

A Case of Iatrogenic Wernicke Encephalopathy Following Prolonged Total Parenteral Nutrition

Uzamış Total Parenteral Nutrisyon Sonrası Gelişen İyatrojenik Wernicke Ensefalopatisi Olgusu

Mustafa Çetiner¹, Sibel Canbaz Kabay¹, Özgül Ocak², Selahattin Ayas¹

¹ Dumlupınar Üniversitesi Tıp Fakültesi, Nöroloji Anabilim Dalı, Kütahya, Türkiye

² Çanakkale Devlet Hastanesi, Nöroloji Kliniği, Çanakkale, Türkiye

ABSTRACT

Thiamine is a water-soluble vitamin that plays a role in the metabolism of glucose. Thiamine deficiency affects cardiovascular and nervous systems. Wernicke encephalopathy is characterized by oculomotor abnormalities, ataxia and cognitive deterioration caused by thiamine deficiency. It is most commonly observed in chronic alcoholics. It may also occur in patients who receive long-term total parenteral nutrition. In this study, we present the case of a 50-year-old male patient who underwent surgery for laryngeal cancer, who stayed in the intensive care unit for a prolonged period and received total parenteral nutrition because of the inability of oral intake.

Keywords: Wernicke encephalopathy, total parenteral nutrition, thiamine deficiency

ÖZ

Tiamin (Vitamin B1) glikoz metabolizmasında rol alan suda çözünen bir vitamindir. Eksikliğinde kardiyovasküler sistem ve sinir sistemi etkilenir. Wernicke ensefalopatisi, klinik olarak okulomotor anormallikler, ataksi ve mental durum değişiklikleri ile karakterize tiamin eksikliğinden kaynaklanan bir ensefalopati tablosudur. En sık olarak kronik alkol bağımlılarında görülmektedir. Uzun süre total parenteral nutrisyon desteği alan hastalarda da gelişebilmektedir. Yazımızda larenks kanseri nedeniyle opere olan, uzun süre yoğun bakımda kalan, oral beslenememesi nedeniyle total parenteral nutrisyon desteği alan ve Wernicke ensefalopatisi tanısı konularak nöroloji yoğun bakıma alınan 50 yaşında erkek hasta sunulmuştur.

Anahtar Kelimeler: Wernicke ensefalopatisi, total parenteral nutrisyon, tiamin eksikliği

Gönderme tarihi / Received: 31.03.2018 Kabul tarihi / Accepted: 22.07.2018

İletişim: Mustafa ÇETİNER, Dumlupınar Üniversitesi Tıp Fakültesi, Nöroloji Anabilim Dalı, Kütahya, Türkiye

ORCID ID: orcid.org/0000-0002-4420-6452

Tel: 0 505 454 3869 E-posta: drcetiner76@gmail.com

INTRODUCTION

Thiamine (vitamin B1) is a water-soluble vitamin which has a role as a coenzyme for neural activity and glucose metabolism and is absorbed by the duodenum and jejunum (1-3). Thiamine deficiency may cause Wernicke encephalopathy (WE), a serious neurological defect characterized by the clinical triad of ophthalmoplegia, ataxia and acute mental confusion (4). Although it is most commonly observed in chronic alcoholics because of chronic malnutrition, it can also acutely occur in cases with prolonged starvation, with hyperemesis gravidarum and undergoing gastrointestinal system surgery (5, 6). Moreover, it can iatrogenically develop because of multivitamin deficiency in patients receiving long-term total parenteral nutrition (1, 7, 8). In recent years, the prevalence of WE has increased because of thiamine deficiency in patients receiving parenteral nutrition (1). Here, we present the case of a 50-year-old male patient who underwent surgery for laryngeal cancer and who stayed in the intensive care unit for a prolonged period and received total parenteral nutrition support because of inability of oral intake.

CASE

Approximately 2 months ago, a 50-year-old male patient had undergone bilateral hemi-

laryngectomy for laryngeal cancer. He received total parenteral nutrition support because of inability of oral intake for the last 1 month. The patient was consulted to us following the development of tendency to somnolence. The patient's medical and family histories were not remarkable. On physical examination, blood pressure was 100/70 mmHg and pulse was 88/minute. Neurological examination revealed tendency to somnolence and the patient opened his eyes to verbal stimulations and responded with one-word answers. He had difficulty in following simple instructions. Pupils were isochoric; direct and indirect light reactions were bilaterally positive. Eye movements could not be evaluated because of the tendency to somnolence. There were no lateralizing symptoms on motor examination. Deep tendon reflexes were normoactive and the plantar reflexes were bilaterally normal. Cranial computed tomography showed normal findings. Diffusion-weighted imaging (DWI) and fluid-attenuated inversion recovery (FLAIR) weighted cranial magnetic resonance imaging (MRI) revealed hyperintense appearance in the bilateral thalamus and tectum (Figure 1).

Apparent diffusion coefficient (ADC) series also showed hyperintense appearance. Cerebral venography examination was normal.

It was thought that the lesions were consistent with vasogenic oedema.

