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Case Report

Gayet Wernicke's encephalopathy with cortical damage following a subtotal gastrectomy: An uncommon association ☆

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ABSTRACT

Wernicke's encephalopathy is a pathological entity caused by Vitamin B1 (Thiamine) deficiency in malnourished individuals, especially alcoholics, patients operated for digestive surgery or suffering from gastrointestinal tract's diseases or incoercible vomiting. Classically it manifests by confusion with oculomotor disorders and ataxia. However, other neurological manifestations are possible. Magnetic resonance imaging is the gold standard imaging technique for diagnosis. It shows signal abnormality on periventricular area around the third and fourth ventricles and on mammillary bodies in the most common cases, however other localizations are possible, in particular the cerebral cortex, which can explain the occurrence of epileptic seizures in some patients. Early administration of Thiamine, intravenously or intramuscularly, allows ad-integrum recovery, while delayed treatment is associated with serious consequences in terms of mortality and morbidity with debilitating neurological sequelae. The presence of cortical lesions is of poor prognosis despite a well-managed treatment. In this article, we report a nonalcoholic Wernicke's encephalopathy case, following a subtotal gastrectomy. Epileptic seizures were the major clinical manifestation, related to the associated cortical lesions. Despite early and well-managed treatment, the patient had a poor prognosis, with progression, after one month, to a persistent chronic vegetative state.

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Introduction

Wernicke's encephalopathy (WE) is a severe metabolic disease of the central nervous system caused by deficiency of thiamine (vitamin B1). It manifests by ocular abnormalities, ataxia and changes in consciousness. MRI is the examination of choice to confirm diagnosis and assess the lesion. Typical imaging findings are symmetric and bilateral signal abnormality in the mammillary bodies, medial thalamus, periventricular regions around the third and fourth ventricles. Now we report a case of WE after subtotal gastrectomy with atypical cortical lesions, in the origin of epileptic seizure as initial symptom.

Case report

A 54-year-old man diagnosed for WE with epileptic seizures and unconsciousness, following a partial gastrectomy. The patient had an estimated weight of 52 kg, with a history of chronic smoking at the rate of 30 pack/years and not known alcoholic. In admission, the patient had epigastralgia, postprandial vomiting evolving since 7 months, hematemesis and weight loss estimated at 15% during the last 6 months. The oeso-gastric fibroscopy shows a gastric wall injury and biopsy confirmed a gastric adenocarcinoma. The patient was admitted in digestive surgery department for a subtotal gastrectomy with gastrojejunal anastomosis. In the postoperative period, patient was conscious, without motor or sensory deficit. A transanastomotic enteral nutrition with parenteral complement were adopted. The patient remained hospitalized in surgery for occurrence of a digestive fistula on the gastrojejunal anastomosis. Four weeks after the intervention, the patient was admitted to intensive care department for a generalized epileptic seizure with altered state of consciousness. The patient was cachexic, unconscious with a Glasgow score of 7/15, without fever. Pupils were symmetric and reactive. The patient presented also dyspnea with snoring on pulmonary auscultation and a hypoxia with 90% SpO_2 under 10 L/min of oxygen, the capillary glycemia was 1.05 g/L. Another generalized epileptic seizure has occurred; the patient is then intubated, ventilated and put on Midazolam, Fentanyl and Levetiracetam 1000 mg/day. Laboratory investigations revealed the following: Anemia with 8 g/dl of hemoglobin, alkalosis at 37.9 mmol/L, hypokalemia at 3 mmol/L, hypochloremia at 92 mmol/L, hypernatremia at 148 mmol/L, hypoalbuminemia at 29.2 g/L, biological inflammatory syndrome with a CRP at 100 mg/L, and leukocytosis at 16,300 e/mm. A meningoencephalitis was evoked at first and a lumbar puncture was performed. In cerebrospinal fluid: protein level at 0.35 g.L, Glucose level at 0.7 g/L, and cell numbers were at 3 elements/mm, within the normal range. The liver balance and the renal function were also without abnormalities.

