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CASE REPORT

Acute alcohol intoxication presenting acquired lesion of the corpus callosum in a young healthy woman: A case of possible Marchiafava–Bignami disease

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Abstract

Background: Marchiafava–Bignami disease is a rare neurological disease characterized by acquired lesions of the corpus callosum. Although the major causative etiology is chronic alcoholism, a case caused by acute alcohol intoxication has not yet been reported.

Case Presentation: A 19-year-old female with no known medical history or a history of chronic alcohol consumption was brought to the emergency department in a coma after binge alcohol consumption. Even after an overnight observation, she remained comatose. After a thorough examination including magnetic resonance imaging, which showed lesions of the corpus callosum, she was treated with thiamine for Marchiafava–Bignami disease. She recovered completely and at the follow-up, the callosum lesion had resolved.

Conclusion: This is a rare case within the spectrum of Marchiafava–Bignami disease caused by acute consumption of alcohol. Clinicians should be aware of this potentially devastating critical condition among patients with severe alcohol intoxication, which might have been overlooked.

KEYWORDS
acute alcohol intoxication, Marchiafava-Bignami disease

INTRODUCTION

Marchiafava–Bignami disease (MBD) is a rare neurological disease characterized by primary degeneration of the corpus callosum. The disease has a high morbidity and mortality.[1–3] Early recognition of this disease is critical because some patients may benefit from early treatment.[2,4] Diagnosis is based on medical history, clinical findings, and magnetic resonance imaging (MRI) of acquired lesions of the corpus callosum (ALCC). Since the psychomotor symptoms are nonspecific, the suspicion of MBD is usually arise from a medical history of chronic alcoholism and malnutrition. However, recent reports have highlighted various etiologies of MBD making diagnosis more complex. Here, we report a perplexing case of an acute alcohol-induced comatose patient presenting with ALCC on MRI, which was considered to be within the spectrum of MBD. To the best of our knowledge, this is the first reported case of MBD caused by acute alcohol intoxication in a young healthy patient.

CASE REPORT

A 19-year-old female was found unconscious in a commercial complex and was brought to our emergency department (ED) by the emergency medical service. It was unclear why the

patient was unconscious at the location at which she was found because there were no witnesses. According to the patient's friend, who accompanied her at the party, the patient had started drinking alcohol 2 h before the emergency call. Prior to copious alcohol consumption, the patient was reported to behave normally. Additionally, her parents and friend informed us that she was a social drinker but had no history of alcohol abuse, medication, or any relevant medical history. In the ED, the patient's Glasgow coma scale (GCS) was three, blood pressure was 107/51 mmHg, pulse rate was 75 beats/min, body temperature was 36.5°C, respiratory rate was 24 breaths/min with a patent airway and a normal breathing pattern, and oxygen saturation was 95% in room air. Although we did not measure the exact body weight in the ED, the patient's body mass index was 20.6 after admission. During the physical examination, only slight abrasions were observed on the face and ankle. Laboratory tests showed slight hypokalemia (3.3 mEq/L), although albumin, total protein, liver enzymes, blood urea nitrogen, creatinine, sodium, magnesium, glucose, hemoglobin, mean cell volume, white blood cell count, and C-reactive protein levels were within the normal range. Blood gas tests showed slight acidemia (pH 7.330) with elevated lactate (2.6 mmol/L) and an anion gap (19.2 mmol/L). Urine toxicology screening test results were negative for methamphetamine, tetrahydrocannabinol, cocaine, opiates, benzodiazepines, barbiturates, and tricyclic antidepressants. Computed tomography of the head revealed no abnormal findings. Based on these findings and the history of binge alcohol consumption at the party, we made a tentative diagnosis of acute alcohol intoxication with dehydration. Normal saline was administered, and the patient was admitted to the observational unit. However, the patient remained in a coma for 12 h after admission. Therefore, detailed investigations were conducted. The cerebrospinal fluid (CSF) test results were normal. Electroencephalography (EEG) showed diffuse slowing of background activity with poor response to stimuli but no epileptiform discharge. The blood gas analysis revealed a mixed disorder of metabolic and respiratory acidemia (pH 7.287, $HCO3^-$ 0.3 mmol/L, BE −6.2 mmol/L, pCO2 42.5 mmHg) with elevated lactate (3.3 mmol/L), although glucose was within the normal range (111 mg/dL). A change in electrolyte concentration from admission was also noted: sodium (from 140 to 145 mEq/L), potassium (from 3.9 to 3.3 mEq/L), chloride (from 104 to 110 mEq/L). Subsequently, the maintenance fluid was modified to a hypotonic solution containing potassium. At this moment (approximately 18 h after admission, just preceding the MRI), the net fluid balance was negative, with a total of 1500 mL of normal saline infusion and 1700 mL of urine output. Brain MRI showed an area of high signal intensity on diffusion-weighted imaging (DWI) with a decreased apparent diffusion coefficient (ADC) in the corpus callosum and hemispheric white matter but showed no abnormal findings on the other sequences (Figure 1). Additionally, we checked the blood alcohol concentration (BAC) at that time (20 h after admission) which was extremely high (435 mg/dL), and vitamins (thiamine, cyanocobalamin, and folate) which were within the normal range (51.5 ng/mL, 331 pg/mL, and 5.4 ng/mL, respectively). As we could not perform these laboratory tests in our hospital, these results were obtained a few days later. We considered a diagnosis of MBD and administered thiamine at a dose of 500 mg/8 h following the MRI and comprehensive vitamin testing (21 h after admission). Six hours after the thiamine injection (i.e., 27 h after admission), the patient gained consciousness and showed a GCS of nine. Approximately 40 h after admission, the patient recovered completely and returned home without any neurological symptoms. At a follow-up visit conducted 50 days after discharge, the lesion observed on the brain MRI had resolved completely, and she had no other symptoms. The patient informed us that she was a social drinker, drank alcohol once or twice a week, and consumed 40 mg of alcohol at a time. Moreover, she started drinking 5 months before hospital admission when she started going to the university. Based on a thorough examination and detailed medical history, we concluded that the ALCC was caused by binge alcohol consumption and was within the spectrum of MBD.

