Case Reports in Neurology

Case Rep Neurol 2019;11:124–131

DOI: 10.1159/000499601
Published online: April 18, 2019

Case Report

Gastrointestinal Beriberi and Wernicke's Encephalopathy Triggered by One Session of Heavy Drinking

Elysia Tjong[a] Yen-Yi Peng[b, c]

[a]A.T. Still University School of Osteopathic Medicine in Arizona, Mesa, AZ, USA; [b]Renown Institute for Neuroscience, Renown Health, Reno, NV, USA; [c]Department of Neurology, University of Nevada, Reno, NV, USA

Keywords
Gastrointestinal beriberi · Wernicke's encephalopathy · Thiamine

Abstract
An otherwise healthy 30-year-old male acquired gastrointestinal beriberi and subsequent Wernicke's encephalopathy after 1 session of heavy drinking. Nausea, vomiting, and anorexia relentlessly progressed. The patient developed external ophthalmoplegia after 2 months. Intravenous 1,000 mg thiamine reversed both neurologic and gastrointestinal symptoms within hours. It is hard to diagnose gastrointestinal beriberi since the symptoms are nonspecific. The patient underwent 11 emergency room visits, 3 hospital admissions, and laparoscopic cystectomy within 2 months, but the gastrointestinal symptoms continued to progress. Two months after the onset of gastrointestinal symptoms, external ophthalmoplegia appeared, and, therefore, intravenous thiamine was given. The simultaneous resolution of the debilitating gastrointestinal symptoms and external ophthalmoplegia was unique. Thiamine deficiency remains underdiagnosed and should be considered in patients who develop unexplained gastroparesis or autonomic nervous failure of the digestive system, even in the nonalcoholic population.

KARGER

Yen-Yi Peng
Renown Health
Department of Neurology, University of Nevada
75 Pringle Way, Reno, NV 89502 (USA)
E-Mail yypeng@taiwaneden.com

Introduction

Thiamine derivative – thiamine pyrophosphate – is an essential coenzyme in the mitochondrial functions. Thiamine deficiency can lead to Wernicke-Korsakoff syndrome, gastrointestinal beriberi, neuropathy, and cardiomyopathy. The characteristic symptoms of Wernicke's encephalopathy (WE) consist of oculomotor abnormalities, ataxia, and confusion (the classic triad). These 3 symptoms do not always coexist in most cases. Caine's criteria of WE require 2 of the following 4 signs: (A) dietary deficiencies, (B) oculomotor abnormalities, (C) cerebellar dysfunction, and (D) either an altered mental state or mild memory impairment; these criteria improve the diagnostic sensitivity and are clinically applicable [1]. Failure to diagnose WE and promptly institute intravenous thiamine could result in Korsakoff's psychosis (KP) or death. KP is characterized by amnesia, confusion, and confabulation [2].

WE symptoms begin with gastrointestinal beriberi. Patients with thiamine deficiency start experiencing nausea, vomiting, and anorexia and are well documented in the literature. Cases of thiamine-responsive gastrointestinal symptoms (gastrointestinal beriberi) have also been reported [3–7].

It is hard to diagnose gastrointestinal beriberi since the symptoms are nonspecific and no specific laboratory tests are available. Thiamine deficiency-induced gastroparesis compromises the absorption of nutrition and further worsens the thiamine deficiency. Gastrointestinal beriberi can be a vicious cycle and can potentially lead to WE. It is critical to consider thiamine deficiency in patients with acute gastroparesis [5].

Case Report

A 30-year-old healthy male had 1 session of heavy drinking (3 cans of beer, 2 shots of Tequila, 1 shot of Vodka, and 2 shots of Whiskey) followed by 1 day of hangover. The patient developed nausea and foreign body sensation in his throat 3 days later and started to seek medical help. Within 2 months, 11 emergency room (ER) visits, 3 admissions, and laparoscopic cholecystectomy took place in this institution. Despite that, nausea, vomiting, anorexia, and weight loss relentlessly progressed. In the beginning, the patient sought medical help for intractable food regurgitation. Toward the end of the first month, the patient could no longer tolerate solid food. He was only able to consume shakes (spinaches, berries, but no dairy). Appetite was impaired in the first month. By the start of the second month, he could no longer tolerate shakes. For the first 2 weeks in the second month, the patient was only able to consume water. Afterward, the patient could not even tolerate water and lost his appetite completely. Towards the end of the 2 months, the patient had more than 10 episodes of bilious vomiting a day. The cholecystectomy did not relieve his nausea, vomiting, and anorexia. The patient lost around 23 kg (118 kg to 95 kg) in weight within 2 months (Fig. 1).

