

Gastrointestinal Beriberi Mimicking a Surgical Emergency in a Well-Nourished Patient: A Case Report

T.N. Diem Vu, MD; Michelle S. Junker, MD; Alexander Kurjatko, MD; Robert C. Albright, Jr, DO; Stephanie F. Heller, MD; and Mariela Rivera, MD

Abstract

Beriberi is a well-documented disease caused by thiamine deficiency. The diagnosis of gastrointestinal beriberi in the clinical setting is uncommon, especially in nonalcoholic patients. Failure to recognize beriberi can result in devastating acute multisystem organ failure; however, timely treatment can result in rapid improvement in a patient's clinical status. We present the case of an 81-year-old nonalcoholic man presenting with abdominal pain, lethargy, and hypotension. The patient was admitted to the intensive care unit and intubated for hemodynamic instability and declining mental status. Further investigations revealed profound lactic acidosis and cardiac hypokinesia. The patient's course changed rapidly after intravenous thiamine administration, and within hours he was weaned off vasopressors. He was extubated, discharged from the intensive care unit, and discharged to home quickly thereafter. To our knowledge, this report is the first description of gastrointestinal beriberi mimicking a surgical emergency in an otherwise well-nourished patient with no history of alcoholism. The rapid improvement the patient experienced with administration of thiamine underscores the importance of considering gastrointestinal beriberi and thiamine deficiency in all moribund patients with unexplained abdominal symptoms, cardiogenic shock, and lactic acidosis.

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From the Department of Surgery (T.N.D.V., M.S.J.), Division of Nephrology and Hypertension (R.C.A.), and Division of Trauma, Critical Care, and General Surgery (S.F.H., M.R.), Mayo Clinic, Rochester, MN; and Adams Memorial Hospital, Decatur, IN (A.K.).

Beriberi is a well-documented disease caused by thiamine deficiency, historically divided into “wet” and “dry” classifications. One 2004 case report described a third form of beriberi characterized by abdominal pain and lactic acidosis in 2 alcoholic patients.¹ The diagnosis of beriberi in the clinical setting is uncommon, especially in nonalcoholic patients. Failure to recognize beriberi can result in devastating acute multisystem organ failure; however, timely treatment can result in rapid improvement in a patient's clinical status. We report the case of a nonalcoholic man with gastrointestinal beriberi who presented with abdominal pain, lethargy, and hemodynamic instability.

REPORT OF CASE

The patient was an 81-year-old man with a medical history notable for rheumatoid arthritis treated with prednisone. He had no history of alcohol abuse. One year previously,

he had presented with a 2-week history of abdominal pain, nausea, and weakness. He underwent exploratory laparotomy with multiple trips to the operating room for lysis of adhesions and eventually proximal ileal resection. During that hospital course, it was noted that the patient had recently been treated for atrial fibrillation with a rapid ventricular rate, but echocardiography revealed no evidence of thrombosis.

This case report focuses on his next hospitalization during the spring of 2018, which started when he presented to the emergency department with severe lethargy and was intubated for airway protection. He was hypotensive, requiring support with norepinephrine. Physical examination findings were notable for a soft, but distended, abdomen.

Laboratory work-up revealed a white blood cell count of $10.8 \times 10^9/L$ (reference range, $3.5-10.5 \times 10^9/L$) with a neutrophil shift. His creatinine level was 2.1 mg/dL

(reference range, 0.8-1.3 mg/dL) (to convert creatinine values to $\mu\text{mol/L}$, multiply by 88.4), which was substantially elevated above his baseline level. Arterial blood gas studies revealed metabolic acidosis with a pH of 7.29, PCO_2 level of 39 mm Hg, bicarbonate concentration of 18 mmol/L, and base deficit of 7 mmol/L. Central venous oxygen saturation was 49%. His liver function profile was within normal limits. The serum albumin level was mildly decreased at 2.3 g/dL (reference range, 3.5-5.0 g/dL) (to convert albumin values to g/L, multiply by 10). His lactate concentration was 6.8 mmol/L (reference range, 0.6-2.3 mmol/L). Computed tomography of the abdomen and pelvis yielded unremarkable results. Echocardiography revealed no evidence of thrombus and a left ventricular ejection fraction of 45%. Ammonia levels, acetaminophen values, and toxin panel results were unremarkable.

Because of the patient's worsening clinical status and lactic acidosis, he underwent exploratory laparotomy for presumed mesenteric ischemia. The results were unrevealing. Postoperatively, he was admitted to the surgical intensive care unit for mechanical ventilation and hemodynamic support. His lactic acidosis continued to worsen, with the lactate level reaching a peak of 14.4 mmol/L. The patient's pH value decreased to a nadir of 7.21, his bicarbonate level decreased to 13 mmol/L, and the base deficit increased to 14 mmol/L. Central venous oxygen saturation was 51%. The patient became progressively more hemodynamically unstable. Point-of-care echocardiography revealed global hypokinesis and ventricular dilatation, concerning for a cardiac etiology of his clinical deterioration. Thus, epinephrine was added to the regimen of norepinephrine and vasopressin. Withdrawal of support was also being considered at this time.

After approximately 20 hours after the exploratory laparotomy, a nephrology consultation was requested for possible dialysis in the setting of worsening acidosis. Thiamine deficiency was proposed as a cause given the unusual clinical picture. The patient was treated empirically with intravenous thiamine delivered at 200 mg daily for 5 days. Whole-blood thiamine concentration measurements were not contemporaneously available. Within

2 hours of the first thiamine administration, the patient was weaned off epinephrine. Within 9 hours, vasopressin was discontinued, and within 16 hours, norepinephrine was stopped as well. Seven hours after thiamine administration, his lactate concentration had decreased to 4 mmol/L, and levels returned to normal the following morning. Acid-base balance had improved to a pH of 7.41 by 4 hours after thiamine administration, with the base deficit decreasing to 3 mmol/L. His bicarbonate concentration improved to 21 mmol/L.

