Case Report

Generalized Edema and Hyperdynamic Circulation. A Possible Case of Beriberi

Marcos F. Minicucci, Leonardo A. M. Zornoff, Mirna Matsue, Roberto M. T. Inoue, Luiz S. Matsubara, Marina P. Okoshi, Katashi Okoshi, Álvaro O. Campana, Sergio A. R. Paiva
Botucatu, SP - Brazil

We report the case of a patient with heart failure caused by beriberi, with involution of the acquired signs and symptoms after receiving thiamine.

Beriberi, a disease caused by thiamine deficiency, has 2 major clinical presentations, the wet and the dry form. Wet beriberi is characterized by signs and symptoms of high output heart failure, due to retention of sodium and water, peripheral vasodilation, and biventricular failure. Although it is an uncommon cause of heart failure, thiamine deficiency may be present in the following individuals: patients using high doses of loop diuretics; patients with diarrhea; patients undergoing peritoneal dialysis or hemodialysis; patients with parenteral nutrition; patients with thyrotoxicosis; and alcoholics. We report the case of a patient with heart failure, whose signs and symptoms subsided after being medicated with thiamine.

Case Report

The patient was a 20-year-old man of mixed heritage, farm worker, who complained of paresthesia and lower limb edema for 10 months, and generalized edema and dyspnea on exertion for 1 month. The patient denied smoking, and reported ingesting 1 to 2 bottles of beer on weekends. On physical examination, his heart rate was 100 bpm, his respiratory frequency was 16 breaths/minute, and his blood pressure was 140/70 mmHg. Lower limb edema (3+/4+), sacral edema, wide pulses, and tremor of the extremities were present. The patient had no jugular stasis, the ictus was not visible, but impulsive and palpable on the fifth left intercostal space in the left lateral decubitus, 2 cm beyond the midclavicular line, occupying 2-3 digital pulps. Heart auscultation showed 2 rhythmic heart sounds of normal intensity, with a rough holosystolic murmur (2+/6+) on the mitral and tricuspid foci. Although the respiratory examination showed no abnormalities, the examination of the abdomen showed liver 2 cm away from the costal margin, no ascites, and no other alterations.

The laboratory tests revealed no proteinuria in the 24-hour urine; normal serum levels of urea, creatinine, albumin and bilirubins; and no alterations in the coagulogram, hemogram, and thyroid tests. The chest X-ray (PA) showed a cardiothoracic index > 0.5, with bulging of the medium arch and accentuation of the vascular web (fig. 1). The electrocardiogram showed sinus tachycardia and incomplete right bundle-branch block (fig. 2). The echocardiogram on hospital admission showed enlargement of the left chambers, in addition to high values of shortening fraction and ejection fraction (tab. I).

The results of the complementary tests in addition to the clinical findings were compatible with a hyperdynamic status of circulation. Because the patient had no anemia, hyperthyroidism, or evidence of arteriovenous fistula, the diagnostic hypothesis of beriberi was established. Three days after the introduction of thiamine, the patient's edema improved, the cardiac murmur disappeared, and, on chest X-rays, the cardiac area decreased (fig. 3). The echocardiography performed 7 days after the introduction of thiamine was normal (tab. I). The patient has been followed up on an outpatient basis, currently being asymptomatic.

Discussion

In this patient's history, the presence of lower limb and sacral edema accompanied by dyspnea is the most relevant sign. The clinical findings and laboratory tests point towards the existence of a hyperdynamic condition of the circulation. Therefore, a drop in peripheral resistance and high cardiac output should be considered. The following diseases should be remembered: anemia, hyperthyroidism, arteriovenous fistula, Paget's disease, and beriberi (thiamine deficiency) [1].

Anemia was ruled out with the normal hematocrit and hemoglobin values. In regard to hyperthyroidism, the most frequent signs and symptoms were lacking, and the values of free T4 and TSH were within the normal range. The lack of antecedents that could explain the arteriovenous fistula, such as trauma and previous surgical interventions, made that diagnosis improbable. In regard to Paget's disease, no serum increase in alkaline phosphatase or radiological changes compatible with the disease were found. Therefore, all diagnostic hypotheses were ruled out, except that of beriberi.

