CASE REPORT

OPSOCLONUS-MYOCLONUS-ATAXIA SYNDROME (OMAS) DUE TO ORGANOPHOSPHATE TOXICITY: CASE REPORT OF A RARE ASSOCIATION

Dur-e-Sabeh[1], Nauman Ismat Butt[2]✉, Muhammad Sohail Ajmal Ghoauri[1], Asfand Yar Ali[1], Fahad Qaisar[1], Muhammad Umair Javed[1]

[1]Bahawal Victoria Hospital, Quaid-e-Azam Medical College Bahawalpur-Pakistan

[2]Azra Naheed Medical College, Superior University Lahore-Pakistan

Opsoclonus-Myoclonus-Ataxia Syndrome (OMAS) is a rare neurological disorder characterized by severe ataxia, multifocal muscle jerky movements and rapid eye oscillatory movements rarely seen as a consequence of organophosphate exposure. A 19-year-old gentleman presented with 2-day history of sudden onset uncontrollable jerky movements of all four limbs, dizziness and vertigo, difficulties in sitting and walking characterized by side-way swaying and increased tendency to fall. There was history of palpitations, vomiting, diarrhea, increased urination, lacrimation and salivation. Four days prior to presentation, there was accidental exposure to chlorpyrifos. On examination, pooling of tears, dribbling of saliva, hypertension, tachycardia, miosis, opsoclonus, myoclonus and ataxia were noted. He was admitted and started on intravenous atropine. MRI scan was done which demonstrated normal cerebellum. He was diagnosed with OMAS caused by Organophosphate Toxicity. By the time of discharge, he was clinically stable and asymptomatic.

Keywords: Opsoclonus-Myoclonus-Ataxia Syndrome (OMAS); Organophosphate Toxicity; Opsoclonus; Myoclonus; Ataxia

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INTRODUCTION

Opsoclonus-Myoclonus-Ataxia Syndrome (OMAS) is a rare neurological disorder usually having a sub-acute or acute course and predominantly affects pediatric population in 2nd year of life.[1] OMAS is characterized by severe ataxia, multifocal muscle jerky movements and rapid eye oscillatory movements. In pediatric population per year, OMAS is seen in 0.2 per 1 million and an even lower incidence is seen in adult population.[2] The etiology of OMAS is not entirely known but it is postulated that immune dysregulation and autoimmunity may play a role in its pathogenesis.[3]

More than half cases of OMAS are paraneoplastic. In pediatric population, OMAS is strongly associated with CNS tumors especially neuroblastoma.[4] However in adults, breast carcinoma and small-cell lung carcinoma are more associated with OMAS.[4] Idiopathic OMAS is more common in adults as compared to children and is usually seen following viral and bacterial illness including salmonella enterica infections, mycoplasma pneumonia, rotavirus, HIV, chicken pox, HCV, mumps and recently SARS-COVID-19 infection.[3,5–7] Very rarely, OMAS may be associated with organophosphate exposure as reported by Haridas *et al*.[8] which remains the sole case report on this rare association available on PubMed.

It is important to diagnose OMAS timely and to look for any underlying malignancy to reduce mortality. In cases of idiopathic OMAS immunosuppressive therapy with corticosteroids, intravenous immunoglobulin, plasmapheresis and rituximab may play a role to improve prognosis.[3]

Herein, we present a case of sudden onset Opsoclonus-myoclonus-ataxia syndrome in a young male after exposure to organophosphate toxicity which is a very rare presentation. However, he recovered readily after treatment with atropine and methylprednisolone.

CASE REPORT

We report the case of a 19 years old gentleman who presented with 2 day history of sudden onset uncontrollable jerky movements of all four limbs.

There was history of dizziness and vertigo for 2 days reported as spinning of his surroundings. Along with this, he reported difficulties in sitting and walking characterized by side-way swaying and increased tendency to fall. There was no history of head trauma, focal sensory or motor loss, loss of consciousness, seizures, photophobia and phonophobia. There was history of palpitations,

vomiting, diarrhea, increased urination, lacrimation and salivation for last 2 days. A farmer by profession, he was unmarried, did not smoke or use illicit drugs.

Four days prior to presentation, there was accidental exposure to chlorpyrifos pesticide spray (an organophosphate) but the patient remained stable initially and did not seek medical attention.

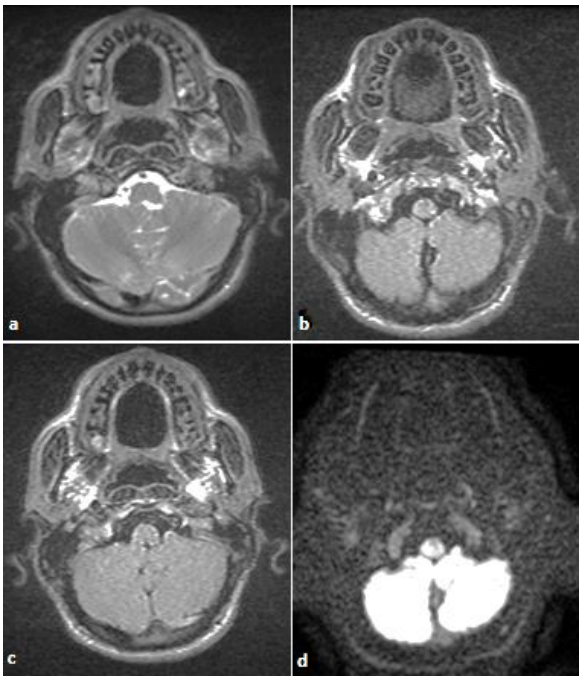

Figure-1: MRI showing no cerebellum abnormalities in T2WI (*a*), FLAIR (b,c) and DWI sequences (*d*)