The patient drank regularly 3 years prior to this event (around 4 cans of beer each day, with an average of 4 days per week) but started to drink alcohol only occasionally since then. Because the gastrointestinal symptoms were not specific, during the 2 months after the onset of symptoms, the patient was treated for probable esophageal stricture, esophageal tear, acid reflux, *Helicobacter pylori* infection, pancreatitis, gastroparesis, cholecystitis, cardiac disease, asthma, and/or anxiety. Among these, in the third admission in this institution, pancreatitis or cholecystitis was regarded as a possible diagnosis.

A gastric emptying test confirmed gastroparesis (Fig. 2). Esophagram was negative. Magnetic resonance cholangiopancreatography showed normal pancreas and biliary structure.

Two upper gastrointestinal endoscopies were also performed and were unremarkable. The gastric biopsy was negative for *H. pylori* infection. Two abdominal/pelvic computed tomography (CT) scans were also unremarkable. Hepatobiliary iminodiacetic acid scan showed a gallbladder ejection fraction of 12% (normal range ≥38%), which revealed gallbladder dyskinesia (Fig. 3). The laparoscopic cholecystectomy was performed around the seventh week. The second abdominal CT was done 3 days after cystectomy and revealed residual barium in the colon left by the esophagram done 1 month ago (Fig. 4). The residual barium remained in the colon of the abdomen according to X-ray 37 days after the esophagram and denoted decreased bowel motility. Two brain magnetic resonance imaging (MRI) scans, cardiac echogram, and abdominal echogram were unremarkable.

Complete blood cell count remained unremarkable. Blood chemistry showed normal alkaline phosphatase during this period of 2 months. Both SGOT and SGPT were not remarkable at the beginning (SGOT was 22 U/L with a normal range of 12–45 U/L; SGPT was 51 U/L with a normal range of 2–50 U/L). The initial blood chemistry did not support the diagnosis of alcoholic hepatitis. Towards the second month, SGOT rose to 52 U/L (approximately 2 times the amount of baseline) and SGPT rose to 161 U/L (approximately 3 times the amount of baseline). Lipase had increased since the second month with the highest level at 573 U/L (normal range 11–82 U/L). Amylase was mildly elevated at 148 U/L (normal range 20–103 U/L). Serum vitamin B_1 (thiamine), lactic acid, and blood pH were never ordered. The patient drank a small amount of beer a couple of times only during these 2 months before he completely lost his appetite. The gallbladder pathological report showed inflammation.

Ten days after laparoscopic cholecystectomy and around 8 weeks after onset, the neurology team was consulted for a new onset of diplopia. During the physical examination, the patient was awake and oriented. The patient was not in acute distress. He denied abdominal pain. Anorexia and bilious vomiting remained intractable. External ophthalmoplegia and diplopia were present (online suppl. Video 1; for all online suppl. material, see www.karger.com/doi/10.1159/000499596). There was no confusion or ataxia. The second MRI of the brain and an ophthalmologic consultation were completed just 1 day prior. No eye disorders or intracranial structural lesions were responsible for this ophthalmoplegia. The diplopia was not fluctuating. One dose of 500 mg intravenous thiamine was promptly administered under the physician's supervision, but the ophthalmoplegia remained [6, 8]. A second dose of intravenous 500 mg thiamine was, therefore, given, without immediate response either. After around 6 h of sleep, the patient woke up and noticed that the diplopia had subsided. Intractable nausea and vomiting also resolved on the same day, and his appetite came back. The clinical manifestations met Caine's criteria of WE because of oculomotor problems and dietary deficiencies. The diagnosis of WE was made 2 months after the patient's single session of heavy drinking [1]. Daily intravenous 1,000 mg thiamine was continued [2]. The patient recovered completely and was discharged 3 days after the intravenous thiamine supplementation (online suppl. Video 2). The patient remained healthy 9 months after this event, and he later started a healthy diet.

