Case Report

ANNALS OF MEDICINE & SURGERY

OPEN

Wernicke encephalopathy revealed by areflexic flaccid tetraparesis associated with gravidarum hyperemesis

Rajae Ben Chaib, MD[a,b,c,*], Ikram Mekkaoui, MD[a,b,c], Zaima Chetouani, MD[a,b,c], Ousmane kaba, MD[a,b,c], Ikam Markaoui, MD[a,b,c], Sanae Elmezzeoui, MD[a,b,c], Houssam Bkiyar, MD[a,b,c], Brahim Housni, MD[a,b,c]

Introduction: Wernicke's encephalopathy is an acute neuropsychiatric syndrome resulting from thiamine deficiency, which is associated with significant morbidity and mortality. The diagnosis of Wernicke's encephalopathy is based on the clinical manifestations and rapid reversal of symptoms with thiamine.

Case presentation: The authors present the case of a 25-year-old female patient at the 19th week of gestation (Gravid 1 para 0) with an unremarkable medical history who was admitted to the hospital for an areflexic flaccid tetraparesis with ataxia after persistent vomiting. The brain and spinal MRIs did not reveal any abnormalities, and the evolution was marked by an important improvement after supplementation with thiamine.

Conclusion: Gayet Wernicke encephalopathy is a medical emergency. Clinical symptoms are inconstant and varied. MRI is the reference examination to confirm the diagnosis, but in 40% of cases it is strictly normal. Early thiamine administration can prevent morbidity and mortality in pregnant women.

Keywords: ataxia, gravidarum hyperemesis, MRI, thiamine, wernicke encephalopathy

Introduction and importance

Wernicke encephalopathy (WE) is defined by the presence of neurological symptoms caused by biochemical injuries of the central nervous system after exhaustion of B-vitamin reserves, in particular thiamine (vitamin B1). The classic triad of symptoms found in WE is ophthalmoplegia, ataxia, and confusion.

In the context of hyperemesis gravidarum, pregnant women have an increased demand for thiamine, which rapidly depletes, leading to WE, and this happens typically between the 14th and 20th weeks of gestation[1].

We report a case of a pregnant woman with WE revealed by areflexic flaccid tetraparesis.

This case report follows Surgical CAse REport (SCARE) Guidelines[2].

HIGHLIGHTS

- Wernicke's Encephalopathy (WE) complicating hyperemesis can be responsible of a big morbidity.
- Clinical symptoms of WE are inconstant and varied; in our case, it has been revealed by areflexic flaccid tetraparesis.
- The brain MRI is essential, but in our case, it did not show any abnormalities.
- Gayet Wernicke encephalopathy is a medical emergency, treatment should not be delayed until confirmation of the diagnosis.
- Early thiamine replacement can prevent morbidity and mortality in pregnant women.
- WE revealed by areflexic flaccid tetraparesis associated with gravidarum hyperemesis (a case report).

Case presentation

A 25-year-old woman (G1, P0) with an unremarkable medical history. She had no history of heavy alcohol consumption, but she had an unbalanced diet since pregnancy. She had never had surgery.

She began vomiting during the 8th week of gestation, for which she was treated with antiemetics, but she continued to have persistent vomiting and complained of general weakness. At 17 weeks of gestation, she complained of tetraparesis. She was brought to the emergency department, where she was treated with intravenous fluids, including dextrose and metoclopramide, and was referred to the ICU in our hospital.

At admission, we found a patient with a Glasgow Coma Scale of 15/15. She was aware of time and place. The neurological examination revealed a loss of equilibrium as well as

[a]Department of Intensive Care Unit, Mohammed VI University Hospital, [b]Faculty of Medicine and Pharmacy, Mohammed First University and [c]Mohammed First University, FMP, Oujda, Morocco

Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

*Corresponding author. Address: Mohammed I University, Universite Mohammed Premier Oujda, Morocco. Tel.: 212654341512. E-mail: rajaebcb@gmail.com (R. Ben Chaib).