Brain CT imaging was without abnormality (Fig. 1). Magnetic resonance imaging (MRI) was performed with a 1.5-Tesla magnet. On the Brain MRI, there were bilateral and symmetric T2-weighted and Flair hyperintensities in the mammillary bodies, medial thalamus, periaqueductal area and in the parietal cortex (Fig. 2). The diffusion-weighted imaging (DWI) showed signal hyper intensities in the above positions without restriction of the apparent diffusion coefficient (Fig. 3). There was no signal abnormality on T1-weighted sequences (Fig. 4). A Gayet WE with cortical damage was the most appropriate diagnosis. Vitamin B1 level was low at 66 mmol /L, with a high level of vitamin B12 at 1795 pg/ml and a normal level of Vitamin B6. Immediately, an injectable vitamin B1 therapy was started at the rate of 500 mg/day for 3 days, followed by an oral relay.

The patient opened spontaneously his eyes and moved his limbs with difficulty. Sedation was stopped 24 hours later. However, the patient remained dependent on the ventilator, a tracheostomy was performed at the 20th day. The patient leaved the intensive care department after 1 month of hospitalization in a persistent chronic vegetative state.

Discussion

Gayet WE is a severe metabolic disease of the central nervous system, related to a deficiency in vitamin B1 and was first described in 1881 by Dr. Carl Wernicke. The classic clinical triad of symptoms observed in WE are acute mental confusion, ataxia, and ophthalmoplegia with serious consequences if untreated [1].

Thiamine (vitaminB1) is a water-soluble vitamin essential in carbohydrate metabolism [2]. Cerebral metabolism is highly dependent on thiamine, and deficiency may result in brain injury, particularly in regions with higher metabolic demands, including primarily the cerebellar vermis, dorsomedial thalamic nuclei, periaqueductal gray matter, and mammillary

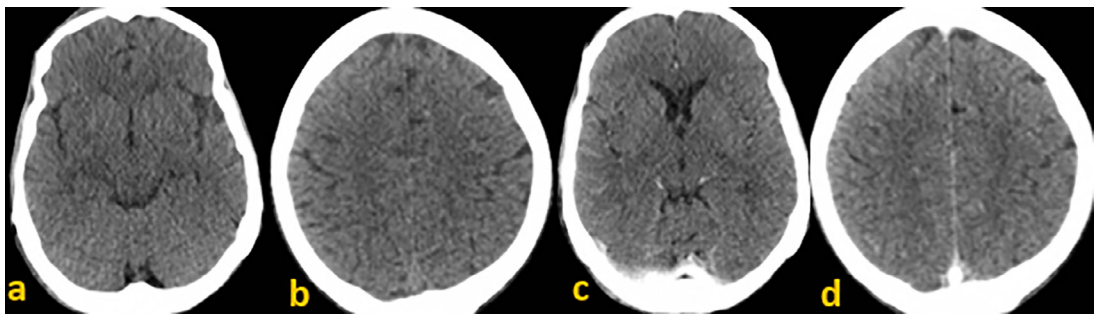

Fig. 1 – Axial computed tomography (CT) without (a, b) and with (c, d) contrast agent injection: no abnormal signal.

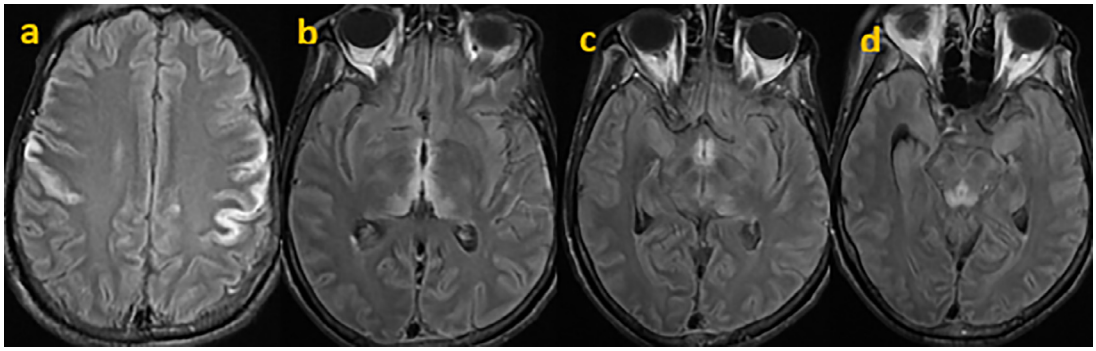

Fig. 2 – Axial sections of MRI in FLAIR sequence: signal abnormalities on parietal cortex (a), in, medial thalamus (b), in periaqueductal area and the mammillary bodies (c, d).