Beriberi is the name given to the clinical alterations caused by thiamine (vitamin B1) deficiency. Thiamine is a water-soluble vitamin, essential for the formation of thiamine pyrophosphate, a coenzyme for the metabolism of carbohydrates.

Clinically, 4 fundamental forms of thiamine deficiency can be identified: Wernicke encephalopathy; dry beriberi, in which peripheral polyneuropathy predominates; wet beriberi, in which the signs and symptoms of high output heart failure are present; and Shoshin beriberi (sho = acute damage, shin = heart) associated with shock [2].

One consequence of thiamine deficiency is peripheral vasodilation, with an increase in arteriovenous shunts and a significant

Medical School of Botucatu, Universidade Estadual Paulista - UNESP
Mailing address: Marcos F. Minicucci - Departamento da Clínica Médica - FMB-UNESP - Botucatu, SP, Brazil - Cep 18618-000
E-mail: mminicucci@uol.com.br
Received: 4/8/03
Accepted: 6/16/03
English version by Stela Maris Costalonga

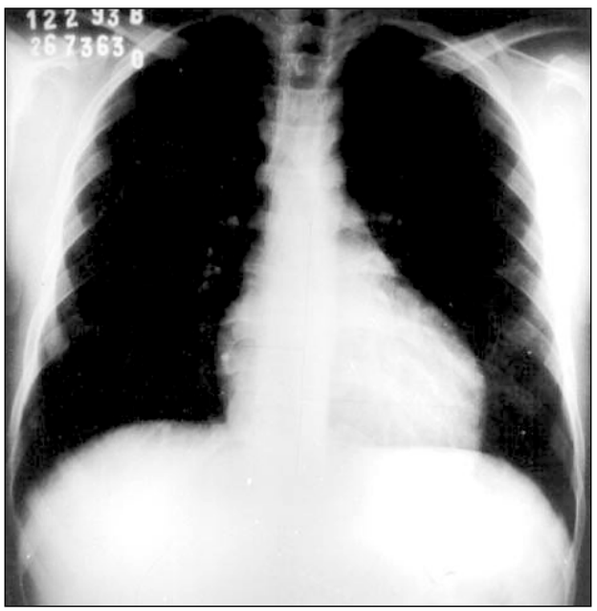

Fig. 1 - Chest radiography (PA) performed on patient's hospital admission.

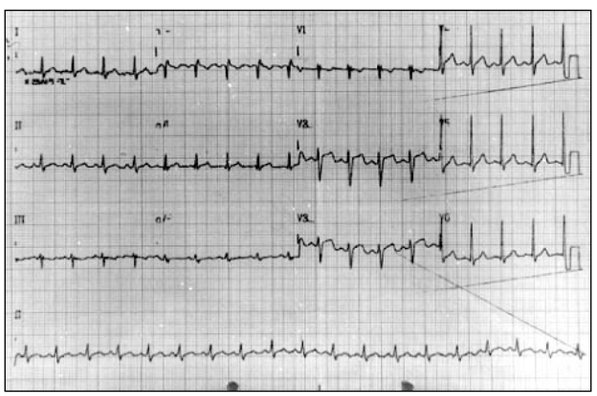

Fig. 2 - Electrocardiogram performed on patient's hospital admission.

change in the small vessel circulation, with a decrease in cerebral and renal blood flows, and an increase in muscle blood flow [3]. On the other hand, these alterations have consequences, such as an increase in peripheral venous pressure and renal retention of sodium and water [4], during which edema may be established, even in the absence of clear evidence of heart failure.

The causes of beriberi are as follows: deficient dietary ingestion; alcoholism; and the use of substances with antithiamine properties. In hospitalized patients, the following causes should be considered: parenteral nutrition; hemodialysis; peritoneal dialysis; and the use of diuretics [5-7]. The deficiency states may be aggravated by situations that acutely increase thiamine requirements, such as physical exercise, fever, and infections.

