Organophosphate Intoxication-related Coital-like Involuntary Movements: Report of A Case

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Abstract- The neurologic manifestations of organophosphate intoxication are many, and different pathophysiologic mechanisms are responsible for the different presentations occurring at different stages of the disease process. Movement disorders constitute one of the neurologic manifestations, which may include Parkinsonism or other dyskinetic movements. However, involuntary coital-like movements have not been reported as one of the organophosphate intoxication-related movement disorders. In this case report, we describe a 71-year-old man who developed involuntary coital-like movements about one and a half months after an event of organophosphate intoxication in an attempt to commit suicide. The involuntary movements were to-and-fro pelvic thrusting and back-rocking movements. The patient was able to suppress the involuntary movements for a short period of time, although they usually persisted all day long. The involuntary movements occurred in all postures including standing, sitting and in supine postures, resulting in great embarrassment. These involuntary movements also interfered with the initiation of sleep, although they discontinued while asleep. With clonazepam and piracetam therapy, the involuntary coital-like movements of this patient decreased in amplitude, although remaining to a degree even after one year of follow-up.

Key Words: Involuntary coital-like movements, Organophosphate intoxication

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INTRODUCTION

Organophosphates have been widely used in agriculture and in industry. Since they can be absorbed through ingestion, inhalation and skin exposure, organophosphate intoxication is not an uncommon problem seen in clinical practice. It has been the leading cause of death in suicide patients in Taiwan[1]. The neurologic manifestations of organophosphate intoxication vary, including acute cholinergic crisis and intermediate- and delayed-phase syndromes[2]. The mechanisms responsible for these different neurologic syndromes are also different. Organophosphate intoxication-associated movement disorders have been reported in several

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cases[1,3-5]; however, involuntary coital-like movement has not, until this report, been mentioned among them. In this report, we describe a case of involuntary pelvic movement after an event of attempted suicide with the ingestion of organophosphates.

CASE REPORT

On April 3, 2004, a 71-year-old male, a retired farmer, was sent to our emergency room with severe low back pain and involuntary pelvic thrusting movements. His family history was unremarkable, although he had history of dementia with delusions and hallucinations for four years. He also had been taking olanzapine 5mg four times a day for two years. Physical examination revealed stable vital signs. Neurologic examinations in the emergency room showed clear consciousness and normal cranial nerve examination. His muscle power was symmetrically normal, although deep tendon reflexes were slightly decreased over bilateral knees and ankles. He had bilateral flexor plantar responses. Vibration and joint position sensations were mildly impaired over the bilateral distal lower legs. Primitive reflexes including glabella signs, palmomental reflexes and grasping reflexes were all positive. In addition, there were no "startled responses" to stimuli, sensory triggers or sensory tricks. The involuntary movements of this patient included to-and-fro pelvic thrusting and back-rocking movements, which resembled "coital-like" movements. The dyskinetic movements were confined in the pelvic and lower back, and there were no other associated dyskinetic movements beyond this area. Upon neurologic interview, the patient was able to suppress the involuntary movements for a short period of time, although they usually persisted all day long. The involuntary movements occurred in all postures including standing, sitting and supine postures, resulting in great embarrassment. The involuntary movements also interfered with the initiation of sleep, although they discontinued when asleep. Brain magnetic resonance imaging revealed multiple old infarctions and subcortical leukoariosis. Electromyographic and nerve conduction studies revealed prolonged latency, decreased compound motor action potentials and slow conduction velocities, which were compatible with axonal sensorimotor polyneuropathy. With clonazepam (1mg tid and 2mg hs) and piracetam (400mg qid) therapy, the amplitude of his involuntary pelvic movements decreased, although they persisted to a degree during 13 months of follow-up.

Before the development of involuntary "coital-like" movements, he had attempted suicide by drinking about 250 ml of organophosphate insecticide (demephion) on February 20, 2004. At that time, he was found to have altered consciousness with a Glasgow coma scale of E1V1M1, subsequently receiving emergent intubation on the way to the emergency room. Neurologic examination in the emergency room revealed meiosis without light reflex, fixed extraocular movements and flaccid quadriplegia. The results of the laboratory examination of his blood were as follows: WBC 16100/uL, Hb 17.8 g/dL, Platelets 273000/uL; arterial blood gas: PH 7.113, PaO_2 272.1 mmHg, PCO_2 41.7 mmHg, HCO_3 13 mEq/L, SaO_2 99.4%; glucose 293 mg/dL, blood urine nitrogen/creatine 19 mg/dL/1.3 mg/dL, AST 50 U/L, Na/K 145/3.2 meq/L, and cholinesterase: 414 U/L. He was treated with atropine 5 mg per day for six days and 2-pyridine aldoxime methiodide (PAM) 2-4g per day for 10 days. His cholinesterase levels returned to 4969 U/L on March 8, 2004. His consciousness and muscle strength improved gradually after the first week of admission. His endotracheal tube was successfully removed on March 17, 2004. However, the involuntary pelvic thrusting and back-rocking movements developed gradually and became more and more severe.