Discussion

This case represents a challenging case of gastrointestinal beriberi, which led to WE within 2 months. The gastrointestinal beriberi was triggered by 1 session of heavy drinking. The above arguments are supported by the following: (A) the dramatic response of neurologic and gastrointestinal symptoms to intravenous 1,000 mg thiamine (online suppl. Video 1 and

2). (B) Based on the medical records in this institution (11 ER visits and 3 hospital admissions), the gastrointestinal symptoms progressively worsened within this 2-month period. Anorexia progressed to complete loss of appetite. Nausea progressed to intractable bilious vomiting. Weight loss was unrelenting (Fig. 1). This information was corroborated by the family's separate report. Within these 2 months, the patient only drank a couple of beers before he lost his appetite completely in the second month. (C) The cholecystectomy did not relieve the gastrointestinal symptoms. The gastrointestinal immobility was pervasive. The clinical manifestations and imaging tests denoted severe gastroparesis and delayed bowel motility on top of the gallbladder dyskinesia. (D) Even though the patient was a nonalcoholic before this event, he did admit that he had an unhealthy diet and he reported eating at fast-food restaurants around 4 times a week. Greasy food was his favorite. He typically ate 2 meals a day and he was overweight (BMI was 40.7). Absorption of thiamine is reduced by malnutrition [8]. The total body stores of thiamine for healthy subjects are small, being approximately 30–50 mg (daily requirements 1–2 mg, increased by alcohol metabolism), and will be depleted within a few weeks by poor intake even if absorption from the gastrointestinal tract remains normal [6, 8]. Therefore, it is reasonable to assume that the patient's reserve of thiamine was already compromised before this event.

Nonalcoholic WE could develop in patients with a thiamine-deficient diet. Therefore, our argument that 1 binge drinking induced the thiamine deficiency remains plausible. The gastrointestinal beriberi made the absorption of thiamine more difficult and spiraled into the cerebral beriberi/WE. Genetic predisposition to develop WE is also a likely possibility [8].

WE is a neurological urgency. Failure to diagnose and promptly institute intravenous thiamine would result in KP or death. KP is characterized by amnesia, confusion, and confabulation [2]. WE can be characterized by the classic triad of oculomotor abnormalities, ataxia, and confusion. However, these symptoms do not always coexist. In this case, the patient only had oculomotor abnormalities. The pathological features in WE and KP are the same. Therefore, WE and KP can be grouped into the Wernicke-Korsakoff syndrome. Thiamine deficiency induces gastrointestinal symptoms, such as nausea, vomiting, and anorexia. It is reasonable to coin these early symptoms of thiamine deficiency as gastrointestinal beriberi [3, 4, 7]. Furthermore, thiamine deficiency could affect the peripheral nerve (dry beriberi) and cardiac system (wet beriberi) [9].

Our team did not order any serum thiamine level of this patient during these 2 months. Intravenous thiamine was given promptly with the high clinical suspicion of WE. Thiamine blood levels do not always reflect brain levels of thiamine diphosphate, nor do they predict the development of WE in an individual because of compromised thiamine transport into the neurons in patients with thiamine deficiency [6]. Unspecific gastrointestinal symptoms and lack of reliable objective tests pose a diagnostic challenge in the early stages of thiamine deficiency.

Thiamine is critical for normal mitochondrial function [6]. Energy-intensive neurons are susceptible to thiamine deficiency. In this case, the authors hypothesize that thiamine deficiency can result in gastroparesis, delayed bowel motility, and gallbladder dyskinesia. More specifically, thiamine deficiency can result in autonomic nervous failure of the gastrointestinal system. The authors also hypothesized that the inflammatory gallbladder on the pathological report could be reactive to the gallbladder dyskinesia. Apparently, the gallbladder inflammation was not the sole cause of the gastrointestinal symptoms since the cholecystectomy did not relieve the patient's symptoms at all.

