

Case report

A case of neuromyelitis optica diagnosed with a chronic subdural hematoma

Akira Tempaku¹

¹ Department of Neurosurgery, Hokuto Hospital, Japan

Abstract

Background: Chronic subdural hematoma (CSDH) is often found in the elderly owing to slight head trauma and is associated with several neurological disorders. Neurological deficits are cured by a simple surgical removal of the hematoma; however, these deficits persist if there is insufficient hematoma removal. It is rare for patients to continue having neurological disorders once the hematoma is removed.

Case report: A 61-year-old woman presented with gait disturbance. She was diagnosed with a subdural hematoma through head computed tomography. After hematoma irrigation, her gait disturbance exacerbated, and she developed urinary tract dysfunction. Ubiquitous neurodegeneration in the midbrain and spinal cord was suspected owing to a hyperintense signal on fluid-attenuated inversion recovery of magnetic resonance imaging. The anti-aquaporin 4 antibody was detected in the patient's serum, and she was diagnosed with neuromyelitis optica (NMO).

Conclusions: Progressive NMO caused gait dysfunction and triggered head trauma, followed by CSDH. Although NMO rarely causes CSDH, it should be considered in uncommon cases of CSDH.

Key words: neuromyelitis optica, chronic subdural hematoma, anti-aquaporin 4 antibody

(*J Rural Med 2017; 12(2): 126–129*)

Introduction

Chronic subdural hematoma (CSDH) is known to occur mainly in the elderly owing to slight head trauma and is as-

sociated with mild hemiparesis and cognitive disorders^{1, 2}. Neurological deficits can be well cured through simple surgical removal of the hematoma; however, neurological dysfunction persists if there is insufficient hematoma removal or postoperative re-bleeding. Nevertheless, it is very rare for patients to show worse neurological disorders after hematoma removal.

Case Report

A 61-year-old woman with diabetes mellitus (DM) had gait disturbance and a mild decline in cognitive function. Approximately 2 months prior, she experienced dizziness, digestive disorders, and gait disturbance. Several internal medical studies, including serological and radiological tests of the abdominal organs (conducted at another hospital), were unable to detect the cause of the symptoms. Head computed tomography (CT) showed no specific abnormalities. She recovered without any specific treatment within several weeks.

Two months later, she had a slight gait disturbance again and visited our neurological clinic. Her consciousness level was normal. Neurological analysis revealed right hemiparesis, including the right upper extremity, which was Barre positive, a discrete movement disorder of the right finger, and ataxic gait. She had no verbal disturbance. She was diagnosed with a subdural hematoma through head CT (Figure 1A). The depiction of the left sulcus of the brain decreased, and a mid-line shift was not observed. Her neurological dysfunction exacerbated within 1 week. She was admitted, and hematoma irrigation was performed. After the surgical intervention, her gait disturbance exacerbated (Figure 1B). Furthermore, urinary tract dysfunction, sensory disturbances, decrease in deep tendon reflexes, and posterior column symptoms developed. Ubiquitous neurodegeneration in the midbrain and cervical spinal cord was suspected owing to a hyperintense signal on fluid-attenuated inversion recovery

Received: December 2, 2016

Accepted: June 5, 2017

Correspondence: Akira Tempaku, Department of Neurosurgery, Hokuto Hospital, 7-5, Inadacho-Kisen, Obihiro, Hokkaido 080-0833, Japan
E-mail address: tempaku@hokuto7.or.jp

This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial No Derivatives (by-nc-nd) License <<http://creativecommons.org/licenses/by-nc-nd/4.0/>>.