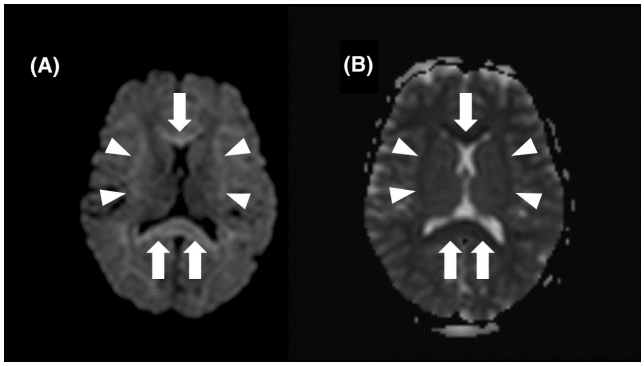

FIGURE 1 Brain MRI from the patient showing an acquired lesion of the corpus callosum (arrow) and hemispheric white matter (arrowhead), hyperintense on DWI (A), and hypointense on the ADC map (B). ADC, apparent diffusion coefficient; DWI, diffusion-weighted imaging; MRI, magnetic resonance imaging.

DISCUSSION

This was a perplexing case of acute alcohol intoxication presenting with ALCC, suggestive of MBD in a patient without any underlying chronic conditions. Although the major risk factor for MBD is chronic alcohol consumption,[2–4] MBD due to acute alcohol intoxication has not been reported before.

The diagnosis of MBD is based on MRI images in combination with medical history and clinical findings. The typical MRI findings of MBD include low T1, high T2 and high FLAIR signal intensity within the body of the corpus callosum which represent demyelination and myeline damages and which resolve entirely in cases exhibiting a benign clinical course. Associated lesions have been documented in the cerebral white matter, predominantly along the frontparietal robe. In our case, acquired lesions were observed in the specific area on DWI and ADC sequences, suggestive of cytotoxic edema in the early phase of MBD, as reported in several cases. The results of a thorough examination

(including toxicological screening and blood, CSF, and EEG tests), medical history, and the clinical course excluded other possible etiologies that present with similar MRI findings such as seizures, drugs (e.g., antiepileptic drugs), infectious diseases, hypoglycemia, hypernatremia, high-altitude edema, Charcot–Marie–Tooth disease, and systemic lupus erythematosus.[1] Eventually, the diagnosis was made based on the entire clinical course and complete recovery of neurological symptoms and MRI findings with thiamine treatment alone.

Several studies have investigated MBD pathophysiology. Some case reports of MBD have been reported in patients with severe osmotic disorder who were not alcoholics, suggesting that abrupt changes in serum osmolality lead to demyelination of the corpus callosum.[5–7] Another report suggested that cytotoxic edema of the corpus callosum caused by various etiologies, such as increased neurotransmitters including glutamate and cellular energy loss, is an important mechanism for this condition.[1,8] This pathophysiology is associated with extremely high BAC, which causes a marked change in blood osmolality and has direct/indirect cytotoxic effects on the brain.[9] Therefore, it is possible that the ALCC presented in this case is within the spectrum of MBD caused by binge consumption of alcohol.

In patients with severe alcohol intoxication, it is difficult to establish when further examination and treatment are required. Heinrich et al. concluded that MBD causes minor long-term disability in 61% of the patients and death in 8% of the patients.[3] Although it is not clear what determines the clinical course, early diagnosis of this disease is critical because some of these patients may benefit from early treatment.[2,4] However, it is not practical to conduct MRI in all unconscious patients with acute alcohol intoxication. Additionally, the routine use of intravenous fluid, dextrose, and thiamine in patients with acute alcohol intoxication is controversial.[10] Our patient had a severely disturbed consciousness and an extra-callosal lesion on the brain MRI which was previously reported to be a bad prognostic factor for MBD.[4] Consequently, the patient's condition could have deteriorated if left untreated. Therefore, thiamine was administered at a dose of 500 mg/8 h, and the patient recovered quickly. It is unclear whether the patient's recovery was due to thiamine administration or a natural course because her BAC was high enough to cause a coma. However, in such cases, routine administration of thiamine might be a better treatment strategy than waiting for a complete examination. Administering thiamine prioritizes the prevention of patient morbidity because of untreated MBD over the rare side effects in a cost-effective manner (given its low cost).

CONCLUSION

In summary, we reported a case of acute alcohol intoxication with ALCC caused by binge consumption of alcohol in an otherwise healthy patient. This case should serve as a caution for clinicians and the public against underestimating the potential harm caused by acute alcohol intoxication.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

ETHICS STATEMENT

Approval of the research protocol: N/A.
Informed consent: The patient and her parents provided a written informed consent for the publication of this case report.
Registry and the registration no. of the study/trial: N/A.
Animal studies: N/A.

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