Considering the deficient food ingestion, the following are the major sources of thiamine: whole grains, cereals, meat (especially pork), poultry, fish, vegetables, and dairy products. Refined rice, sugar, fat, and several processed foods low in vitamin B_1 [5,8].

A relevant fact is that high-energy diets (mainly those rich in carbohydrates) increase the need for thiamine. The habit of alcohol ingestion is frequently associated with beriberi. In alcoholism, thiamine deficiency seems to be associated with lower ingestion of food, lower thiamine absorption, and an alteration in the intermediate metabolism of that vitamin [9].

Another aspect to be considered is the existence of substances with antithiamine activity, which may be present in teas (fermented tea leaves, extracts of tea leaves), in the nuts of certain types of palm trees, in raw fish, seafood, and coffee [5].

In an attempt to identify the causative agent in the present case, a food frequency questionnaire was used to assess food ingestion. It is a semiquantitative method that allows for the detection of dietary deviations and the assessment of the ingestion of food, such as coffee, tea, alcohol, and nuts, which can have an antithiamine action [10].

The inquiry showed that the patient had high-energy ingestion, which was 2.42 times greater than his basal energy expenditure. Thiamine ingestion, minus the loss occurring during the process of food preparation, was close to that recommended [11]. However, the ingestion of coffee and alcoholic beverages, in addition to the increased need for thiamine due to the increased energy expenditure, contributed to that vitamin deficiency.

The following criteria are used for diagnosing the cardiovascular manifestation of beriberi [12]: 1) absence of another etiological factor; 2) history of thiamine-deficient food ingestion for at least 3 months; 3) association with peripheral polyneuritis; 4) enlargement of the cardiac area, sinus tachycardia, and peripheral edema; 5) rapid response to thiamine administration.

Due to the lack of identification of another etiological factor that could explain the clinical findings, which had characteristics

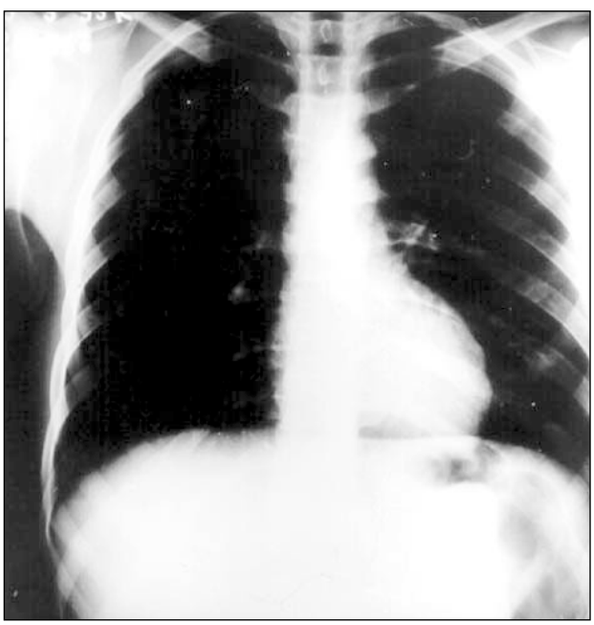

Fig. 3 - Chest radiography (PA) performed on patient's hospital discharge.

Table I - Variables related to the dimensions of the cardiac structures and left ventricular function obtained on echocardiographic study

Echocardiogram	1st	2nd
Diameter of the aorta	25 mm	27 mm
Left atrial diameter (systole)	44 mm	32 mm
Left ventricular diameter (diastole)	62 mm	55 mm
Left ventricular diameter (systole)	35 mm	37 mm
% shortening	43,0%	32,7%
Ejection fraction	0,82	0,70
Interventricular septum (diastole)	7,0 mm	7,5 mm
Posterior wall (diastole)	6,0 mm	7,5 mm

E = left; V = ventricular.

corresponding to the diagnostic criteria of thiamine deficiency, that was considered an acceptable hypothesis. During hospitalization, the patient complained of muscle aches, and the neurological examination was compatible with that of peripheral polyneuropathy. This could be a new element to reinforce the possibility of vitamin B1 deficiency. During hospitalization, in addition to rest and diuretics, vitamin B1 was added to the prescription, at the dosage of 100 mg in an intravenous bolus and 25 mg by the oral route per day for 2 weeks. The clinical findings improved, with the edema and cardiac murmur disappearing.