The appetite and hunger sensations depend on the interaction between the nervous system of the gastrointestinal system and the brain [10]. The loss of hunger sensation in the second month of this case suggests the breakdown of the gut-brain connection. The appetite and hunger sensations came back hours after intravenous thiamine supplementation in this case, which suggests the critical role of thiamine in maintaining human hunger perception.

As documented in the literature and as also demonstrated in this case, gastrointestinal beriberi usually precedes WE [2]. The threshold of thiamine deficiency for gastrointestinal beriberi is much lower than that of WE. Furthermore, considering the evolution of gastrointestinal symptoms in this case, the authors hypothesized that the gastrointestinal symptoms of the hangover after drinking might be secondary to transient thiamine deficiency. Most people could recover from this transient thiamine deficiency within hours and days spontaneously. This case is an exception probably because of an underlying low reserve of thiamine due to the patient's poor diet and occasional drinking.

Failure to diagnose thiamine deficiency and promptly institute intravenous thiamine could result in high medical costs, WE, KP, or death. Thiamine deficiency remains underdiagnosed and should be considered in patients who develop unexplained gastroparesis or autonomic nervous failure of the digestive system, even in the nonalcoholic population [5, 7, 11].

Acknowledgement

Austine Peng assisted in proofreading the manuscript.

Statement of Ethics

Our institution does not require ethical approval for reporting individual cases. Written informed consent was obtained from the patient for his anonymized information to be published in this article.

Disclosure Statement

All authors declare no conflicts of interest.

Funding Sources

There were no funding sources.

Author Contributions

E.T. prepared the manuscript.
Y.-Y.P. was the neurologist for this patient.

References

1 Caine D, Halliday GM, Kril JJ, Harper CG. Operational criteria for the classification of chronic alcoholics: identification of Wernicke's encephalopathy. J Neurol Neurosurg Psychiatry. 1997 Jan;62(1):51–60.
2 Thomson AD, Marshall EJ, Bell D. Time to act on the inadequate management of Wernicke's encephalopathy in the UK. Alcohol Alcohol. 2013 Jan-Feb;48(1):4–8.
3 Duca J, Lum CJ, Lo AM. Elevated Lactate Secondary to Gastrointestinal Beriberi. J Gen Intern Med. 2016 Jan;31(1):133–6.
4 Prakash S. Gastrointestinal beriberi: a forme fruste of Wernicke's encephalopathy? BMJ Case Rep. 2018;2018:bcr-2018-224841.
5 Shah IA, Asimi RP, Kawoos Y, Wani M, Saleem T, Baba WN. Nonalcoholic Wernicke's Encephalopathy: A Retrospective Study from a Tertiary Care Center in Northern India. J Neurosci Rural Pract. 2017 Jul-Sep;8(3):401–6.
6 Thomson AD, Marshall EJ. The natural history and pathophysiology of Wernicke's Encephalopathy and Korsakoff's Psychosis. Alcohol Alcohol. 2006 Mar-Apr;41(2):151–8.
7 Donnino M. Gastrointestinal beriberi: a previously unrecognized syndrome. Ann Intern Med. 2004 Dec;141(11):898–9.
8 Thomson AD, Guerrini I, Marshall EJ. The evolution and treatment of Korsakoff's syndrome: out of sight, out of mind? Neuropsychol Rev. 2012 Jun;22(2):81–92.
9 Lanska DJ. Chapter 30: historical aspects of the major neurological vitamin deficiency disorders: the water-soluble B vitamins. Handb Clin Neurol. 2010;95:445–76.
10 Ahima RS, Antwi DA. Brain regulation of appetite and satiety. Endocrinol Metab Clin North Am. 2008 Dec;37(4):811–23.
11 Pilling S, Yesufu-Udechuku A, Taylor C, Drummond C; Guideline Development Group. Diagnosis, assessment, and management of harmful drinking and alcohol dependence: summary of NICE guidance. BMJ. 2011 Feb;342:d700.