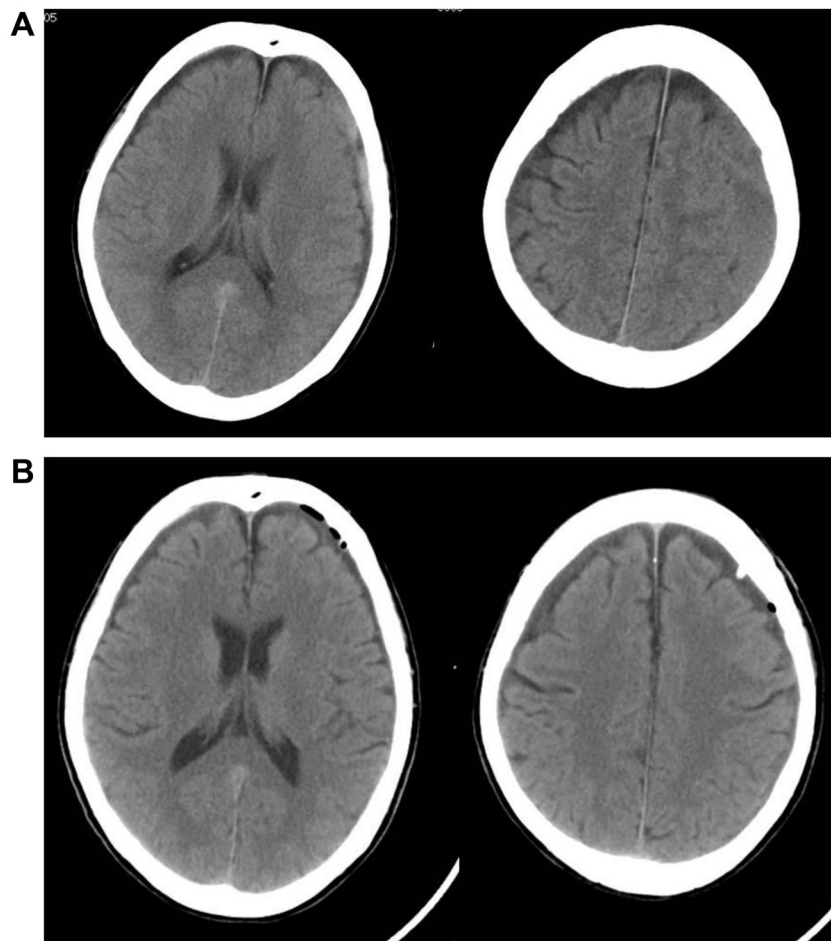


Figure 1 Head computed tomography scan images at admission (A) and post irrigation (B).

ery (FLAIR) and diffusion-weighted sequences of magnetic resonance imaging (MRI) (Figure 2). The oligoclonal band was negative. The myelin basic protein level in the cerebrospinal fluid increased to 110.3 pg/mL. The anti-aquaporin 4 antibody (AQP4Ab) was detected in the serum through a cell-based assay (CBA) (Cosmic Corporation, Tokyo, Japan), and she was diagnosed with neuromyelitis optica (NMO). The FLAIR hyperintense signal was improved by high-dose administration of a steroid (1 g Predonine intravenously for 3 days) (Figure 2). Her neurological disorders was improved.

Discussion

This patient with a moderate CSDH continued to have central nervous system disorders after the removal of the intracranial hematoma. A further study revealed another underlying neurodegenerative disease, which had caused the gait disturbance. The gait disturbance resulted in head

trauma by sequential fall down, which was accompanied by CSDH. CSDH was proven to be the result of trauma due to a neurodegenerative disease, and was not the cause of neurological disturbances.

Both multiple sclerosis (MS) and NMO affect the remission and exacerbation courses in the central nervous system. Both neurodegenerative diseases cause neural dysfunctions including hemiparesis, minute exercise difficulty, and gait disturbance^{3, 4}. These symptoms caused head trauma, followed by CSDH. As this patient had never been diagnosed with MS or NMO until CSDH formation, initiation of the appropriate treatment was delayed. She had a clinical history of digestive disorders and gait disturbance 1 year prior to the CSDH formation. These symptoms were suspected to be the clinical features of NMO. A postoperative decrease in limb mobility and progression of antagonistic muscle coordination disturbances were signs of NMO exacerbation.

It is rare for NMO to cause CSDH, but it is necessary to consider NMO in atypical cases of CSDH. A PubMed search