In addition, the chest radiography prior to treatment (fig. 1) was compared with that performed 8 days after treatment (fig. 3), when the patient had no edema. The cardiac area decreased, the cardiothoracic index being 0.41. In addition, the heart medium arch (PA view), which bulged on the first radiography, had a normal contour on the second. Comparing the first echocardiogram with that performed after clinical improvement (14 days after the initial one), the hyperdynamic condition of the circulation showed improvement (tab. I). The rapid clinical response to thiamine in young patients has been reported in the literature and has been used as a diagnostic criterion [12,13].

In regard to laboratory diagnosis, stimulation of the erythrocytic transketolase activity with thiamine diphosphate is the test recommended for confirming that vitamin deficiency [10]. Currently, this methodology is not available at our hospital.

We believe that this clinical case is relevant. In the first place, thiamine deficiency, although easily treatable, may lead to death. In addition, the usual treatment of heart failure with diuretics may worsen the case. Another aspect to be considered is the importance of echocardiography as an essential examination for the etiological diagnosis, because it identified the hyperdynamic state of the circulation in our patient. Finally, it is worth noting the importance of the clinical history and dietary inquiry for patient diagnosis and follow-up.

References

1. Okoshi MP, Okoshi K, Cicogna AC. Insuficiência cardíaca de alto débito. Doenças do coração Prevenção e tratamento (1 ed.). Rio de Janeiro: Guanabara Koogan, 1998.
2. Pereira VG, Masuda Z, Katz A, Tronchini VJ. Emergência de shoshin beribéri no Brasil: aspectos clínicos, hemodinâmicos e terapêuticos. Rev Ass Méd Brasil 1985; 31: 17-9.
3. Seligmann H, Halkin H, Rauchfleisch S et al. Thiamine deficiency in patients with congestive heart failure receiving long-term furosemide therapy: a pilot study. Am J Med 1991; 91: 151-5.
4. Blacket RB, Palmer AJ. Haemodynamic studies in high output beriberi. Br Heart J 1960; 22: 483-501.
5. Tanphaichitr V. Thiamin. In Shils ME et al. Modern Nutrition in Health and Disease (9ªed). Baltimore: Williams & Wilkins, 1998, p. 381-9.
6. Shimon I, Seligmann H, Vered Z et al. Thiamine supplements improve left ventricular function in patients with chronic heart failure. J Am Coll Cardiol 1993; 21: 366A.
7. da Cunha S, Albanesi Filho FM, da Cunha Bastos VL, Antelo DS, Souza MM. Thiamin, selenium, and copper levels in patients with idiopathic dilated cardiomyopathy taking diuretics. Arq Bras Cardiol 2002; 79: 454-65.
8. Haas RH. Thiamin and the brain. Annu Rev Nutr 1988; 8: 483-515.
9. Anderson SH, Charles TJ, Nicol AD. Thiamine-deficiency at a District General-Hospital - report of 5 cases. QJ Med 1985; 55: 15-32.
10. Willet W. Nutritional Epidemiology. New York: Oxford University Press, 1998. p514.
11. IOM. Thiamin. In: Dietary Reference Intake for thiamin, riboflavin, niacin, vitamin B6, folate, vitamin B12, pantothenic acid, biotin and choline, edited by Institute of Medicine. Washington: National Academic Press, 1999, p. 58-86.
12. Blacher C, Barbisan J. Relato de um caso da forma fulminante (shoshin) e revisão da literatura. R AMRIGS 1985; 29: 136-41.
13. Kawai C, Wakabayashi A, Matsumura T, Yui Y. Reappearance of beriberi heart disease in Japan. A study of 23 cases. Am J Med 1980; 69: 383-6.