Figure 1. Hyperintensity on diffuse MRI and flair weighted axial images in bilateral thalamus and tectum

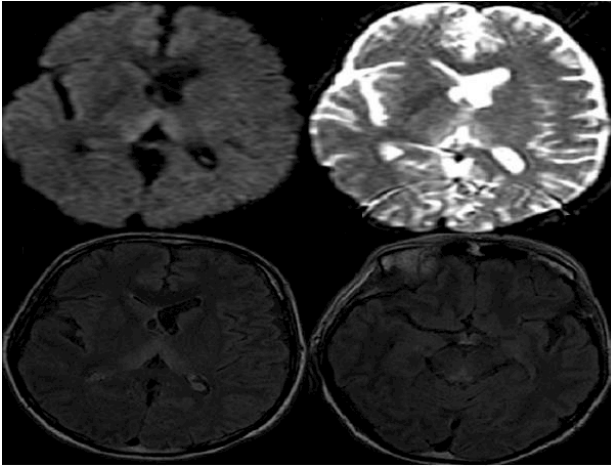
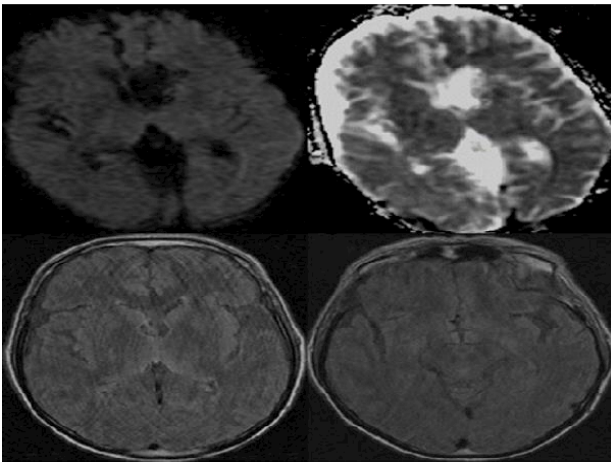


Figure 2. The lesions are regressed after the thiamine replacement on diffuse MRI and flair weighted axial images



The patient was admitted to the neurology intensive care unit. The patient was first evaluated for the presence of metabolic causes. Complete blood count and biochemistry were normal. Vasculitic parameters and

cerebrospinal fluid examinations were normal. The patient was considered to have WE based on clinical and imaging findings. The patient was monitored while he received thiamine supplementation and supportive treatment. The patient was maintained on enteral nutrition through an orogastric tube. Vital signs remained stable. There was remarkable improvement in neurologic deficits after 15 days. The patient has become fully cooperative. There was no gaze palsy. No motor deficit was detected. Control magnetic resonance imaging of the brain showed remarkable regression of the lesions (Figure 2).

DISCUSSION

Thiamine (vitamin B1) is a water-soluble vitamin absorbed from the duodenum and jejunum and acts as a coenzyme that is essential for glucose metabolism and neural activity (1-3). The prevalence of WE is considerably high in patients receiving prolonged total parenteral nutrition (TPN) without thiamine supplementation. Francini-Pesenti has emphasised that the prevalence of WE is high in such patients (1). The present patient underwent head-neck surgery because of malignancy and received prolonged TPN support.

A normal individual's body stores up to 30–50 mg thiamine and requires 1–2 mg/day of thiamine. Thiamine storage is depleted

within 3–4 weeks of thiamine-deficient nutrition, and the symptoms of WE then manifest (9). The present patient received TPN support for one month. Clinical symptoms began to appear at the end of the one-month period.

WE is an acute neuropsychiatric syndrome caused by thiamine deficiency, and it is characterised by a triad of ataxia, oculomotor dysfunction and mental status change (5). Only one-third of the patients exhibit all symptoms. The most prevalent symptom is mental confusion, followed by ataxia and ocular dysfunction; 19% of the patients may exhibit no symptoms at all. Mental status change may further progress to apathy and deep coma if it's not treated (5, 10). The patient was in stupor at the time of consultation. Therefore, other clinical signs could not be evaluated.

The diagnosis of WE is based on clinical evaluation. The guidelines of the European Federation of Neurological Societies recommend the presence of at least two of the following four symptoms to make a clinical diagnosis of WE in non-alcoholic patients: dietary deficiencies, eye symptoms, cerebellar dysfunction and altered mental state or mild memory impairment (4). The present patient had two among the four above mentioned symptoms: prolonged malnutrition and altered mental state.

Cranial MRI is a valuable imaging method for diagnosis. T2-weighted and FLAIR images represent symmetric mammillary bodies and hyperintense signals in the periventricular thalamus and periaqueductal grey matter and midline structures in the periphery of the third ventricle. In addition, atypical changes may also be observed. These lesions may cause increased diffusion and DWI (9, 11–13). MRI performed after the initial neurological examination in the present patient demonstrated hyperintensity in bilateral thalamus and midbrain tectum in FLAIR sequences, and DWI showed hyperintensity that was associated with the vasogenic oedema. Following parenteral thiamine replacement, neurological symptoms and radiological images showed significant improvement.

WE is an emergency medical condition. It's treatment should be initiated immediately after the suspicion of the condition without testing thiamine levels (14). Patients with severe thiamine deficiency must be treated with intravenous thiamine replacement, with a dose of 50 mg/day for 3–5 days, followed by 250 mg/day for the following 3–5 days or until the symptoms disappear. Upon initiation of oral food intake, administration of 100 mg/day of thiamine is recommended until the risk factors are eliminated (15). However, there is uncertainty regarding the standard

dosage and duration of treatment in clinical practice because current data mostly relies on observational studies and case series (16). High-dose thiamine treatment (≥ 500 mg/day) was found to be effective and safe in a study by Nishimoto et al. (17) The symptoms may rapidly disappear. Some patients show a remarkable improvement after the administration of the first dose (14). In the present patient, WE was considered based on the patient's medical history, radiological findings and neurological symptoms, and intravenous thiamine treatment was promptly initiated. The patient received intravenous thiamine of 500 mg/day for the first 3 days, intravenous thiamine of 200 mg/day for the following 3 days and i.v. thiamine of 100 mg/day for the following 10 days. Radiological images and clinical status of the patient showed improvement after approximately 2 weeks.

WE must be considered in the differential diagnosis even if the classical triad is not observed. WE should be especially suspected when neuropsychiatric symptoms occur in patients who receive TPN in the postoperative period. Early diagnosis and treatment may prevent severe TPN.

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