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Case Report

Cervical cord lesions in Wernicke's encephalopathy[☆]Sho Ishikawa, MD^{a,*}, Kumiko Ando, MD, PhD^a, Takashi Katakami, MD^b, Michi Kawamoto, MD, PhD^b^a Department of Diagnostic Radiology, Kobe City Medical Center General Hospital, 2-1-1 Minatojima-minamimachi, Chuo-ku, Kobe, Hyogo, 650-0047, Japan^b Department of Neurology, Kobe City Medical Center General Hospital, 2-1-1 Minatojima-minamimachi, Chuo-ku, Kobe, Hyogo, 650-0047, Japan

ARTICLE INFO

Article history:

Received 10 March 2022

Revised 4 April 2022

Accepted 5 April 2022

Keywords:

Wernicke's encephalopathy

Spinal lesion

Vasogenic edema

MRI

FLAIR

ABSTRACT

A 30-year-old woman suffering from an eating disorder and alcoholism presented with a progressively worsening gait disturbance lasting 2 weeks. Her neurological findings included impaired ocular motility and trunk ataxia. Fluid-attenuated inversion recovery imaging of the brain showed hyperintensity in the dorsal brainstem, aqueduct, thalamus, and cerebral cortex. A long hyperintense segment on T2-weighted imaging was visible in the central gray matter of the cervical spinal cord. No restricted diffusion was observed; thus, T2 elongation in the spine was suggested to be due to vasogenic edema. We diagnosed the patient with Wernicke's encephalopathy and initiated vitamin supplementation. Thereafter, her symptoms rapidly improved; magnetic resonance imaging on the 11th day of hospitalization showed normalization of the signals in her brain and spinal cord. As our case demonstrates, Wernicke's encephalopathy can induce vasogenic edema of the spinal cord, which can rapidly improve with early therapeutic intervention.

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Introduction

Wernicke's encephalopathy is a neurological disorder caused by a deficiency of vitamin B1. The three main symptoms are impaired consciousness, abnormal eye movements, and ataxia. Alcoholism, gastrectomy, malabsorption syndrome, and malignancy are known risk factors [1,2]. Brain magnetic resonance imaging (MRI) shows high signal in the midbrain

aqueduct, periventricular third ventricle, thalamus, and mammillary bodies on T2-weighted imaging (T2WI) and fluid-attenuated inversion recovery (FLAIR) imaging, and is useful in diagnosing Wernicke's encephalopathy [2–4]. There are no reports of spinal cord involvement in Wernicke's encephalopathy. In this report, we describe a case of Wernicke's encephalopathy with a reversible abnormal signal area in the spinal cord.

[☆] Competing Interests: The authors have no conflicts of interest to disclose.

* Corresponding author.

E-mail address: shoishikawa0819@gmail.com (S. Ishikawa).<https://doi.org/10.1016/j.radcr.2022.04.004>1930-0433/© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

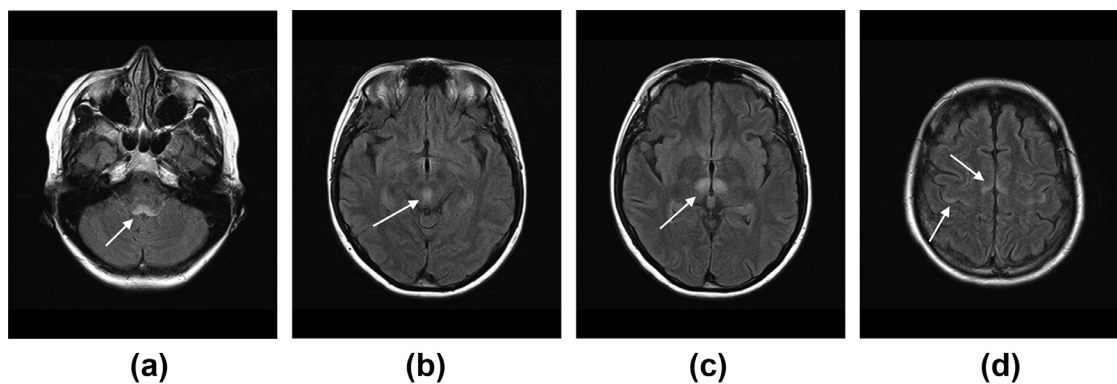


Fig. 1 – Brain magnetic resonance imaging (MRI) before treatment. Axial fluid-attenuated inversion recovery (FLAIR) imaging (TR: 8000 msec, TE: 116 msec, TI: 2500 msec, FA: 170, slice thickness: 5.0 mm) of the brain showed hyperintensities in the dorsal medulla oblongata (A) (arrow), periventricular gray matter surrounding third ventricle (B) (arrow), thalamus (C) (arrow), and cerebral cortex (D) (arrows). These lesions showed no water diffusion restriction (not shown).