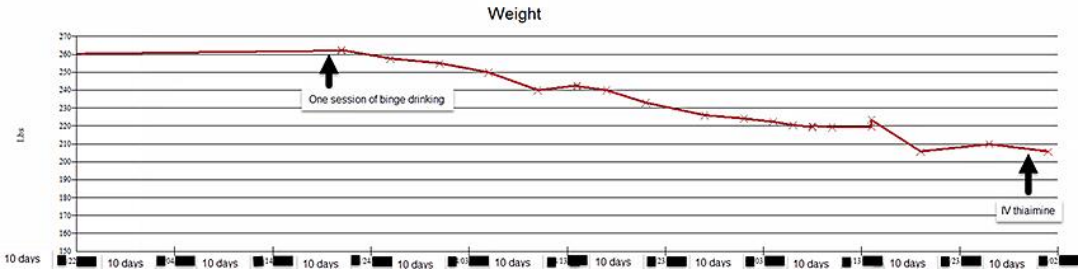

Fig. 1. The weight loss in the following 2 months after 1 session of heavy drinking: each cross-mark represents 1 ER visit or 1 hospital admission.

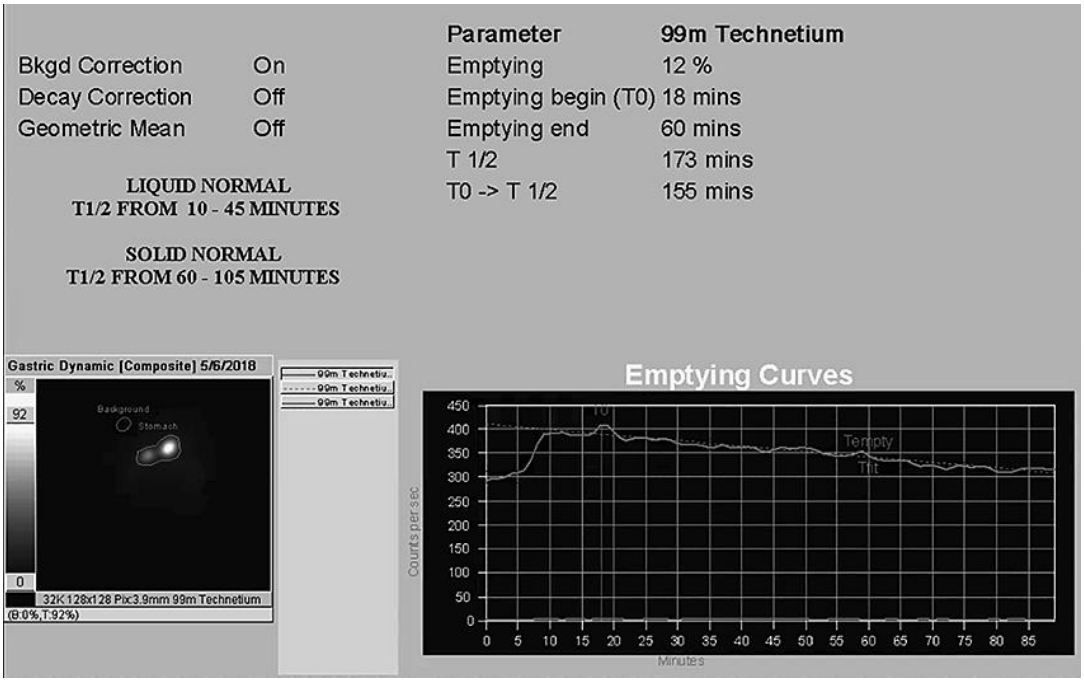

Fig. 2. Gastric emptying scintigraphy: there was no evidence of gastroesophageal reflux during 90 min of continuous observation. Gastric half-emptying time was 173 min. Delayed gastric emptying suggested gastroparesis.

