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CASE REPORT

Non-alcoholic beriberi, Wernicke encephalopathy and long-term eating disorder: case report and a mini-review

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Abstract

Introduction Nowadays, reports of beriberi are rare in developed countries. Wernicke encephalopathy may be present in about 25% of patients with beriberi.

Case report We report the case of a woman with history of depression and chronic eating disorder, who complained Wernicke encephalopathy and beriberi. Sural nerve and muscular biopsy were performed, showing severe axonal neuropathy. Thiamine supplementation was started with rapid improvement of the pulmonary and cardiac affections; improvement of peripheral neuropathy was incomplete.

Conclusions Thiamine deficiency can be misdiagnosed. Beriberi is an important cause of acute flaccid paralysis; hence, clinicians should consider this diagnosis and prompt start thiamine treatment to avoid permanent neurological sequelae.

Introduction

Thiamine deficiency is a condition that can present with peripheral cardiac failure, neuropathy and encephalopathy. Reports of beriberi are rare in developed countries and can occur in alcohol abuse, anorexia nervosa, dieting, or malabsorption following bariatric surgery. Concurrent symptoms of Wernicke encephalopathy (WE) may be present in about a quarter of patients with beriberi [1]. Herein, we present a case of concomitant WE and beriberi in a woman with the history of depression and chronic eating disorder.

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Case report

A 47-year-old woman was admitted to hospital with a 5-day history of weight loss, abdominal pain, vomit, paraesthesia and pain in lower limbs and low-grade fever. She had a medical history of monoclonal gammopathy of undetermined significance (MGUS), hypothyroidism in replacement therapy following autoimmune thyroiditis and a chronic eating disorder (anorexia nervosa).

Abdominal and thoracic computed tomography scan was normal.

Few days after the hospitalization, she referred diplopia on right side gaze. Neurological examination revealed deficit of the right superior oblique and the inferior rectus muscles, nystagmus and weakness at left foot dorsiflexion (MRC 2/5) and at plantar flexion (MRC 4/5). Deep tendon reflexes were lost in the lower limbs.

Nerve conduction study (NCS) disclosed reduced amplitude of motor nerve action potentials in upper limbs and of bilateral median and left ulnar nerves compound muscle action potential (CMAP), with normal conduction velocities. Needle electromyography (EMG) revealed minimal myopathic signs in the proximal musculature of the lower limbs in the absence of acute denervation.

Cerebrospinal fluid (CSF) examination revealed normal count of cell, protein and glucose and a PCR positivity for Adenovirus (224 copies/ml).

Sural nerve biopsy was performed and showed severe axonal neuropathy: there was a very serious reduction of myelinated fibers with endonevrial connective tissue replacement and only rare remyelinating fibers. Right deltoid skeletal muscle biopsy was normal except for a slight neurogenic pattern (Fig. 1).

Total-body-^{18}FDG-PET excluded infection foci, but showed bilateral hypometabolism in the temporo-occipital and parietal cortex, whereas widespread hypermetabolism was detected in the bone marrow. Brain MRI revealed a FLAIR hyperintensity of the distal segment of the right intraorbitary optic nerve, associated with contrast enhancement.

Osteomedullary biopsy was performed and it showed dyserythropoiesis and lambda gammopathy (3–4%).

Serological testing for viral hepatitis, Borrelia, Adenovirus and HIV was negative, vascular endothelial growth factor (VEGF) was within normal limits.

Two weeks later, the neurological conditions worsened evolving into severe tetraparesis with associated dyspnoea and tachycardia (until 130 bpm). A transthoracic echocardiogram showed biventricular hypokinesia, severe systolic dysfunction with ejection fraction of 22%, minimal right retro-atrial pericardial and pleural effusion.

Beriberi syndrome was suspected due to her chronic eating disorder. Serum thiamine levels were < 2 nmol/L (nv 8–30). Parenteral thiamine supplementation (300 mg three times a day) was, therefore, started, replaced by oral administration (600 mg daily) a few days later.

A rapid improvement of both pulmonary and cardiac conditions occurred. However, improvement of peripheral neuropathy was quite slow and so far incomplete. Indeed, one year after the onset, the patient still presented a moderate tetraparesis and NCS revealed a severe four limbs polyneuropathy with acute denervation and initial signs of re-innervation in all the examined muscles at EMG. Serum thiamine levels were into the normal limits.

Discussion

In the last years, important progress has been made in the understanding and treatment of the medical complications of eating disorders, but a gap in many different clinical areas between current medical knowledge and more effective and evidence-based medical treatment knowledge exists [2]. As in our case, it is not frequent, for clinicians, to think about thiamine deficiency as a possible cause of polyneuropathy, particularly in patients without WE or heart failure at the initial phase.