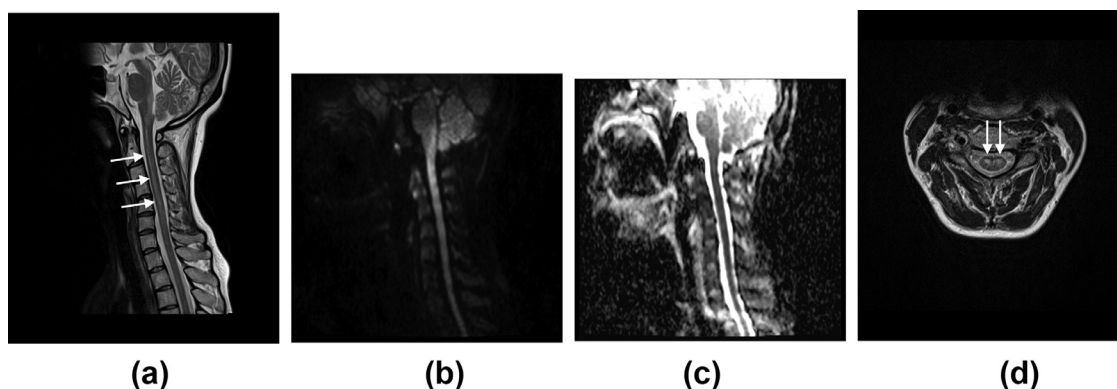


Fig. 2 – Cervical spine magnetic resonance imaging (MRI) before treatment. Sagittal (TR: 4000 msec, TE: 91 msec, slice thickness: 4.0 mm) and axial (TR: 3610 msec, TE: 82 msec, slice thickness: 4.0 mm) T2-weighted imaging of MRI of the cervical spine (A, D) showed longitudinal hyperintensity in the central gray matter of the spinal cord (arrows). The lesion showed hyperintensity on sagittal diffusion-weighted imaging (TR: 3100 msec, TE: 78 msec, slice thickness: 4.0 mm, $b = 600 \text{ s/mm}^2$) with a high apparent diffusion coefficient (B, C).

Case report

A woman in her thirties presented with a 1-month history of severe anorexia and a 2-month history of dizziness and difficulty walking. She had a history of depression, insomnia, eating disorders, and heavy alcohol consumption.

The neurological findings at the time of admission showed eye movement disorder. In addition, myotonia was observed throughout the body, tendon reflexes were enhanced, and pathological reflexes were also observed.

Blood tests on arrival showed metabolic acidosis, but there were no other significant abnormal findings. Urinalysis and spinal fluid examination also showed no abnormal findings.

Brain MRI was performed and hyperintensity on FLAIR imaging were found in the dorsal medulla oblongata, periven-

tricular gray matter surrounding third ventricle, thalamus, and cerebral cortex. These lesions showed no water diffusion restriction (Fig. 1).

Cervical spine MRI also showed extensive hyperintensity on T2WI without water diffusion restriction in the central gray matter of the spinal cord (Fig. 2).

Based on clinical and imaging findings, the patient was considered to have Wernicke's encephalopathy and vitamin supplementation therapy was started.

After the start of treatment, the eye movement disorder rapidly improved, and her gait and muscle weakness also improved in 2 weeks.

Results of blood tests after the start of treatment showed decreases in vitamins and trace elements and negative results for anti-aquaporin 4 antibody. The results were consistent with Wernicke's encephalopathy.

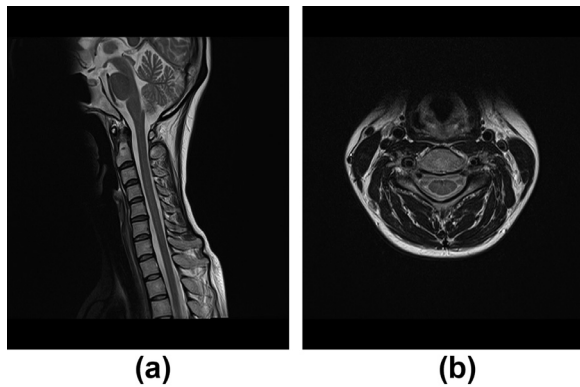


Fig. 3 – Cervical magnetic resonance imaging (MRI) 11 days after treatment. Sagittal and axial T2-weighted imaging of cervical spine showed regression of hyperintensities in the cervical spine (A, B).

Brain and cervical spine MRI 11 days after the start of treatment demonstrated disappearance of abnormal hyperintensities on T2WI and FLAIR imaging (Fig. 3).

Discussion

Vitamin B1 is a water-soluble vitamin that acts as a coenzyme in the glycolytic and citric acid circuits [1,2]. Wernicke's encephalopathy is suggested to be caused by vitamin B1 deficiency. The treatment is vitamin supplementation, and the earlier the treatment is started, the better the prognosis. If treatment is delayed, the prognosis is poor, and severe cases often progress to irreversible Korsakoff's syndrome, characterized by residual mental disorders, memory impairment, retrograde amnesia, and language impairment. When untreated, the mortality rate is 10%-20% [1].

