4	56	33
	Median, Lt	3.75	5.5	39	3.1	36	58	30
	Ulnar, Rt	3.33	3.07	47	2.8	34.6	60	34
	Ulnar, Lt	3.07	9.2	49	3	28.5	62	30
	Tibial, Rt	5.83	5.57	36	-	-	-	54
	Tibial, Lt	5.57	6.3	33	-	-	-	52
	Peroneal, Rt	6.04	3	38	3.9	7	45	-
	Peroneal, Lt	6	3.2	36	4.2	7.2	48	-
	Sural, Rt	-	-	-	3.8	10.1	47	-
	Sural, Lt	-	-	-	4.1	12	50	-

Note: Rt: Right, Lt: Left. Case 1 with spontaneous activity in distal and proximal muscles with normal Motor Unit Action Potentials (MUAPs); subacute axonal type radiculopathy, which is more prominent in upper limbs. Case 2 with spontaneous activity in distal and proximal muscles with decreased recruitment of MUAPs; diffuse motor polyneuropathy which is more prominent in proximal of upper limbs.

Patient 2

A 35 years old man presented with behavioral change, agitation and encephalopathy since 2 weeks before admission. In addition, he had complained from abdominal pain and constipation for several months. The patient was addicted to oral opium since three years ago. On admission, the neurologic examination revealed proximal weakness in all limbs that were more prominent in upper limbs (i.e. MIBS). The muscle strength for proximal muscles of upper and lower limbs were MRC=3 and MRC=4, respectively. The strength of distal muscles was in normal range. The sensory examination revealed

hypoesthesia in distal extremities, as well. The DTRs were all reduced in upper and lower limbs. Electro diagnostic studies showed sub-acute non-length-dependent axonal, sensory and motor, polyneuropathy. Therefore, additional studies were requested. Anemia (hemoglobin=9.5 g/dL, MCV=89 fL) was detected. The lead level was 64 mg/dL, while all other workups were in normal limits. Consequently, he received EDTA for treatment. The muscle strength became significantly improved after one month that he could walk without any difficulty and the strength of upper limbs became MRC=4.

RESULTS AND DISCUSSION

Typically, Man-In-The-Barrel Syndrome (MIBS) is defined to have a central lesion, characteristically after ischemic damage within border zone territories of cerebral arteries. However, this syndrome has diverse etiologies involving central and peripheral nervous system. We report two cases with typical MIBS that had proven diagnosis of lead toxicity and had favorable outcome with chelation therapy.

Lead-induced neuropathy predominantly involves motor fibers. In the case of time course, it may have different presentations including acute (Acute motor axonal neuropathy (GBS-like) with respiratory failure [6] or acute motor neuron disease), subacute, or chronic presentation. In addition, lead can also cause sensory and autonomic neuropathies, especially in more protracted exposures [7]. Lead characteristically cause axonal neuropathy but there are some reports of primarily demyelinating neuropathy in severe toxicity [8]. The exact mechanism of nerve injury in lead toxicity is unknown. One of the proposed mechanisms is destruction of the blood-nerve-barrier with endoneurial edema that may lead to damage to schwann cells and interruption of endoneurial microcirculation [8]. Another theory denotes to substitution of calcium and zinc with lead at the synapse can interfere with neurotransmitter release and signal transduction [9].

We reported two cases presented with subacute onset MIBS, mild encephalopathy, behavioral changes, and anemia that were diagnosed as lead toxicity. The electrodianostic studies revealed subacute axonal sensory-motor polyneuropathy with predominantly involvement of motor fibers. Although lead toxicity typically involve upper extremities in the territory of radial nerves [6], but there are some rare reports of MIBS as a presentation of lead-induced neuropathy. Okhovat and his collegues reported two cases of lead toxicity with MIBS [10]. One of their cases was a 60 years old man with weakness and atrophy of proximal upper limbs lasted for 3 months with electrodianostic evidences of active motor neuron disease in proximal muscles of the upper limbs. The other case was a 65 years old man with acute onset brachial diplegia and six months' history of anemia and GI symptoms. The electrodiagnostic studies revealed acute axonal motor polyradiculoneuropathy. Both cases respond well with chelation therapy [10]. Similar to our second case, both of their cases had a history of opium addiction for 5 to 10 years. Opium addiction along with occupational and environmental sources is an important source of lead poisoning in low-income countries. It seems opium has been contaminated with lead to increase its weight for more profit. In this regards, there were several reports of lead toxicity in opium users recently in our country [11].

CONCLUSION

Man-In-Barrel Syndrome may be a clinical manifestation of lead toxicity. Physicians should be aware of this presentation and put lead toxicity in their differential diagnosis when encounter with patients suffered from MIBS. Therefore, we recommended that lead toxicity should be considered in all patients presented with MIBS, especially if associated with other suggestive features like anemia, abdominal pain and encephalopathy. On the other hand, favorable outcome with chelation therapy in our cases and similar cases suggests that early diagnosis and early treatment of lead toxicity in these situations may prevent irreversible neuronal damage. This issue is notable in countries like Iran where increasing cases of lead poisoning were reported in illicit opium consumers.

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REFERENCES

1. Orsini M, Mello MP, Nascimento OJ, Henrique C, Reis M, Rg De Freitas M, et al. Man-In-The-Barrel Syndrome: History and different etiologies. Rev Neurocienc. 2009;17(2):138-140.
2. Shah P, Wadia P. Reversible Man-In-The-Barrel Syndrome in myasthenia gravis. Ann Indian Acad Neurol. 2016;19(1):99.
3. Akhaddar A, Asmaro K, Pabaney AH, Rammo R, Tahir R, Kole MK, et al. Man-In-The-Barrel Syndrome: Case report of ventral epidural abscess and review of the literature. Surg Neurol Int. 2018;9:8.
4. Soltaninejad K, Shadnia S. Lead poisoning in opium abuser in Iran: A systematic review. Int J Prev Med. 2018;9(1):8.
5. Razmeh S, Abdollahi M, Poranian M. Motor neuron disease due to exposure to lead. Clin Med Diag. 2016;6(5):126-127.
6. Beigmohammadi MT, Aghdashi M, Najafi A, Mojtahedzadeh MO, Karvandian K. Quadriplegia due to lead-contaminated opium. Middle East J Anaesthesiol. 2008;19(6):1411-1416.
7. Mitra P, Sharma S, Purohit P, Sharma P. Clinical and molecular aspects of lead toxicity: An update. Crit Rev Clin Lab Sci. 2017;54(7):506-528.
8. Thomson RM, Parry GJ. Neuropathies associated with excessive exposure to lead. Muscle Nerve. 2006;33(6):732-741.
9. Gordon JN, Taylor A, Bennett PN. Lead poisoning: Case studies. Br J Clin Pharmacol. 2002;53(5):451-458.
10. Okhovat AA, Ansari B, Basiri K, Fatehi F. Neuromuscular manifestations of lead poisoning in opium and herbal users in Iran. Rev Neurol (Paris). 2020;176(10):856-863.
11. Hayatbakhsh MM, Oghabian Z, Conlon E, Nakhaee S, Amirabadizadeh AR, Zahedi MJ, et al. Lead poisoning among opium users in Iran: An emerging health hazard. Subst Abus Treat Prev Policy. 2017;12(1):43.