CASE REPORT

Weakness and Diplopia in a 4-Year Old Boy. An Unusual Diagnosis

How to Cite This Article: Alizadeh A, Jajarmi H, Saeed Zand N. Weakness and Diplopia in a 4-Year Old Boy. An Unusual Diagnosis. Iran J Child Neurol. Autumn 2022; 16(4): 75-79

Anahita ALIZADEH MD[1],
Homa JAJARMI MD[2],
Nazanin SAEED ZAND MD[2]

1.Fellowship of Clinical Toxicology, Department of Clinical Toxicology, MTRC Medical Toxicology Research Center, Mashhad University of Medical Sciences, Mashhad, Iran
2. Department of Pediatric, Mashhad University of Medical Sciences, Mashhad, Iran

Corresponding Author
Alizadeh A .MD
Pediatrician, Fellowship of Clinical Toxicology, Mashhad University of Medical sciences, Mashhad, Iran
Email: Alizadehan@mums.ac.ir

Received: 14-Nov-2020
Accepted: 12-Jul-2021
Published: 23-oct-2022

Abstract

Ptosis, diplopia, and overall weakness in children may have serious underlying causes such as myasthenia, botulism, Guillain-Barré syndrome, and poisoning, which require a systematic and timely evaluation and proper management.(1,2,3) In pediatrics, clinical presentations may be atypical, and diagnostic overlap sometimes makes the final diagnosis challenging. This can be addressed through taking accurate history, performing a physical exam, conducting a comprehensive assessment, and using appropriate diagnostic algorithms.

Keywords: Children; Poisoning; lathyrus sativus
DOI: 10.22037/ijcn.v16i4.32888

Case presentation

A 4-year-old boy was evaluated for a 2-day history of bilateral ptosis and diplopia. He also reported having had ataxia and bilateral lower limb weakness, all began after ingestion of an unknown herb two days ago. He had no history of the consumption of canned foods. There were no prodromal symptoms. There was no myalgia, sensory symptoms, or sphincter malfunction. On admission, the patient had an ataxic gate, and his examination showed normal pupils reactive to light, bilateral ptosis, and ophthalmoplegia (left and right lateral rectus palsy). DTR was detected in 4 limbs, but the muscle power was decreased bilaterally (2/5) in the lower limbs. The Babinski sign was not seen on the right and was equivocal on the left side. Examination of other cranial nerves was normal, and there was no sensory deficit. (The patient had temporary low back pain. All vital signs were normal. The gag reflex was normal.

Over the 3-day course of his hospitalization, DTR in the lower limbs became undetectable and muscle power in the upper limbs

also significantly decreased by 2/5. He did not show any sign of respiratory involvement or new neurological manifestations.

The patient was evaluated for acute overall weakness, and investigations showed normal blood count, liver and renal function tests, electrolytes, blood gas, and CSF analysis which was performed one week after the onset of the symptoms. (Table 1.) Brain MRI was also unremarkable, but spinal MRI with GAD showed cauda equine nerve root enhancement. The Nerve conduction study demonstrated pure motor polyneuropathy.

The patient was treated with IVIG (1gr/kg for three days) and showed mild improvements in his weakness and diplopia. He was discharged early on parental consent with the recommendation to start physiotherapy. At discharge, Ophthalmoplegia was diminished, but mild ataxia and hypotonia remained. After ten therapy sessions and a 5-month follow-up appointment, he fully recovered, and all symptoms disappeared.

Fig 1. *lathyrus sativus. Was there any relationship between the ingestion of the herb by the patient and his symptoms? (The patient had a history of consumption of an unknown amount of the plant Lathyrus sativus two days before the disease onset*

Fig 2. *lathyrus sativus. The patient had poly neuropathy according to electrophysiology results and also ataxia and ophthalmoplegia, areflexia, and a history of a *plant ingestion (*Lathyrus sativus; Kheler or Gav terkunak)*

Table 1: Laboratory results

Laboratory test	Patient levels	Normal range
WBC (cell/µL)	15.300	4000-11,000
Neutrophil (%)	55.9	33-42
Lymphocyte (%)	35.3	50-59
RBC (cell/µL)	4,100,000	3,800,000-5,500,000
Hb (g/dL)	11.4	11.5-13.5
HCT (%)	35	34-40
PLT (cell/µL)	230,000	150,000-450,000
PT (sec)	12	10-11.30
PTT (sec)	20	24-36
BS (mg/dL)	120	60-100
Na (mEq/L)	134	135-145
K (mEq/L)	3.5	3.4-4.7
Cl (mEq/L)	109	98-107
Mg (mg/dL)	1.9	1.6-2.4
Ca (mg/dL)	8.8	9-11
Urea (mg/dL)	20	25-40
Cr (mg/dL)	0.5	0.3-0.7
AST (U/L)	31	20-60
ALT (U/L)	15	25-40
PH	7.37	7.35-7.45
Pco2 (mmHg)	38	35-45
Hco3 (mEq/L)	20	22-26
ESR (mm/hr)	15	4-20
CRP (mg/dL)	Negative	0-0.5
Lumbar punction	WBC =0 RBC =0 Glucose=89 Pt =45	Normal

Discussion

Was there any relationship between the ingestion of the herb by the patient and his symptoms? (The patient had a history of consumption of an unknown amount of the plant Lathyrus sativus two days before the disease onset (Fig 1,2)