Annals of Medicine & Surgery (2023) 85:2127–2129

Received 24 January 2023; Accepted 22 March 2023

Published online 11 April 2023

http://dx.doi.org/10.1097/MS9.0000000000000579

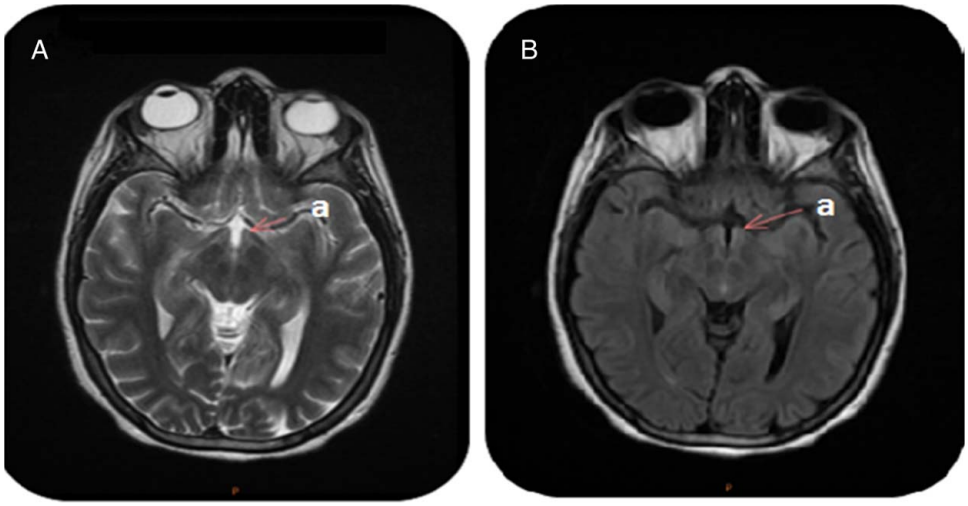

Figure 1. (A) MRI T2 image showing no brain abnormalities: the mammillary body T2 image of case 3 showing no brain abnormalities (B) T2 Axial flair MRI.

incoordination of gait (ataxia). The pupils were bilaterally equal and reactive, and the ocular fundus was normal. The cranial nerve examination was normal.

She had a deficit of the thumb index clamp and of the dorsiflexion of the wrist, and concerning the inferior members, she also had a deficit in the dorsiflexion at 3/5 and in the extension at 4/5. Reflexes were absent, the sensory exam was normal, and Babinski was indifferent.

Her physical examination revealed that she had a weight of 59 kg, a temperature of 37.3°C, a blood pressure of 109/61 mmHg, and a pulse rate of 98 beats per min.

Her initial laboratory workup was as follows: white blood cell count, 9030/µl ; C-reactive protein, 22.09 mg/l; procalcitonin, 0.21 ng/l; electrolytes; the level of thyroid hormones was normal; serum creatinine level was 63 mol/l (normal range (NR): 50–100 mol/l), and urea level was 4,2 mmol/l (NR: 2.5–6.1 mmol/l). Liver function tests showed an L-aspartate aminotransferase level of 21 IU/l (NR < 40) and an L-alanine aminotransferase level of 17 IU/l (NR < 45). The serum potassium level was 2.8 mEq/l. And the level of thiamine was at 10 nmol/l (NR 74–222 nmol).

The hypokaliemia was initially corrected, but there was no improvement in clinical symptoms. She was treated with intravenous fluids, including dextrose and electrolytes. Metoclopramide and Zofran were introduced to control vomiting.

Then we completed our exploration with a cranial magnetic resonance imaging (MRI) that was normal (Fig. 1), and the spinal MRI and ENMG did not reveal any abnormalities.

The patient was first treated for 3 days with 500 mg of thiamine, parenterally injected three times a day once the diagnosis was evident, and then the treatment was maintained with 100 mg per day for another 14 days.

On the 19th day of hospitalization, she improved and was able to walk. She was sent home on the 37th day with mild ataxia.

Clinical discussion

WE is common, often missed and preventable, with a high morbidity and a 10–20% mortality. In the Western world, WE is most commonly found in malnourished alcoholics, but it is also found in other conditions associated with vitamin deficiency, for example, after bariatric surgery, in anorexia nervosa–bulimia, and in hyperemesis gravidarum (in which the vitamins and nutrition may be vomited and not absorbed), as well as with dialysis, if thiamine therapy is not given after or during dialysis. It also occurs occasionally if parenteral nutrition is given without thiamine[3].