About one-third of patients with anorexia nervosa presents thiamine deficit [3]. The classical presentation

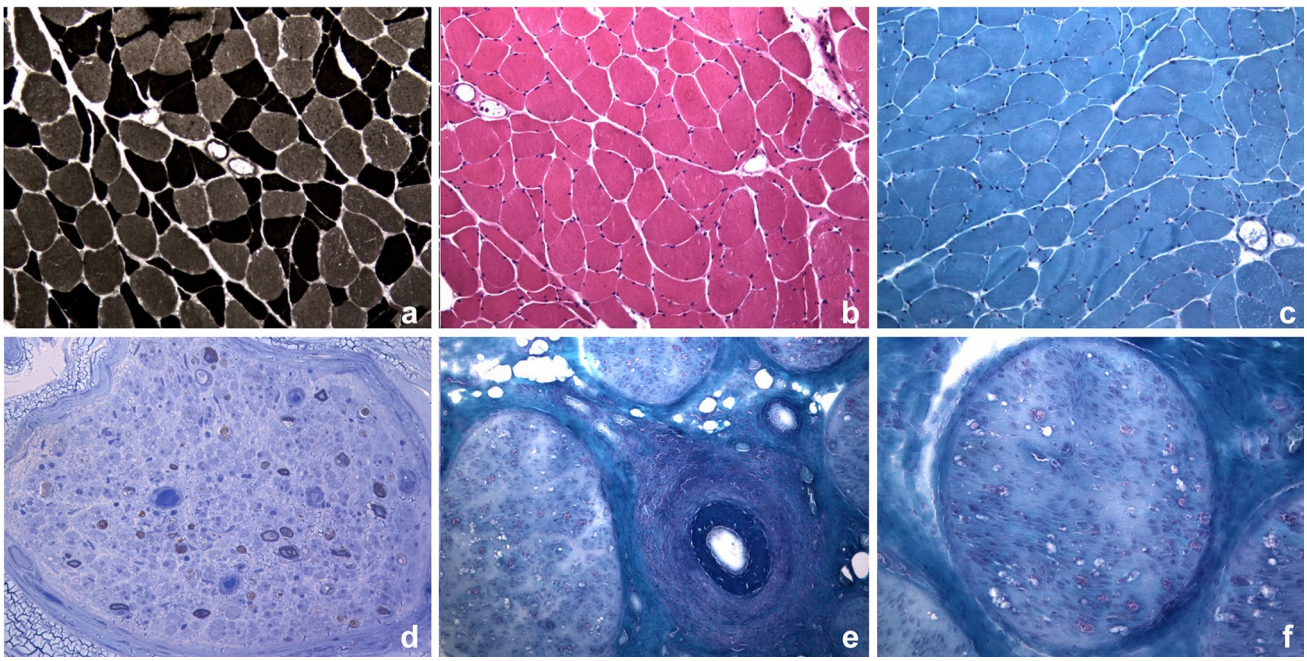

Fig. 1 Representative light microscopy images from muscle biopsy showing a mild fiber size variability with type 2 hypotrophic fibers (**a**–**c**) and representative light microscopy images from sural nerve biopsy showing severe reduction of myelinated fibers, with endoneurial connective tissue replacement (**d**, **f**), with important perivascular cellular infiltrate (**e**). **a** ATPase pH 9.4 ×200, **b** Hematoxylin & eosin staining ×200, **c** Gomori Trichrome staining ×200, **d** Toluidine blue ×400, **e** Gomori Trichrome staining ×100, **f** Gomori Trichrome staining ×200

of marked thiamine deficiency is characterized by WE and beriberi [4]. Beriberi may present in two forms: dry beriberi with progressive axonal neuropathy, neuropathic pain and gait disturbance; wet beriberi, defined by the combination of dry beriberi with cardiac failure, often accompanied by peripheral oedema. Beriberi polyneuropathy may develop gradually over weeks to months, or acutely and, therefore, difficult to distinguish from Guillain-Barré syndrome [5].

Beriberi and WE are medical emergencies and proper and prompt diagnosis is fundamental to start an appropriate treatment with thiamine. In beriberi neuropathy, NCS usually show sensory predominantly sensory axonal neuropathy, typically with absence of sural-sparing pattern and normal CSF protein count [1].

WE and beriberi were reported rarely in patient with anorexia nervosa. The recognition of signs and symptoms of WE is often late, probably due to the overlap between symptoms of thiamine deficiency and symptoms of WE [6]. To our knowledge, only two cases describing WE and beriberi secondary to anorexia nervosa are reported: one in an adolescent boy [7] and one in a 25-year-old woman [8].

It is reported that anorexia nervosa may also induce myopathy [9]. In our patient, the minimal myopathic signs at EMG were not confirmed at skeletal muscle biopsy in which only slight neurogenic signs were detected.

Other cases reported only neuropathy or Korsakoff's syndrome in paediatric [10–12] and in adult patients [13–16] with malnutrition, even mimicking a Guillain-Barrè syndrome. Peripheral neuropathy and/or WE secondary to thiamine deficiency are reported in patients with alcohol abuse, dialysis, malabsorption or hyperemesis gravidarum [17–19]. In particular, some cases of WE have been described following restrictive bariatric surgery procedures [20, 21].

In conclusion, the present case shows that thiamine deficiency involves multiple systems and can be misdiagnosed. More specifically, beriberi is an important cause of acute flaccid paralysis, even more in the presence of a history of dietary deprivation. Even in Western countries, clinicians should consider this diagnosis and start prompt empirical thiamine treatment to avoid permanent neurological sequelae. High-dose thiamine supplementation is inexpensive and rapidly effective.

What is already known on this subject?

Although cases are rare today, thiamine deficiency can present with neuropathy and encephalopathy in anorexia nervosa. In fact, about 1/3 of patients with anorexia nervosa presents thiamine deficit.

What does this study add?

Prompt thiamine treatment is important to avoid permanent neurological sequelae. In Western countries thiamine deficiency is rare and misdiagnosed, so it is important to consider this diagnosis.

Compliance with ethical standards

Conflict of interest The authors declare no actual or potential conflicts of interest including any financial, personal, or other relationships with other people or organizations. No financial support has been related to the manuscript being submitted. All authors approved the contents of the manuscript and validated the accuracy of the data.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from the patient for the publication of this case report.

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