The patient was extubated on his third hospital day and was transferred from the intensive care unit on his fourth hospital day. He was discharged to home on his eighth hospital day after completing a 5-day course of intravenous thiamine.

DISCUSSION

Vitamin B₁, or thiamine, is an essential nutrient first discovered in 1897 as a water-soluble vitamin. It is synthesized by bacteria, plants, and fungi from thiazole and pyrimidine. In humans, thiamine is absorbed by active transport in the jejunum and ileum.² When transported in the bloodstream, thiamine is protein-bound mainly to albumin, although a binding protein specific to thiamine also exists.³ Thiamine is then taken up by cells through both active and passive diffusion. Within the cell, thiamine is phosphorylated by thiamine pyrophosphokinase into thiamine pyrophosphate, or thiamine diphosphate. This is the most metabolically active form of thiamine, serving as a cofactor for transketolase, branched-chain α -ketoacid dehydrogenase, and pyruvate dehydrogenase.⁴ Therefore, thiamine deficiency impairs the activity of these enzymes, diminishing the flux of the tricarboxylic acid cycle and allowing the substrate pyruvate to accumulate. Pyruvate also accumulates due to the decreased activity of pyruvate dehydrogenase, which would normally convert pyruvate into acetal coenzyme A. The pyruvate built up by the impairment of these pathways is then converted to lactate through the anaerobic activity of lactate dehydrogenase. In summary, the clinical findings in thiamine deficiency are due to the accumulation of pyruvate and thus lactate caused by the dysfunction of important enzymes that require thiamine for function.

The daily requirement of thiamine is dependent on an individual's carbohydrate intake and usually amounts to 1 to 2 mg per day. To process 1000 kcal, 0.5 mg of thiamine is required. Body stores of thiamine are usually between 30 and 50 mg and can be depleted within a span of 4 to 6 weeks.⁵ The National Institutes of Health guidelines on the measurement of thiamine status state that plasma levels of thiamine are not reliable indicators of thiamine status.⁶ Instead, erythrocyte transketolase activity and erythrocyte thiamine diphosphate concentration are the criterion standards for assessing intracellular concentration of thiamine.⁷

The constellation of symptoms caused by thiamine deficiency has historically been referred to as beriberi. The research of Christiaan Eijkman had suggested that beriberi was caused by the diet, noting a correlation between symptoms and a diet of white rice as opposed to brown (unpolished) rice.⁸ It has been well documented that a history of alcohol abuse is associated with thiamine deficiency, due to both poor availability in the diet and interference in the absorption of thiamine hydrochloride.⁹ States of malnutrition such as anorexia nervosa, systemic malignancy, HIV/AIDS, and prolonged total parenteral nutrition use without supplementation are also associated with thiamine deficiency. Bariatric surgery has arisen as a contemporary cause of thiamine deficiency due to absorptive changes.¹⁰ Experimental models have also documented deficient thiamine transport in diabetes mellitus.¹¹ Because thiamine is excreted renally, excessive losses of thiamine through long-term dialysis or chronic diuresis have been documented as causes of thiamine deficiency. More obscure causes of thiamine deficiency include the consumption of certain fish and shellfish that harbor thiaminases in their flesh,¹² which would result in normal, but functionally inactive, thiamine levels. Overconsumption of polyphenols found in teas, chocolates, nuts, fruits, and vegetables has also been documented to have similar effects.¹³

There are 2 common forms of beriberi, termed *dry beriberi* and *wet beriberi*. Dry beriberi is characterized by neurologic symptoms including ataxia, numbness, loss of deep tendon reflexes, paralysis, and confusion.

Wet beriberi is characterized by cardiovascular effects, resulting in peripheral edema, dyspnea, and heart failure. A third form of beriberi has been described with gastrointestinal symptoms. In the 1940s, a study of induced thiamine deficiency demonstrated the onset of gastrointestinal symptoms such as nausea, vomiting, and abdominal pain.¹⁴ In case reports, varying levels of lactic acidosis have been characteristic of the disease process.^{15,16}

This case illustrates that, although rare, thiamine deficiency can present in patients without a history of alcoholism or malnutrition. To our knowledge, this report is the first description of gastrointestinal beriberi mimicking a surgical emergency in an otherwise well-nourished patient with no history of alcoholism. A prior report of 2 patients featured similar clinical narratives, although the 2 patients described were inadequately nourished and 1 had a history of alcoholism.¹ We hypothesize that our patient's previous small bowel resection could have predisposed him to this condition. The rapid improvement the patient experienced with therapeutic administration of thiamine underscores the importance of recognizing this uncommon disease process. Failure to recognize gastrointestinal beriberi as a cause of severe lactic acidosis could result in preventable morbidity and mortality. Therefore, thiamine deficiency should be considered in the differential diagnosis in all moribund patients with unexplained abdominal symptoms, cardiogenic shock, and lactic acidosis.

CONCLUSION

As evidenced by the rapid improvement experienced by the patient in this case, gastrointestinal beriberi and thiamine deficiency should be considered in critically ill patients with unexplained abdominal pain, cardiogenic shock, and lactic acidosis.

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Correspondence: Address to T.N. Diem Vu, MD, Department of Surgery, Mayo Clinic, 200 First St SW, Rochester, MN 55905 (Vu.trangl@mayo.edu).

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