DISCUSSION

This 71-year-old man passed through the acute and intermediate stages of organophosphate intoxication[1,6-8] including massive bronchorrhea, excessive sweating, salivation, meiosis, incontinence, muscle twitching and fasciculation, quadriplegia, respiratory failure and coma. As a result of the previous admission, this patient had the following neurologic sequelae of organophosphate intoxication: 1) Polyneuropathy and 2) Involuntary pelvic movements. The polyneuropathy seen in this

patient was of the axonal sensorimotor type, which is compatible with the reported findings of organophosphate intoxication-related polyneuropathy[9], and is known as an effect of inhibition of neuropathy target esterase (NTE) activity in the peripheral nervous system[10,11].

Extrapyramidal symptoms caused by organophosphate intoxication may include Parkinsonism, opisthotonos, torticollis, dystonia, and choreoathetosis [1,3-5], and they usually appear 4 to 40 days after organophosphate exposure, a condition usually reversible several weeks later. The exact pathophysiologic mechanism of these organophosphate intoxication-related extrapyramidal symptoms is unclear, but an imbalance between the cholinergic and dopaminergic system in the central nervous system has been implicated[1]. However, symptoms of involuntary coital-like movements after an event of organophosphate intoxication, as shown in this case, have not been reported before.

Spinal myoclonus involving the abdominal wall and paraspinal muscles resembling belly dancer's dyskinesia has been reported[12]. Sensitivity to stimulus is a common feature of spinal myoclonus, a condition which was not present in our patient. Furthermore, the sustained muscle contraction in our patient was less likely to be myoclonus. Belly dancer's dyskinesias has an unique feature of alternate abdominal muscle contraction, which produces slow, writhing movements of abdomen[13]. The jerky synchronous tortipelvic and back-rocking movements in our patient are not typical manifestations in belly dancer's dyskinesia. Repetitive copulatory rocking movements of pelvic region and the lumbar spine have been described in patients with metoclopramide and antipsychotic-induced tardive dyskinesia. Further electromyographic study has identified this colpulatory movement as truncal tardive dystonia, variants of tardive syndromes[14]. The pelvic thrusting movements of tardive dyskinetic movements are often associated with typical bucco-lingual-facial dyskinesia, laryngeal, respiratory dyskinesia and/or akathisia, which compose a part of tardive syndrome. Other associated dyskinetic movements that have been reported were not present in our case. Chorea often describes an involuntary movement disorder consisting of sudden, rapid, irregular, flowing, nonstereotyped movements that are distally predominant[15]. The absence of a "flowing" phenomenon and the relatively stereotyped pelvic movements in our patient make chorea unlikely. The coital-like movements of this case were unlikely to be related to a psychogenic movement disorder because it persisted even while the patient was walking, yielding to imbalance and falling down if without support.

In the literature[16], a case of "coital-like" pelvic movement resembling rhythmic sleep-related movement disorder--restless leg syndrome, has been described. The authors postulated that the association of this "coital-like" pelvic movement with rhythmic sleep-related movement disorders, such as restless leg syndrome, periodic limb movements in sleep and rhythmic movement disorder, were probably related to a dysfunction of the dopaminergic system at the level of the basal ganglion or the diencephalic-spinal nucleus. The involuntary pelvic to-and-fro thrusting and back-rocking movements, as occurred in our patient, are similar to the rhythmic movements described by Lombardi et al.[16], although the other described abnormal movements during sleep was not found in our patient. Nevertheless, similar pathophysiologic mechanisms may be responsible for the involuntary movements in our case. Organophosphates may bind and inhibit cholinesterase enzymes irreversibly, affecting both nicotinic and muscarinic receptors. This interference may result in excessive cholinergic stimulation and further dopaminergic system dysfunction. Furthermore, chemical or genetic reduction of neuropathy target esterase (NTE) activity may result in a neurologic phenotype of hyperactivity, which has been reported in an animal study[11]. The catalytic activity of NTE has been proposed as a link to the control of motor activity and may contribute to some hyperactivity disorders in humans[11]. A decrease of NTE activity is also well known in patients with organophosphate intoxication, although the pathophysiologic mechanisms of this relationship have yet to be verified.

There is no recommended therapeutic regimen for organophosphate intoxication-related involuntary coital-

like movements. Dopamine agonist, pramipexole, completely abolished the sensory and motor symptoms in a case reported by Lombardi et al[16]. Recent studies suggest dopaminergic dysfunction as the cause of restless leg syndrome, and dopamine agonist has been shown with a therapeutic effect in patients with restless leg syndrome[17]. Although the exact reason is not known, our patient responded partially to combined clonazepam and piracetam therapy. Whether dopaminergic agents are also effective in organophosphate intoxication-related coital-like movements may warrant further large-scale studies experiences.

In conclusion, the exact cause of the involuntary coital-like movements, as shown in our patient, remains unclear; delayed organophosphate-intoxication effect is a possible cause. Research with organophosphates has led to the discovery of several neurologic syndromes and different targets of neurotoxicity[1,2,6,9]. Due to the advancement of emergent management and intensive care, delayed neurologic symptoms will increase in the future. As the leading cause of suicide attempts in Taiwan, more detailed observation for organophosphate-intoxicated patients is needed in order to explore delayed neurologic complications.

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