The classic triad of symptoms of Wernicke's encephalopathy - impaired consciousness, oculomotor disturbances, and ataxia - is present in only 16%-38% of cases [5]. Low vitamin levels in blood tests can help diagnose Wernicke's encephalopathy, but it takes several days to get the results of blood tests. In addition, serum thiamine levels do not necessarily reflect those of the spinal fluid, and even normal serum levels do not rule out the diagnosis [6]. Therefore, Wernicke's encephalopathy is usually suspected based on clinical information and diagnostic treatment is given.

Brain MRI is a useful tool for early diagnosis of Wernicke's encephalopathy. The characteristic findings on brain MRI are symmetric T2-hyperintensities in the mammillary bodies, periventricular gray matter surrounding third ventricle, and medial thalamus. Water diffusion restriction may or may not be seen. Hemorrhages on T2*-weighted imaging and contrast enhancement by gadolinium contrast medium may be seen in acute lesions [2-4].

Wernicke's encephalopathy is thought to be due to damage to astrocytes in the early stages of the disease. Early le-

sions in the brains of thiamine-deficient mice often show edematous swelling of astrocytes, with little or no change in neurons or oligodendrocytes. As the tissue damage progresses, changes are seen in neurons and oligodendrocytes [7]. Diffusion-weighted imaging of brain lesions may or may not be restricted. The presence of diffusion restriction is thought to reflect cellular edema caused by damage to the cell membrane. The mechanism of cellular edema is thought to be a lack of intracellular ATP, resulting in decreased resistance to oxidative stress, decreased intracellular pH, and accumulation of toxic intermediate products, leading to cell death. The absence of diffusion restriction is thought to reflect extracellular edema caused by damage to the blood-brain barrier. Vitamin B1 deficiency depletes ATP, lowers the pH of the synaptic cleft, disrupts astrocytes, and leads to disruption of the blood-brain barrier [2].

Spinal cord lesions in Wernicke's encephalopathy are rare and have not been reported in the literature. The mechanism of the damage to specific areas in central nervous system in Wernicke's encephalopathy is not yet understood. In this case, extensive abnormal T2-hyperintensities of MRI were observed in the central gray matter of the spinal cord and also gray matters of the brain, including the periventricular gray matter surrounding the third ventricle, and the medial thalamus. These gray matter signal alterations in the spine and brain might be caused by the same mechanism. Of note, the spinal lesions of the patient improved with treatment, as did the brain lesion.

Wernicke's encephalopathy can present with extensive spinal cord involvement. Early therapeutic intervention can improve symptoms and abnormal MRI signals both in the brain and spine.

Patient consent

Informed written consent was obtained from the patient for publication of the case report and all imaging studies.

REFERENCES

- [1] Sechi G, Serra A. Wernicke's encephalopathy: new clinical settings and recent advances in diagnosis and management. *Lancet Neurol* 2007;6:442-55. doi:10.1016/S1474-4422(07)70104-7.
- [2] Ota Y, Capizzano AA, Moritani T, Naganawa S, Kurokawa R, Srinivasan A. Comprehensive review of Wernicke encephalopathy: pathophysiology, clinical symptoms and imaging findings. *Jpn J Radiol* 2020;38:809-20. doi:10.1007/s11604-020-00989-3.
- [3] Antunez E, Estruch R, Cardenal C, Nicolas JM, Fernandez-Sola J, Urbano-Marquez A. Usefulness of CT and MR imaging in the diagnosis of acute Wernicke's encephalopathy. *AJR Am J Roentgenol* 1998;171:1131-7. doi:10.2214/ajr.171.4.9763009.
- [4] Manzo G, De Gennaro A, Cozzolino A, Serino A, Fenza G, Manto A. MR imaging findings in alcoholic and nonalcoholic acute Wernicke's encephalopathy: a review. *Biomed Res Int* 2014;2014:503-96. doi:10.1155/2014/503596.

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- [5] Harper CG, Giles M, Finlay-Jones R. Clinical signs in the Wernicke-Korsakoff complex: a retrospective analysis of 131 cases diagnosed at necropsy. *J Neurol Neurosurg Psychiatry* 1986;49:341–5. doi:[10.1136/jnnp.49.4.341](https://doi.org/10.1136/jnnp.49.4.341).
- [6] Davies SB, Joshua FF, Zagami AS. Wernicke's encephalopathy in a non-alcoholic patient with a normal blood thiamine level. *Med J Aust* 2011;194:483–4. doi:[10.5694/j.1326-5377.2011.tb03068.x](https://doi.org/10.5694/j.1326-5377.2011.tb03068.x).
- [7] Hazell AS. Astrocytes are a major target in thiamine deficiency and Wernicke's encephalopathy. *Neurochem Int* 2009;55:129–35. doi:[10.1016/j.neuint.2009.02.020](https://doi.org/10.1016/j.neuint.2009.02.020).