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Case Report

Moldy sugarcane-induced toxic encephalopathy in a child: a case report

Xin Chen, Wansheng Peng, Lei Wang, Sehua Qu, Zhouzhi Ding

Department of Pediatrics, The First Affiliated Hospital of Bengbu Medical College, Bengbu 233004, Anhui, China

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Abstract: A female child aged 2 years 3 months developed toxic encephalopathy 3 h after consuming moldy sugarcane. She presented with convulsions and coma, followed by aphasia and paralysis, as well as extrapyramidal symptoms, including nystagmus, dystonia, and spasmodic torticollis. Cranial magnetic resonance imaging (MRI) revealed symmetrical, fan-shaped, long, abnormal T2 signals in the globus pallidus bilaterally. This was a typical case of toxic encephalopathy caused by moldy sugarcane. Several case reports on toxic encephalopathy have been published in China, but none have reported the typical cranial MRI findings of the disease.

Keywords: Sugarcane, mold, food poisoning, encephalopathy, magnetic resonance imaging

Introduction

The consumption of moldy sugarcane may lead to toxic encephalopathy and extensive damage to the central nervous system, resulting in disability and, possibly, death [1-4]. Several case reports on this topic have been published in China and the intoxication is relatively common in 13 of 34 Provinces in China (**Table 1**) [1, 5, 6]; however, none have reported cranial magnetic resonance imaging (MRI) results. We report a case of a young child who had toxic encephalopathy after consuming moldy sugarcane.

Case report

A female child aged 2 years 3 months was admitted at our hospital on May 3rd, 2014. Twenty minutes prior to admission, the child had a sudden onset of frequent but non-projectile vomiting of gastric contents, followed by convulsions manifested as tonic-clonic seizures. The patient was taken to the emergency department after the intravenous injection of 3 mg of diazepam for convulsion control. The patient had urine and fecal incontinence (no abnormal appearance) and no fever. The child had consumed "reddish" sugarcane at home 3 h prior to the onset of symptoms. On admission, a physical examination revealed a body temperature of 37°C, respiratory rate of 28 breaths per min, heart rate of 120 beats per min, and blood pressure of 85/45 mmHg. Her body weight was 12.5 kg and length was 85 cm. The patient had coma (Glasgow coma score, 12 points) and smooth breathing. No jaundice, rash, or hemorrhagic spots were observed on the skin or mucosa. Her pupils were round, with a diameter of approximately 4 mm, and reactive to light. Her neck had no rigidity. No abnormalities were found in the heart, chest, or abdomen. Decreased muscle tone of the extremities was observed (muscle tension, 0 level), and the extremities responded after stimulation. The patellar reflex was bilaterally positive with negative pathological signs.

The primary diagnosis was poisoning with moldy sugarcane ("reddish" sugarcane). The patient was subjected to gastric lavage, catharsis, fluid infusions, and diuretics to reduce the intracranial pressure. No abnormalities were found in the blood indexes, electrolyte levels, liver and kidney functions, or myocardial enzymes. We contacted the Provincial Center for Disease Control and Prevention and requested the family members to send a sample of the remaining "reddish" sugarcane for analyses. On the second day of hospitalization, the child

Table 1. Cases of moldy sugarcane poisoning reported in the English literature

Reference	Summary
Liu et al. [5]	Moldy sugarcane poisoning is a fatal food poisoning that is epidemic in 13 provinces in China. 3-nitropropionic acid was identified as the causative agent.
Ming [1]	The main symptom was dystonia. Cranial CT showed bilateral lenticular lucencies.
Present case report	The main symptoms were aphasia, nystagmus, and flaccid paralysis of the extremities. CT was normal, but MRI revealed symmetrical, fan-shaped, long, abnormal T2 signals in the bilateral globus pallidus.

CT: computed tomography; MRI: magnetic resonance imaging.

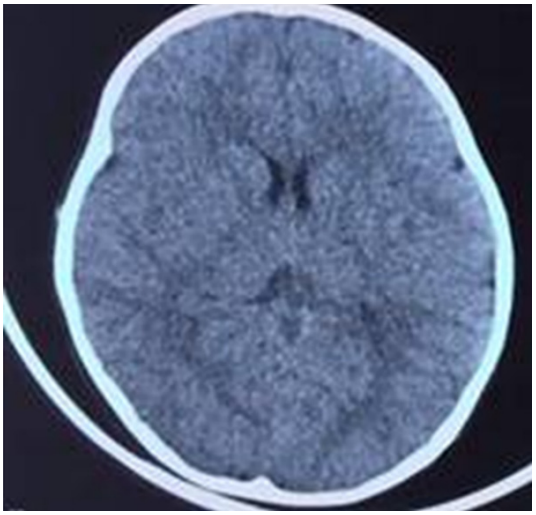

Figure 1. Cranial CT scan showing no obvious abnormalities.

