A Case of Pulmonary Hypertension in a 67-Year-Old Woman with Thiamine Deficiency Following Partial Gastrectomy and Exacerbated by Diuretics

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Patient: Female, 67-year-old
Final Diagnosis: Pulmonary hypertension caused by thiamine deficiency
Symptoms: Leg edema
Medication: —
Clinical Procedure: —
Specialty: Cardiology

Objective: Unusual clinical course
Background: Thiamine deficiency often occurs in patients with alcohol abuse and unbalanced diets. However, gastric surgery and/or use of diuretics can also cause this situation. Importantly, thiamine deficiency can cause pulmonary hypertension, which is completely reversible. This report is of a case of a 67-year-old woman who presented with pulmonary hypertension and thiamine deficiency following partial gastrectomy and exacerbated by diuretics. We found that her blood thiamine concentration was extremely low. We diagnosed her as having thiamine deficiency caused by gastrectomy and administered diuretics. After intravenous thiamine administration, her symptoms showed immediate improvement, associated with the normalization of the pulmonary hypertension. After detailed analysis of the cause of her pulmonary hypertension, including Swan-Ganz catheterization and echocardiography, we concluded that her pulmonary hypertension was caused by thiamine deficiency following partial gastrectomy and exacerbated by diuretics.

Conclusions: This case highlights the importance of recognizing that thiamine deficiency can be a cause of pulmonary hypertension, and that thiamine deficiency can be associated with gastrectomy and the use of diuretics.

Keywords: 2-(1-hydroxyethyl)thiamine pyrophosphate • Diuretics • Hypertension, Pulmonary

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Background

The clinical classification of pulmonary hypertension is categorized into 5 groups according to clinical presentation, pathological findings, and hemodynamic characteristics. Metabolic disorders are classified into group 5 (unclear and/or multifactorial mechanisms), but the classification does not include thiamine deficiency [1]. Thiamine is a water-soluble vitamin essential for carbohydrate metabolism. Thiamine deficiency often occurs in patients with alcohol abuse and unbalanced diets. Moreover, it has been shown that thiamine deficiency can also result from gastrectomy [2] and adverse effects of diuretics [3]. Importantly, in rare situations, it has been shown that pulmonary hypertension can result from thiamine deficiency [4].

This report is of a case of a 67-year-old woman who presented with pulmonary hypertension and thiamine deficiency following partial gastrectomy and exacerbated by diuretics.

Case Report

A 67-year-old woman who had a history of partial gastrectomy and jejunum interposition because of non-Hodgkin lymphoma at the age of 36 years and a sigmoid colectomy because of colon cancer at the age of 58 years exhibited leg edema for several months. To reduce her edema, indapamide had been administered at another hospital previous to the current presentation. However, her edema gradually worsened despite use of diuretics (indapamide), and she also developed dyspnea on exertion. Finally, she was referred to our hospital for further medical care for this condition.

She denied drinking alcohol and having an unbalanced diet. Physical examination revealed jugular vein distention, increased second heart sound, and bilateral legs pitting edema. Her neurological examination result was normal. Her blood pressure was 97/64 mmHg, body temperature was 35.4°C, and pulse was 89 beats/min. Her respiration rate was 16 breaths/min with oxygen saturation of 95% in room air.

Chest radiography revealed cardiomegaly and congested lungs (Figure 1A). Twelve-lead electrocardiography (ECG) revealed a sinus rhythm and RSR’ pattern in V1 (Figure 1B). Echocardiography revealed a right ventricular dilatation (Figure 1C), and a D-shaped left ventricular cavity in later systole and throughout diastole. Maximal tricuspid valve regurgitation increased up to 3.4 m/s (estimated pressure gradient between the right ventricle and right atrium: 49 mmHg; Figure 1D). The left ventricular systolic function was preserved. Little pericardial effusion was observed. The blood test result was almost normal except for an elevated brain natriuretic peptide level of 483.8 pg/mL. The hemodynamic evaluation using Swan–Ganz catheterization revealed a pulmonary artery pressure of 48/17 (30) mmHg, pulmonary capillary wedge pressure of 11 mmHg, cardiac index of 2.47 L/min/m², and pulmonary vascular resistance of 5.2 Wood units.

On the basis of these data, we diagnosed her as having pulmonary hypertension and then changed her medication from indapamide to torsemide. However, her body weight gradually increased, and her dyspnea did not improve. Thus, we started intravenous infusion of dobutamine (3 μg/kg/min). After 8 days of hospitalization, we found that her thiamine concentration was decreased to 10 ng/ml (normal range: 24–66 ng/ml). At that time, we diagnosed her as having thiamine deficiency caused by gastrectomy and administered diuretics (Figure 2). Intravenous thiamine (100 mg/day) was prescribed. After starting thiamine therapy, the patient’s urine output was increased significantly, and her dyspnea also improved even after withdrawal of dobutamine and diuretics. On day 18, the hemodynamic evaluation using a Swan-Ganz catheter revealed a pulmonary pressure decrease to 27/8 (16) mmHg, pulmonary capillary wedge pressure of 7 mmHg, cardiac index increase to 2.94 L/min/m², pulmonary vascular resistance decrease to 2.6 Wood units. She was discharged on day 22 with a prescription for oral thiamine. She is currently attending our hospital without recurrence of pulmonary hypertension.