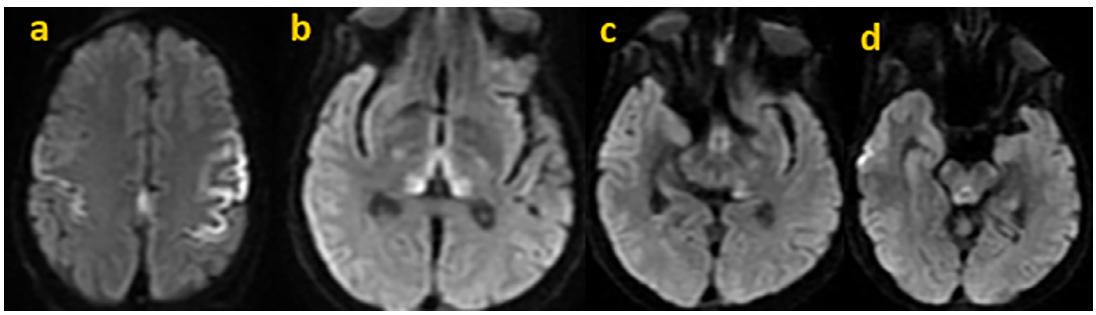

Fig. 3 – Axial section of MRI in diffusion sequence B1000: signal abnormalities on parietal cortex (a), in, medial thalamus (b), in periaqueductal area and the mammillary bodies (c, d).

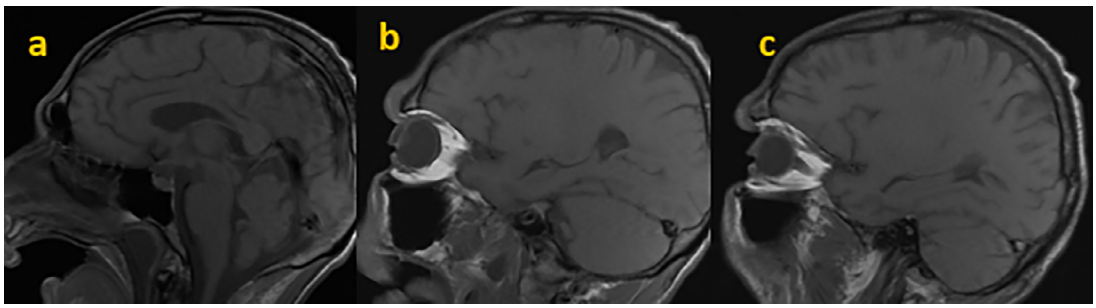

Fig. 4 – No signal abnormality on T1-weighted ponderation (a-c).

bodies [3]. As animals and humans cannot produce thiamine, it should be obtained from exogenous sources; thus, thiamine can be depleted within 2-3 weeks after deficient diet or malabsorption [3].

Most cases of WE are due to chronic alcoholism. 30%-80% of alcoholics had signs or laboratory studies compatible with thiamine deficiency. Nonalcohol-related conditions also predispose to thiamine deficiency, as demonstrated in gastrointestinal surgical procedures. Extensive gastrectomy for gastric neoplasia is one of these surgeries. Thiamine deficiency is directly proportional to the amount of resected gastric mucosa [3]. In our patient, subtotal gastrectomy and enteral nutrition through a jejunostomy, causing foodstuff to circumvent the duodenum and jejunum tract, where absorption of thiamine mainly occurs, certainly played a role in the development of WE [4].