Up to WE complicating hyperemesis during pregnancy, On the fetal side, WE can lead to miscarriage, preterm birth, and intrauterine growth retardation[4], and it happens especially if the diagnosis is delayed because of atypical symptoms or the presence of disturbances in liver function tests, which are frequent in hyperemesis and can lead one to initially suspect hepatic encephalopathy. Steven C[5] describes in his manuscript a case of spontaneous abortion.

The diagnosis of WE is based on the clinical manifestations and rapid reversal of symptoms with thiamine. Determination of blood transketolase activity and thiamine pyrophosphate reflects the thiamine status in the body[6]. Our patient presented with areflexic flaccid tetraparesis following intractable vomiting during her active pregnancy of 16 weeks of amenorrhea without thiamine supplementation.

In imaging, MRI shows abnormalities in 60% of cases, which implies that normal imaging does not exclude the diagnosis[7,8]. This is the case of our patient, but we can observe, in the days following the onset of clinical signs, hypersignals in T2, FLAIR, and diffusion, typical by their location and their symmetrical character, around the aqueduct of Sylvius, the third Ventricle (V3), the medial face of the thalami, and above all at the level of the mammillary tubercles. Diffusion sequences reveal hypersignal zones that predict long-term neurological sequelae[9].

Once the diagnosis is established, treatment must be administered parenterally as soon as possible; in our case, we had to correct hypokalemia first, which is a common cause of tetraplegia. The evolution was favorable under vitamin therapy.

Conclusion

Clinical symptoms are inconstant and varied. Early thiamine replacement can prevent morbidity and mortality in pregnant

women. The WE was revealed in our clinical case by areflexic flaccid tetraparesis in the first place, followed by ataxia in the context of pregnancy-induced vomiting. The brain and spinal MRIs did not reveal any abnormalities, and the evolution was marked by an important improvement after supplementation with thiamine.

Ethical approval

The ethical committee approval was not required give the article type (case series). However, the written consent to publish the clinical data of the patients was given and is available to check by the handling editor if needed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author contribution

Dr R.B.C. is principal investigator that collected and analyzed data, wrote the manuscript, and prepared the final draft for the submission. B.H. and H.B.: supervised the research project and approved the final draft for publication. All authors approved the final version of the manuscript.

Conflicts of interest disclosure

The authors state that they have no conflicts of interest for this report.

Research registration unique identifying number (UIN)

This is not an original research project involving human participants in an interventional or an observational study, but a case report. This registration was not required.

Guarantor

Ben Chaib Rajae.

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

[1] Oudman E, Oey MJ, Batjes D, *et al*. "Wernicke-Korsakoff syndrome diagnostics and rehabilitation in the post-acute phase". *Addiction Neuroscience*. 2022;4:100043.
[2] Agha RA, Franchi T, Sohrabi C, *et al*. The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. Int J Surg 2020;84:226–30.
[3] Young GB. Wernicke's Encephalopathy. Encyclopedia of the Neurological sciences, 2nd edition. Elsevier; 2014:753–4. doi: 10.1016/B978-0-12-385157-4.00349-3.
[4] Michel ME, Alanio E, Bois E, *et al*. Wernicke encephalopathy complicating hyperemesis gravidarum: a case report. Eur J Obstet Gynecol Reprod Biol 2010;149:117–23.
[5] Spruill S C, Kuller J A. Hyperemesis gravidarum complicated by Wernicke's encephalopathy. Obstet Gynecol 2002;99(5 Pt 2):875–7.
[6] Netravathi M, Sinha S, Taly AB, *et al*. Hyperemesis gravidarum induced Wernicke's encephalopathy: serial clinical, electrophysiological and MR imaging observations. J Neurol Sci 2009;284(no. 1-2):214–6.
[7] Zuccoli G, Santa Cruz D, Bertolini M, *et al*. MR imaging findings in 56 patients with Wernicke encephalopathy: nonalcoholics may differ from alcoholics. AJNR Am J Neuroradiol 2009;30:171–6.
[8] Ebels EJ. Underlying illness in Wernicke's encephalopathy: analysis of possible causes of under diagnosis. Eur Neurol 1974;12:226–8.
[9] Chu K, Kang DW, Kim HJ, *et al*. Diffusion-weighted MR abnormalities in Wernicke's encephalopathy: reversible cytotoxic oedema. Arch Neurol 2002;59:123–7.