Discussion

The case presented here illustrates 2 important issues: 1) there are cases of reversible pulmonary hypertension due to thiamine deficiency, potentially caused by gastrectomy and/or use of diuretics, and 2) empirical treatment using thiamine merits consideration in cases of pulmonary hypertension of unknown origin.

Thiamine is an essential water-soluble vitamin for carbohydrate metabolism. Thiamine deficiency causes tricarboxylic acid (TCA) cycle impairment. Thiamine is found in yeast, organ meat, pork, legumes, beef, whole grains, and nuts [5]. Alcohol abuse and unbalanced diet are well-known causes of thiamine deficiency. Beriberi, which is the best-known type of thiamine deficiency, is characterized by high-output heart failure. In the present case, the cardiac output was not high and did not meet the diagnostic criteria of beriberi [6]. The causes of beriberi are long-term alcohol drinking (32.26%), digestive system disease and surgery (29.03%), imprisonment (9.68%), furosemide administration (6.45%), complex factors (16.13%), other causes (3.23%), and unknown (3.23%) [7]. In our case, the cause of thiamine deficiency was considered to be both gastrointestinal surgery and use of diuretics.

Thiamine is known to be absorbed through the thiamine transporter solute carrier family 19 member 2 (SCL19A2). SCL19A2 is expressed in the following order: liver, stomach, duodenum,
jejunum, colon, cecum, rectum, and ileum [8]. Thus, the stomach and jejunum have important roles for thiamine uptake. In patients who had undergone a gastrectomy, thiamine levels decrease even if they return to normal activity after surgery. The body store of thiamine is limited. If thiamine is not administered orally, the body’s thiamine supply may become depleted within a few weeks [2]. Our patient had a distal gastrectomy 31 years before. Thus, the effect of distal gastrectomy on thiamine deficiency may be limited. She received diuretics from the previous hospital. Diuretics are also known to promote vitamin excretion [9]. In this case, loop and thiazide diuretics may have promoted the thiamine deficiency via urinary loss.

So far, only 2 cases of beriberi in a patient with a history of gastrointestinal surgery exacerbated by the use of diuretics have been reported, and both patients improved immediately after thiamine supplementation [10,11]. However, there is a published case exhibiting thiamine-responsive pulmonary hypertension not associated with high-output heart failure, suggesting that high-output heart failure and pulmonary hypertension may be an independent factor, at least in our case and the previous case [12]. It is interesting to note that the clinical manifestations described above were quite similar those of our patient.
Conclusions

This case highlights the importance of recognizing that thiamine deficiency can be a cause of pulmonary hypertension, and that thiamine deficiency can be associated with gastrectomy and the use of diuretics.

Ethics Approval and Consent to Participate

The publication of this case report was approved by the Clinical Research Ethics Committees of Kanazawa University.

Declaration of Figures’ Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

References:

1. Nazzareno G, Marc H, Jean-Luc V, et al. 2015 ESC/ERS guidelines for the diagnosis and treatment of pulmonary hypertension. Rev Esp Cardiol (Engl Ed). 2016;69(2):17
2. Iwase K, Higaki J, Yoon HE, et al. Reduced thiamine (vitamin B1) levels following gastrectomy for gastric cancer. Gastric Cancer. 2002;5(2):77-82
3. Sica DA. Loop diuretic therapy, thiamine balance, and heart failure. Congest Heart Fail. 2007;13(4):244-47
4. Park JH, Lee JH, Jeong JO, et al. Thiamine deficiency as a rare cause of reversible severe pulmonary hypertension. Int J Cardiol. 2007;121(1):e1-3
5. Paolo MS, Robert MR. Vitamin and trace mineral deficiency and excess. In: Harrison’s Principles of Internal Medicine. 20th ed. J. Larry J, Anthony SF, Dennis L K, et al, Eds. McGraw-Hill, New York, 2018;2309-10
6. Wakabayashi A, Yui Y, Kawai C. A clinical study on thiamine deficiency. Jpn Circ J. 1979;43(11):995-99
7. Lei Y, Zheng MH, Huang W, Zhang J, Lu Y. Wet beriberi with multiple organ failure remarkably reversed by thiamine administration: A case report and literature review. Medicine (Baltimore). 2018;97(9):e0010
8. Reidling JC, Subramanian VS, Dudeja PK, Said HM. Expression and promoter analysis of SLC19A2 in the human intestine. Biochim Biophys Acta. 2002;1561(2):180-87
9. Rieck J, Halkin H, Almog S, et al. Urinary loss of thiamine is increased by low doses of furosemide in health volunteers. J Lab Clin Med. 1999;134(3):238-43
10. Akahori H, Tsujino T, Masutani M, et al. Postgastrectomy beriberi exaggerated by diuretic use: A case report. J Cardiol. 2007;49(1):49-53
11. Tsujino T, Nakao S, Wakabayashi K, et al. Loop diuretic precipitated beriberi in a patient after pancreaticoduodenectomy: A case report. Am J Med Sci. 2016;352(5):543
12. Asakura T, Kodera S, Kanda J, Ikeda M. Thiamine-responsive pulmonary hypertension. BMJ Case Rep. 2013;2013:bcr201207938