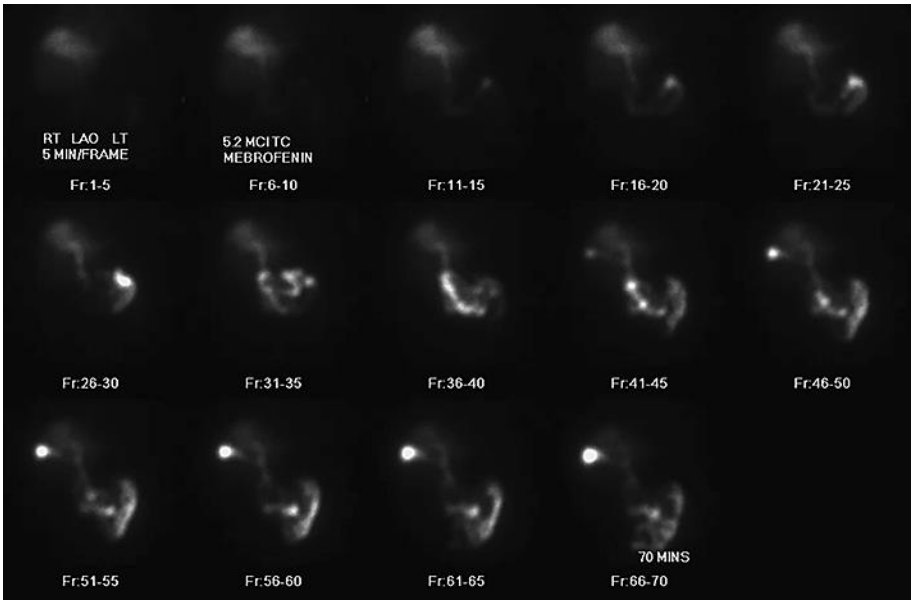

Fig. 3. Hepatobiliary iminodiacetic acid scan: there was a prompt uptake of the tracer in the hepatic parenchyma. The gallbladder filled with activity within 60 min and activity was excreted normally into the small bowel. The gallbladder ejection fraction was 12% (normal gallbladder ejection fraction is 38% or greater). The patient reported mild pain with cholecystokinin administration.

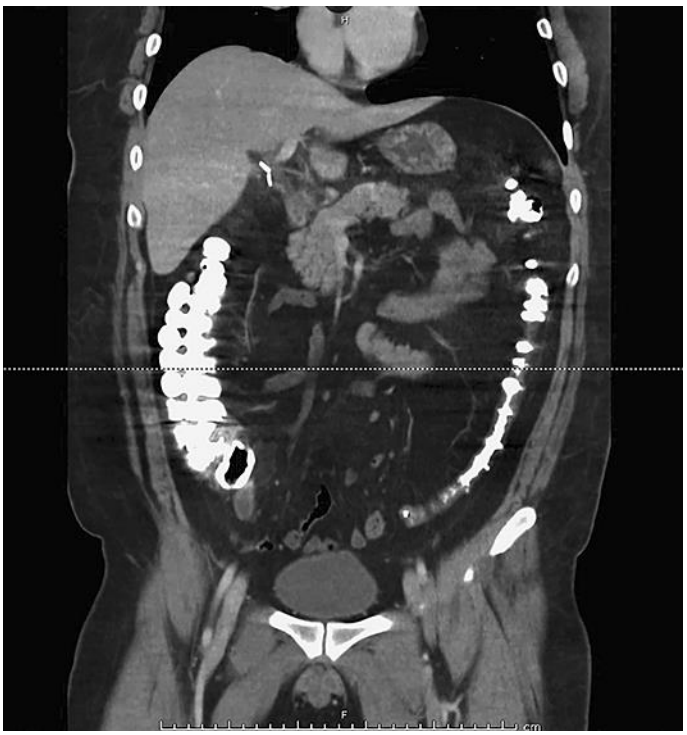

Fig. 4. The second abdominal/pelvic computed tomography scan was done 3 days after the cholecystectomy: interval cholecystectomy with expected postsurgical change, barium within the colon apparently from the esophagram 1 month ago, suggesting decreased/delayed bowel motility with no evidence of bowel dilatation or obstruction.