On examination, a young man with pooling of tears and dribbling of saliva were noted. His pulse was 120 beats per minute, blood pressure 160/100 mmHg and respiratory rate 24 per minute. Pupils were round and equally constricted bilaterally. There were involuntary, rotary, rapid, repetitive conjugate eye movements with no inter-saccadic intervals, oscillopsia, opsoclonus but normal extra-ocular movements in all directions. He had truncal ataxia and an ataxic gait with equal tendency to fall on both sides.

Sudden, involuntary myoclonic movements were also noted during the examination of limbs.

Plantar reflexes were down going bilaterally and signs of neck irritation (Kernig and Brudzinski) were negative. There was no facial asymmetry or focal neurological loss of cranial nerves, sensory or motor systems. Examinations of precordium, chest and abdomen were unremarkable.

He was admitted on basis of organophosphate toxicity and started on intravenous atropine in addition to supportive therapy. His respiratory functions were adequate and he did not require assisted ventilation. Pralidoxime was not prescribed as more than 48 hours had elapsed from organophosphate exposure to hospital presentation.

Due to the neurological features, he was given intravenous methyl-prednisolone 1000 mg once daily for 5 days. MRI scan was done which demonstrated normal cerebellum as shown in Figure-1. Echocardiography and CT scan of chest, abdomen and pelvis were also within normal parameters. After complete atropinization and steroid therapy, the patient recovered and his eye and muscle symptoms had settled. He was subsequently discharged with advice regarding safe handling and use of pesticide. He was asymptomatic at 4 week follow up and was doing his vocational activities efficiently. Based on clinical presentation, radiographic findings and rapid response to treatment, he was diagnosed with Opsoclonus-Myoclonus-Ataxia Syndrome (OMAS) caused by Organophosphate Toxicity. Because of temporal association with organophosphate exposure, response to therapy and monetary constraints, autoimmune antibodies for OMAS were refused by the patient and his attendants.

DISCUSSION

More than half cases of OMAS have an underlying malignancy such as neuroblastoma, breast carcinoma and small-cell lung cancer.[4] In our patient, a malignancy screen compromising of detailed history, clinical examination, MRI brain and CT scan of chest, abdomen and pelvis were negative suggesting idiopathic OMAS. Idiopathic OMAS is usually seen following viral and bacterial illness.[3,5,6,7] There was no history of any recent viral or bacterial illness in our patient. However, the patient had a strong temporal association of OMAS onset with organophosphate (chlorpyrifos) exposure making this the most likely causative factor in the present case. Haridas *et al*.[8]

Reported drugs such as organophosphates and cocaine have been rarely associated with OMAS. Furthermore, organophosphates are a common source of poisoning, both accidental and intentional in tropical countries such as Pakistan and India causing both peripheral and central nervous system manifestations.

Organophosphate pesticides are routinely used by farmers and toxic exposure may occur through ingestion, inhalation or dermal contact.[9] It is estimated that more than 3 million people may be exposed to organophosphates yearly causing more than 300,000 deaths.[10] Organophosphates can stimulate both the sympathetic and parasympathetic nervous systems.[11,12] Features of sympathetic overstimulation include tachycardia, mydriasis,

hypertension, muscle weakness and fasiclulations. Features of parasympathetic overstimulation include bradycardia, miosis, vomiting, bronchospasm, excessive lacrimation, salivation, urination and defecation. Mortality is usually due to respiratory failure caused by respiratory muscle paralysis, CNS respiratory depression, bronchospasm and bronchoconstriction.[10] Neurological symptoms usually start 24–96 hours after exposure.[11,12]

Atropine, which competes with acetylcholinesterase at muscarinic receptors, is the definite treatment in cases of organophosphate toxicity starting with an initial dose of 2–5 mg intravenously and doubling the dose every 3–5 minutes until respiratory secretions have cleared and bronchospasm has resolved.[13] Pralidoxime, which works on muscarinic receptors, is only recommended to be given within 48 hours after exposure to organophosphate.[14] Our patient presented after more than 48 hours since organophosphate exposure, therefore pralidoxime was not given. The exact etiology of OMAS is not known but immune dysregulation and autoimmunity are thought to play a role in its pathogenesis. Therefore, in cases of idiopathic OMAS immunosuppressive therapy with corticosteroids, intravenous immunoglobulin, plasmapheresis and rituximab is usually employed.[3,15]

In the present case, intravenous methylprednisolone was prescribed in addition to atropine therapy for organophosphate toxicity. Our patient showed dramatic response with therapy and was asymptomatic at the time of discharge and 4-week follow-up.

Consent

Detailed informed consent was taken from the patient and his father prior to data collection and manuscript writing.

Conflict of interest

None

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Address for Correspondence:

Nauman Ismat Butt, Department of Medicine & Allied, Azra Naheed Medial College, Superior University Lahore-Pakistan

Cell: +92 345 465 1049

Email: nauman_ib@yahoo.com

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