Guillain-Barre syndrome (GBS) is an autoimmune disturbance with a possible infection trigger (2). The weakness usually follows a nonspecific gastrointestinal or respiratory infection

approximately ten days before. Polyneuropathy mainly involves motor neurons, but sensory neurons and autonomic nerves are sometimes involved. Weakness usually begins in the lower extremities and ascends, progressively involving the trunk, upper limbs, and bulbar muscles. But the involvement of the extraocular forces is rare. Ophthalmoplegia and ataxia are not common in the classic Guillain-Barre syndrome, but some variants of GBS, such as Miller-Fisher syndrome, may be seen (4). Miller-Fisher syndrome (MFS) is an uncommon GBS variant associated with ophthalmoplegia, ataxia, and areflexia. Weakness of the extremities is less common than GBS. (4)

In GBS, on magnetic resonance imaging (MRI) of the spinal cord, typical findings include thickening of the cauda equina and intrathecal nerve roots with gadolinium enhancement. (2, 4)

Nerve conduction studies and electromyography are helpful for the early diagnosis of peripheral nerve inflammation in GBS. Motor and sensory nerve conduction velocities are reduced in this disorder. (2)

Furthermore, many poisons, toxins (poisonous substances from plants or animals), drugs, metabolic diseases, metals, and some infections can cause peripheral neuropathy. (3)

The patient had poly neuropathy according to electrophysiology results and also ataxia and ophthalmoplegia, areflexia, and a history of a *plant ingestion (*Lathyrus sativus; Kheler or Gav terkunak)(Fig 1,2)

Lathyrus sativus has been used as food by poor farmers in India, Bangladesh, Nepal, China, and Ethiopia. Generally, during droughts and famines, it is consumed as a staple food as it sustains the harshest of agro-climatic conditions. It is a nutritious legume rich in protein with reasonable quantities of essential amino acids. Although it has a high nutritional value, the presence of neurotoxic chemicals limits its use as daily food. This plant contains toxic substances such as ODAB, and L-ODAP (β-N-oxalyl-l-α diamino propionic acid), which is the neurotoxic amino acid of this plant. This glutamate analog is harmful to humans. There are several reports of different types of neurologic disorders (such as spastic paraparesis) induced by Lathyrus sativus, which is known as neurolathyrism (5-8)

The toxins of Lathyrus sativus can induce neurolathyrism. Three modes of presentation of neurolathyrism have been described in humans, the most common being a sudden onset of leg weakness on falling asleep or awakening.

Some patients complain of subacute onset of walking difficulty, whereas others experience a gradual progression of spastic paraparesis extending over months. (6, 7, 8)

Overall, we believe that the presented scenario was a case of neurolathyrism as Guillain-Barre syndrome, from the Miller Fisher subtype. Unfortunately, the Miller Fisher antibody assay was not available in our center.

Conclusion

In every child with unusual presentations, poisoning should be considered one of the most probable diagnoses. Some herbs can induce severe and dangerous poisoning, therefore, identifying the clinical symptoms and using appropriate diagnostic algorithms is essential for timely diagnosis and proper treatment of specific cases of toxicity. Moreover, physicians, health personnel, veterinarians, and parents' awareness about possible dangerous plants is critical and should be included in educational programs.

Acknowledgment

I would like to thank my patient's parents for their information and cooperation. Also, many thanks to my dear friend Dr. Sepideh Tabarestani for her guidance in writing this article

Author's Contribution

All of authors participated in gathering data and diagnosis and writing the article

Conflict of Interest

There is no conflict of interest

References

1. Bharucha-Goebel DX Disorders of Neuromuscular Transmission and of Motor Neuron myasthenia gravis, Kliegman RM, Blum N J, Shah S, Geme J W ST , Tasker RC, Wilson KM, Behrman RE in Nelson Textbook of Pediatrics Elsevier 21th ed 2020; chapter 630, 3305-3307
2. Rya M M Guillain-Barré Syndrome. Kliegman RM, Blum N J, Shah S, Geme J W ST , Tasker RC, Wilson KM, Behrman RE in Nelson Textbook of Pediatrics Elsevier 21th ed 2020; chapter 634 p 3335 e
3. Sarnat HB Kliegman RM, Blum N J, Shah S, Geme J W ST , Tasker RC, Wilson KM, Behrman RE in Nelson Textbook of Pediatrics Elsevier 21th ed 2020; Toxic Neuropathies Chapter 632 p3330-3333
4. Mehndiratta MM, Wadhwa A. Electrophysiological features of Gullian Barre syndrome: Newer insights. Neurol India 2016;64:921-2
5. Zasshi Y Review [Research in Motor Neuron Diseases Caused by Natural Substances: Focus on Pathological Mechanisms of Neurolathyrism] . 2019;139(4):609-615. [Article in Japanese] The Pharmaceutical Society of Japan
6. Singh SS , Rao SLN Lessons from neurolathyrism: A disease of the past & the future of Lathyrus sativus (Khesari dal) Indian J Med Res 138, July 2013, pp 32-37
7. Yan ZY,. Spencer P S, Li ZX, Liang YM, Wang YF, Wang CY, Li FM Lathyrus sativus (grass pea) and its neurotoxin ODAP Review Phytochemistry Volume 67, Issue 2, January 2006, Pages 107-12 Lathyrus sativus (grass pea) and its neurotoxin ODAP, Phytochemistry Issue 2, January 2006;Vol 67 Pages 107-121
8. Yadav M Lathyrus sativus - a path to Neurolathyrism Health and Medicine 2018 27 Feb