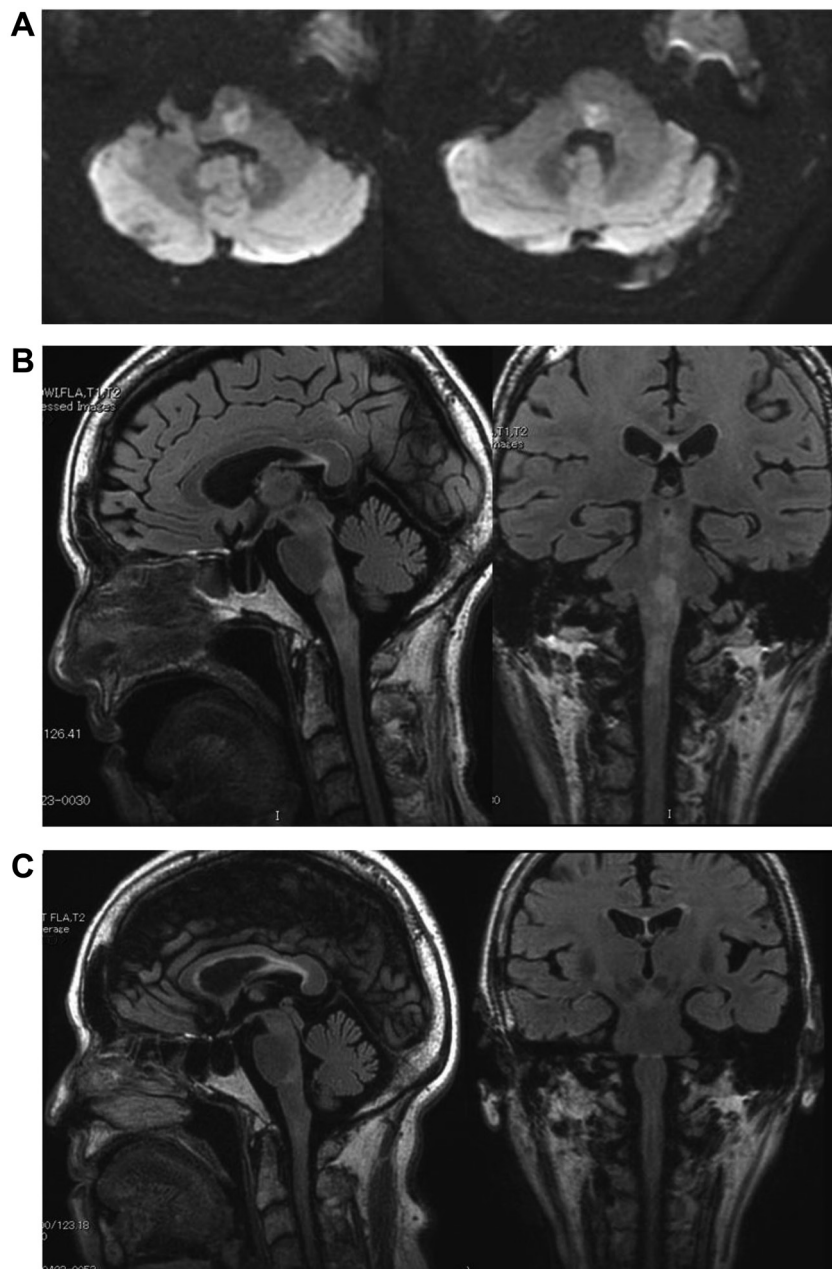


Figure 2 Head magnetic resonance imaging of the neurological disorder worsening over time (A, B) and at remission (C). A shows diffusion weighted imaging findings of the axial plane on the medulla level. B and C show fluid-attenuated inversion recovery findings of the sagittal and coronal planes of the brain stem region.

showed no previous reports on CSDH caused by NMO. An association of CSDH with MS and Parkinson's disease has been reported⁹. The present case shows that there is a possibility that NMO causes CSDH, following head trauma, that is accompanied by gait disturbance. It is necessary to consider other causes in patients with CSDH without any un-

derlying disease. MRIs of whole brain and spine are useful to detect NMO, MS, or other neurodegenerative diseases. In terms of medical economic restriction, however, we should refrain from the MRI for prior-diagnosed patients with CSDH. Any discrepancy between neurological disorders and radiological imaging results should be handled careful-

ly, and additional imaging tests should be performed immediately. In this patient, it was retrospectively recognized that the digestive disorders and gait disturbance were the first symptoms. However, it might have been difficult to suspect a neurodegenerative disease from these symptoms at the primary care hospital. Furthermore, because the symptoms disappeared immediately without any special medication, it became difficult to make an accurate diagnosis.

In this case, the irrigation of CSDH did not contribute to recovery from any neurological disorder; in fact, her symptoms worsened postoperatively. Such conditions should be suspected to coincide with other basic neurological diseases, and an MRI or serum test should be performed.

An enzyme-linked immunosorbent assay (ELISA) can detect AQP4Ab in the serum⁶. However, in this case, AQP4Ab could not be detected using this method and only CBA could detect AQP4Ab. It has been reported that CBA is a more sensitive test for detecting AQP4Ab than is ELISA⁷⁻⁹. It was inferred that a low AQP4Ab level resulted in a contradiction of AQP4Ab identification between the ELISA and CBA methods.