regained consciousness but had aphasia, nystagmus, and flaccid paralysis of the extremities. The myodynamia of the extremities was grade II with decreased muscle tone and bilateral patellar hyporeflexia. No marked abnormalities were detected in the cranial computed tomography (CT) scan (day 2; **Figure 1**), routine cerebrospinal fluid tests, or biochemical analyses (day 2). The cranial MRI (day 3) revealed a symmetrical, fan-shaped, long, abnormal T2 signals in the bilateral globus pallidus (**Figure 2**). On the fourth day of hospitalization, the Provincial Center for Disease Control and Prevention reported that 3-nitropropionic acid (3-NPA) was detected in the sugarcane sample. Therefore, the patient was diagnosed with toxic encephalopathy caused by moldy sugarcane and treated with hyperbaric oxygen therapy. On the 12th day of hospitalization, the child still manifested aphasia and nystagmus. The flaccid paralysis of her extremities gradually progressed to spastic paralysis. The myodynamia of the extremities was grade III with increased muscle tone and bilateral patellar hyperreflexia accompanied by spasmodic torticollis. The patient was treated with oral benzhexol hydrochloride (1 mg tid) to relieve the extrapyramidal symptoms. The child was followed up for 2 years; her verbal function and myodynamia had almost returned to normal with residual nystagmus and increased muscle tone of the extremities.

Discussion

Arthrinium spp., the main pathogen in moldy sugarcane, produces 3-NPA, which is a neurotoxin [3-5]. Upon absorption, the toxin passes through the blood-brain barrier within a short time, causing changes in the nigrostriatal dopaminergic system and striatal degeneration or necrosis [7]. 3-NPA inhibits the production of energy, which leads to adenosine triphosphate depletion and neuronal excitability poisoning [8]. The latency period of symptom onset normally ranges from 15 min to 8 h; however, up to 48 h has been reported. The main manifestations of mild poisoning include functional disturbances of the gastrointestinal tract, such as nausea, vomiting, abdominal pain, and occasional diarrhea, which may be accompanied by headaches, dizziness, and blurred vision. Generally, the symptoms are rapidly relieved. Patients with moderate poisoning may have paroxysmal and tonic convulsions, unconsciousness, motor aphasia, nystagmus, upward gaze, visual hallucinations, enlargement or contraction of the pupils, and tendon hyperreflexia. Routine cerebrospinal fluid testing and biochemical analyses are generally normal. Ocular fundus is normal, but retina edema is common. Patients gradually recover within 1-2 weeks but may have possible sequelae, including speech disorders, consciousness disturbances, and dyskinesia. The main manifestations of severe poisoning include not only aggravated symp-

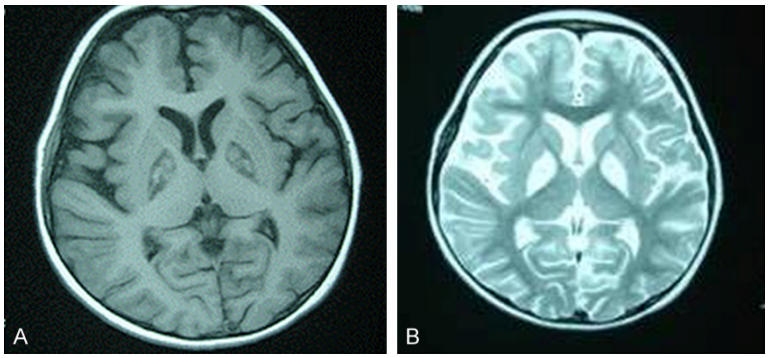

Figure 2. Cranial MRI results in a child who consumed moldy sugarcane. A. T1-weighted MRI scan (T1W1). B. T2-weighted MRI scan (T2W1). The lesions in the bilateral globus pallidus consisted of mixed low-intensity signals on T1W1 and high-intensity signals on T2W1.

toms but also deep coma, status epilepticus, and fever. Hematuria, black tarry stools, and pulmonary edema are not rare during the course of the disease. Patients often die from respiratory failure. Children who survive tend to have severe sequelae of the nervous system, especially of the extrapyramidal system. Cranial CT scans of children with moderate or severe poisoning reveal comparatively symmetrical low-intensity lesions in the bilateral basal ganglia with significant changes in the globus pallidus. Spotty and patchy bleeding may be present in these lesions; diffuse brain atrophy is observed during the late period [9]. Currently, cranial MRI findings from patients with moldy sugarcane poisoning have not been reported yet. Nevertheless, Roberts found symmetrical, long, abnormal T2 signals in the bilateral striatum on cranial MRI in rats with 3-NPA poisoning [7]. MRI may be superior to CT because it unveils lesions that CT generally misses during the early period. In this case report, the CT scan was normal on the second day after symptom onset, while the MRI scan revealed symmetrical, fan-shaped, long, abnormal T2 signals in the bilateral globus pallidus, which was consistent with the findings of Roberts. Currently, no effective therapeutic agents are available for the treatment of moldy sugarcane poisoning. The main therapies include immediate gastrointestinal lavage and catharsis, fluid infusions to treat dehydration and acidosis, sedatives for the convulsions, diuretics and hyperbaric oxygen for the cerebral edema, and antifungal agents. Recent animal studies revealed different treatments that could have some efficacy against 3-nitropropionic acid poisoning, but they have yet to be tried in humans [10-15]. The child in this case report had moderate poisoning. Despite aggressive treatment, the extrapyramidal symptoms remained 2 years after symptom onset. her prognosis is not optimistic.

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Disclosure of conflict of interest

None.

Address correspondence to: Xin Chen, Department of Pediatrics, The First Affiliated Hospital of Bengbu Medical College, 287 Changhuai Road, Bengbu 233004, Anhui, China. Tel: +86-13965263225; Fax: +86-21-57643271; E-mail: 44783939@qq.com

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