Chorea as the First Neurological Symptom of Delayed Encephalopathy after Carbon Monoxide Intoxication

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Abstract

Delayed encephalopathy after carbon monoxide (CO) intoxication involves a triad of characteristic symptoms consisting of mental deterioration, urinary incontinence, and gait disturbance. Additionally, while it is not normally associated with involuntary movement, delayed encephalopathy after CO intoxication can also, in extremely rare cases, lead to chorea. We report a patient with delayed encephalopathy due to CO intoxication, suffering from chorea involving a unilateral lower limb, and subsequent cognitive impairment and other extrapyramidal symptoms.

Key words: carbon monoxide, delayed encephalopathy, chorea


Introduction

Delayed encephalopathy after carbon monoxide (CO) intoxication is clinically characterized by deterioration and relapse of cognitive ability and behavioral movement (1, 2). This recurrence is preceded by a temporary asymptomatic period of variable duration (from 2 to 40 days) following recovery from the acute stage of CO intoxication (1, 3). Delayed encephalopathy after CO intoxication involves a triad of characteristic symptoms consisting of mental deterioration, urinary incontinence, and gait disturbance (4). Additionally, while delayed encephalopathy is not normally associated with involuntary movement, it could also lead to chorea in rare cases (2, 5, 6). We encountered a young patient experiencing chorea of a unilateral lower limb as the first manifestation of delayed encephalopathy following acute CO intoxication, which had occurred 20 days after the acute intoxication.

Case Report

A 16-year-old man was admitted to the emergency room of the nearest general hospital, his chief complaint being of stupor consciousness. He had been found by rescue team in a closed room, which was full of briquette gas. On the basis of first admission hospital reports, we assumed that he attempted to commit suicide by briquette gas inhalation. But the exposure time of the gas was not clear and there was no drug ingestion history. His blood pressure was 110/70, heart rate 85/min, and respiration rate 22/min. He was immediately treated with hyperbaric oxygen therapy under the diagnosis of acute CO intoxication, a diagnosis which was based on a complete history and an examination of his blood carboxyhemoglobin levels (17%). The brain CT performed on the day of admission and EEG performed at second admission demonstrated no definite abnormal findings except for a small amount of symmetrical theta waves on both hemispheres. Two days later, he had completely recovered and was discharged after the 8th day. However, by the 20th day after acute insult, restlessness and involuntary movements of the right foot and leg developed but his mental state was alert and language functions, including comprehension and fluency, were normal, according to the statement of his parents. Gait disturbance and cognitive impairment followed a few days later. Based on this, he was admitted to the movement department of our hospital on the 25th day after acute insult. He had no past history or family history of hypertension, diabetes, cardiac disease, liver disease, or neurological disorders such as epilepsy. On admission, he was alert, but confused, and his cognitive functions were impaired. A neuropsychological test was performed on the day of admission;
Figure 1. Brain magnetic resonance images demonstrate high signal intensities from bilateral periventricular white matter and deep white matter in T2-weighted (A) and diffusion-weighted images (B) and low signal intensities from a comparable area in diffusion coefficient maps (C).

A

B

C

mini-mental state examination (MMSE) score was 8 and clinical dementia rate (CDR) was 2 (sum of box: 11). Involuntary, non-rhythmic, brief and rapid movements were evident in the right leg, ankle and foot. He also exhibited a short step gait with decreased arm swing and increased muscle tone; however, there was no indication of motor weakness, sensory change, ankle clonus, or Babinski signs. Laboratory findings from a complete cell count, urinalysis, serum electrolytes analysis, thyroid function test, and chemical work-up were normal. Chest X-ray and electrocardiography were also normal; however, electroencephalography demonstrated diffuse mixed slowing waves in both hemispheres. The brain magnetic resonance image (MRI) revealed high signal intensities from the bilateral periventricular white matter and deep white matter in T2-weighted and diffusion-weighted images and low signal intensities in apparent diffusion coefficient (ADC) maps, a finding which is suggestive of late developing cytotoxic edema (Fig. 1).

Risperdal (1 mg/day) was prescribed, and after three days, the choreic movement of the right lower limb ceased. In response to cognitive decline, we selected donepezil hydrochloride, a drug thought to prevent the breakdown of acetylcholine, a neurotransmitter in the brain that is important in memory function. On the 18th hospital day, follow-up MMSE and CDR score showed 9 and 2, respectively. Ultimately, there was no improvement for any symptom other than choreic movement and the patient was discharged on 20th hospital day.

Discussion

Carbon monoxide intoxication is the leading cause of lethal poisoning in the world; however, the incidence of delayed encephalopathy following CO intoxication is low, ranging from 0.06% to 2.8% of all cases of CO intoxication (3). Neurological and psychiatric symptoms of delayed encephalopathy including mental deterioration, parkinsonism, gait disturbance, speech disorder, agnosia, ataxia, epileptic seizure, apraxia, and amnesic disturbances may occur (7). Even though neurologic extrapyramidal symptoms such as parkinsonism are well-recognized CO intoxication sequelae, movement disorders after CO intoxication are rare (8). Furthermore, the incidence of movement disorder as CO intoxication sequelae is still unclear (8).

Chorea is symptomatic of many diseases of the nervous system, although most of them are related to rheumatic fever. It is a major feature of Huntington’s chorea, in which the movements tend to be more typically choreoathetotic. Intoxication with phenothiazine drugs or haloperidol and rarely, hyperthyroidism, polycythemia vera, lupus erythematosus, or stroke may also cause chorea (6). Despite this, the incidence of chorea after CO poisoning is still unclear, and may be extremely rare (2, 5, 6). For example, previous studies have demonstrated that in two series of 2,360 patients and 21,143 patients with CO intoxication, none had chorea (4, 9). Further, another study regarding delayed-onset
movement disorders following CO intoxication demonstrated that only three patients among 242 with delayed encephalopathy due to CO intoxication had chorea (2). It is known that chorea, induced by CO intoxication, occurs mainly at ages younger than 40 years and women are more often affected (6). Additionally, it is understood that the duration of chorea after CO intoxication can range from days to months, and that it is usually alleviated through the use of neuroleptic agents (6). In the present case, the choreic movement involved only one lower extremity as an initial manifestation of the delayed neurological sequelae after CO poisoning, an unusual feature compared with previous reports. In contrast, similar to previous reports, the present case demonstrated the disappearance of choreic movement of the unilateral lower limb at three days after administration of a neuroleptic agent.

With regard to anatomical lesions, there is no pathological predilection for chorea at any site, although it commonly suggested that chorea may be associated with the caudate nucleus (2, 5). Furthermore, prior studies have suggested no correlation between neuroimaging findings and the development of chorea. This conclusion was based on the fact that brain images in that study revealed bilateral lesions in the basal ganglia and white matter of the cerebral cortex; however, these lesions can also be seen in non-choreic patients with CO poisoning (6). In the present case, there were no basal ganglia lesions, only bilateral white matter. The first symptom of delayed encephalopathy was the choreic movement of a unilateral lower limb. In addition, the pathophysiological patterns of choreic movement may likely result from excessive dopaminergic output on the receptors of the striatum given that chorea is alleviated by neuroleptic agents.

We encountered a patient with extremely rare unilateral lower limb chorea as the first manifestation of delayed encephalopathy after CO intoxication. Further study is necessary to clarify the lesion sites and pathogenesis of involuntary movements in this form of delayed encephalopathy.

References

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