The International Panel for Neuromyelitis Optica Diagnosis determines that neuromyelitis optica spectrum diseases can be diagnosed with or without AQP4Ab¹⁰, and this case conformed to this criterion, prior to the identification of AQP4Ab. A suspected myelitis lesion was detected with the special multiplicities using FLAIR of MRI. The lesion was distributed from the brain stem to the cervical spinal cord and was more than three vertebral bodies high. Although AQP4Ab detection was negative on ELISA, the patient's clinical features fulfilled the four required criteria, and she had two core symptoms (acute myelitis and acute brain stem symptoms). Prior to the detection of AQP4Ab using CBA, a clinical diagnosis of NMO was made. Furthermore, AQP4Ab detection using CBA confirmed the NMO diagnosis. In any case, the iteration serum test using plural assays was necessary to make the diagnosis.

NMO should be considered as a cause of uncommon cases of CSDH. CSDH is common in patients with DM, renal dysfunction, and hepatic disorders². It develops easily in the elderly and patients receiving anti-platelet or anti-coagulation therapy¹¹. Unlike the causes of CSDH reported in the past, neurodegenerative diseases are easy to be overlooked. However, if a medical history of CSDH is not found in the precedent reports, underlying unknown neurodegenerative diseases should be considered.

Conclusion

NMO must be considered as a cause of CSDH development in patients without any known basal disorders. A fall owing to NMO exacerbation may cause CSDH.

Conflicts of Interest: There are no potential conflicts of interest to disclose. The author has no personal financial interest in any of the drugs, materials, or devices mentioned in this article.

References

1. Markwalder TM. Chronic subdural hematomas: a review. *J Neurosurg* 1981; 54: 637–645. [[Medline](#)] [[CrossRef](#)]
2. Chen JC, Levy ML. Causes, epidemiology, and risk factors of chronic subdural hematoma. *Neurosurg Clin N Am* 2000; 11: 399–406. [[Medline](#)]
3. Jasiak-Zatonska M, Kalinowska-Lyszczarz A, Michalak S, *et al.* The immunology of neuromyelitis optica-Current knowledge, clinical implications, controversies and future perspectives. *Int J Mol Sci* 2016; 17: 273. [[Medline](#)] [[CrossRef](#)]
4. Kawachi I, Lassmann H. Neurodegeneration in multiple sclerosis and neuromyelitis optica. *J Neurol Neurosurg Psychiatry* 2016; 26. pii: jnnp-2016-313300.
5. Harding AE. Subdural haematoma in two patients with chronic neurological disorders. *Br Med J (Clin Res Ed)* 1984; 288: 1986–1987. [[Medline](#)] [[CrossRef](#)]
6. Hayakawa S, Mori M, Okuta A, *et al.* Neuromyelitis optica and anti-aquaporin-4 antibodies measured by an enzyme-linked immunosorbent assay. *J Neuroimmunol* 2008; 196: 181–187. [[Medline](#)] [[CrossRef](#)]
7. Sato DK, Nakashima I, Takahashi T, *et al.* Aquaporin-4 antibody-positive cases beyond current diagnostic criteria for NMO spectrum disorders. *Neurology* 2013; 80: 2210–2216. [[Medline](#)] [[CrossRef](#)]
8. Ruiz-Gaviria R, Baracaldo I, Castañeda C, *et al.* Specificity and sensitivity of aquaporin 4 antibody detection tests in patients with neuromyelitis optica: A meta-analysis. *Mult Scler Relat Disord* 2015; 4: 345–349. [[Medline](#)] [[CrossRef](#)]
9. Waters P, Reindl M, Saiz A, *et al.* Multicentre comparison of a diagnostic assay: aquaporin-4 antibodies in neuromyelitis optica. *J Neurol Neurosurg Psychiatry* 2016; 87: 1005–1015. [[Medline](#)] [[CrossRef](#)]
10. Wingerchuk DM, Banwell B, Bennett JL, *et al.* International Panel for NMO Diagnosis. International consensus diagnostic criteria for neuromyelitis optica spectrum disorders. *Neurology* 2015; 85: 177–189. [[Medline](#)] [[CrossRef](#)]
11. De Bonis P, Trevisi G, de Waure C, *et al.* Antiplatelet/anticoagulant agents and chronic subdural hematoma in the elderly. *PLoS ONE* 2013; 8: e68732. [[Medline](#)] [[CrossRef](#)]