WE remains a diagnosis based on clinical findings, requiring a high index of suspicion. Confirmation may be obtained by measuring blood thiamine concentrations, or measuring the red blood cell trans-ketolase activity. However, these tests are limited due to technical difficulty and lack of specificity [4]. The diagnosis is supported by the response and resolution of neurologic signs with administration of parenteral thiamine. Despite its low sensitivity of 53%, MRI is now considered confirmatory for WE, as it has a 93% specificity to rule out the disorder [4]. Bilateral and symmetric signal abnormalities can be seen in the paraventricular regions of the thalamus, mammillary bodies, periaqueductal region, the floor of the fourth ventricle, and the midline cerebellum [3]. WE associated lesions appear as hypointense on T1-weighted images, hyperintense on FLAIR, and T2-weighted images. Reversible cytotoxic edema is considered the most distinctive lesion of WE and it

is easily shown on MRI on DWI as restricted diffusion. Hong et al. and Chung et al. reported hyperintensity on both DWI and apparent diffusion coefficient in acute WE suggesting a vasogenic edema, as the pathological mechanism for lesions, such as in our case [5]. Involvement of cerebral cortex is rare, less than 40 cases have been reported in the literature until this day [6]. WE with cortical damage (WEc) was reported generally and according to literature data in nonalcoholic patients [7]. The hypothesis of a possible protective effect of alcohol on the brain was raised. While Keita Sakurai et al. noted, through a literature review, that only 30% of WEc were alcoholics [1].

Impairment of the cortical gyrus is rarely reported and usually results in a worse prognosis or sequelae [6]. The frontal and the parietal lobes were the most susceptible areas, especially the central sulcus. Symmetrical banding signs may be a distinctive characteristic in the early stages of the condition. The condition can be reversed if patients receive timely treatment. Persistent hyperintensity on T2-FLAIR or gadolinium enhancement predicts poor prognosis. However, in postmortem examination in WE, cortical involvement is not an uncommon pathologic change. The histological changes in the motor gyrus are identical to those in other typical WE lesions such as in the mammillary body. Furthermore, cortical laminar necrosis can be found and ascribed to thiamine deficiency. Therefore, the clinical manifestations and radiological changes in the cortex may resolve due to elimination of cytotoxic edema, as in other areas [6].

Several treatment regimens were studied [4,8] in patients highly suspected of suffering from WE. Empirical treatment with 500 mg of thiamine hydrochloride infused over 30 minutes, 3 times a day for 2-3 days should be administered [3]. If vitamin absorption is restored, the patient may be shifted to high potency oral B-vitamin complex supplementation and maintained until deficiency factors are eliminated [3]. On the other hand, when no clinical response is demonstrated, the infusion should be discontinued. In the patient case, treatment was started with intravenous injections at a dose 500 mg of thiamine per day.

A preventive approach can also be taken and a prophylactic thiamine administration in patients at risk, specifically alcoholics, pregnant women, and patients with compromised absorption should be undertaken. A routine thiamine supplement therapy and monitoring of thiamine levels are recommended in patients after gastrointestinal surgery [10]. Attention should also be paid to elderly patients with multiple risk factors for nutritional deficiency, especially following gastrectomy. The early recognition of WE is crucial, considering the poor prognosis without appropriate treatment. It should also be kept in mind that undergoing gastrojejunostomy is a risk factor for nonalcoholic WE in elderly patients, particularly in those with poor intake status [4].

Conclusion

Cortical impairment in WE is not a common manifestation. However, cortical lesions lead to worse outcomes. The frontal and the parietal lobes were the most susceptible areas, especially the central sulcus. MRI is essential for diagnosing and monitoring patients with WE with cortical abnormalities due to the risk of lesions irreversibility, which may progress to cortical laminar necrosis and hemorrhage. The condition can be reversed if patients receive timely treatment [9].

Informed consent

I, the undersigned, ROUATI Taoufik, of Moroccan nationality and bearer of the national identity card number: CB218731.

As much as cousin of Mr. BELHADDAOUY Khalid hospitalized in intensive care department, I declare to have been informed of the publication of informations and radiological images of my cousin for scientific purposes, in the form of a paper entitled: Gayet Wernicke's encephalopathy with cortical damage following a subtotal gastrectomy: an uncommon association.

This article will not include the patient's name or personal photo.

For these purposes, I approve my consent.

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