Follow-Up of Brain Single-Photon Emission Computed Tomography (SPECT) and Magnetic Resonance Imaging (MRI) in a Case of Seizure Caused by Osmotic Demyelination Syndrome

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Patient: Male, 38-year-old
Final Diagnosis: Osmotic demyelination syndrome
Symptoms: Seizure
Medication:
Clinical Procedure: —
Specialty: Nuclear Medicine • Radiology

Objective: Challenging differential diagnosis
Background: Osmotic demyelination syndrome (ODS) is an uncommon neurological disorder. Until the mid-1980s, the mortality rate was 90–100%, but more than half of patients now have a good prognosis. Early suspicion of ODS is important. However, radiologic findings of ODS are variable and scintigraphy findings have not been reported.

Case Report: A 38-year-old man with alcohol abuse history was admitted due to electrolyte imbalance. On the 10th day of his hospital stay, he had a generalized tonic-clonic seizure. Brain perfusion SPECT showed asymmetrically hyperperfused and hypoperfused lesions. Brain MRI revealed diffuse T2 hyperintensity with mild diffusion restriction in the pons and hyperperfused lesions on brain SPECT. He was treated based on the diagnosis of hyponatremia and osmotic demyelination. After treatment, the asymmetric hyperperfusion was decreased. MRI showed that the cortical hyperintensity had resolved, with encephalomalacic change shown in the pons.

Conclusions: To the best of our knowledge, this is the first report showing changes in brain perfusion SPECT and MRI in an ODS patient with a seizure. This case report may be helpful to neurologists, radiologists, and nuclear physicians.

MeSH Keywords: Demyelinating Diseases • Follow-Up Studies • Magnetic Resonance Imaging • Tomography, Emission-Computed, Single-Photon

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Background

Osmotic demyelination syndrome (ODS) is an uncommon neurological disorder found in about 0.4–0.56% of all neurological admissions in tertiary hospitals [1–3].

ODS is caused by damage to the myeline sheath of brain cells [4,5]. Although the exact pathogenesis is unclear, rapid correction of chronic hyponatremia is the most frequent predisposing factor [3,5–7]. During rapid correction of hyponatremia, if the brain is unable to recover organic osmolytes quickly enough, cellular dehydration and myeline sheath damage occur [8]. Other related conditions include alcoholism, liver cirrhosis, severe burns, and malnutrition [5,3]. Reflecting the greater susceptibility of white matter tracts of the pontine, central-pontine myelinolysis (CPM) is the classical presentation [5]. However, extrapontine myelinolysis (EPM) is also reported [5].

Clinical manifestations of ODS are highly variable and are correlated with the site affected. Neurologic symptoms such as seizures, paresis, dysarthria, ataxia, and coma are reported [5]. Until the mid-1980s, the mortality rate was 90–100% [3], but more than half of patients now have a good prognosis [5]. However, there is still a possibility of death and disability, especially in patients with liver transplantation [5]. A recent review found that one of the reasons for the current favorable prognosis is earlier detection of ODS by imaging study and progress in intensive care medicine, such as precise electrolyte management [3]. Early suspicion of ODS is important, but radiologic findings of ODS are variable [9,10]. Scintigraphy findings of seizure due to ODS and imaging follow-up studies have not been reported.

Here, we report a case with brain perfusion SPECT and MRI images in an alcoholic patient with seizure due to ODS. Follow-up brain perfusion SPECT and MRI were also done. To the best of our knowledge, this case is the first case report describing changes of brain perfusion SPECT and MRI in this syndrome in a patient with seizure.

Case Report

A 38-year-old man patient presented at our hospital with palpitation, nausea, vomiting, and cold sweats. He has been an irregular and casual drinker for 8 years. Before admission, he drank several bottles of soju for 3 days in a row. He showed mild hand tremor and his speech did not make sense. Laboratory testing revealed elevated liver function: AST (94.4 U/L) and ALT (66.3 U/L). An electrolyte imbalance was also revealed: hyponatremia (115 mEq/L, reference range 136–146 mEq/L), hypokalemia (2.5 mEq/L, reference range 3.5–5.0 mEq/L), and hypochloremia (69 mEq/L, reference range 98–110 mEq/L).

He was admitted and the electrolyte imbalance was treated intravenously. During the first 2 days, the correction speed for hyponatremia was 10 mmol/L/day, and was then slowed to 2–3 mmol/L/day. The hyponatremia was corrected after 7 days, and his liver function also normalized. He had been listless during those days. On the 6th and 7th days, he complained of headache with numeric rating scale (NRS) 5 points. On the 8th day, he was free from headache. On the 10th day of admission, he complained of moderate headache (NRS 6 points) from 5 a.m. At 5:55 a.m., he had a generalized tonic-clonic seizure. It was his first attack of seizure. He was sedated with lorazepam. Right after the seizure, cerebral perfusion single-photon emission computed tomography (SPECT) was performed with brain perfusion tracer (Tc-99m HMPAO). Brain perfusion SPECT revealed hyperperfusion in the right parietal, right occipital, and right lateral temporal areas. T2 hyperintensity with diffusion restriction in the right parietal, left parietal, and temporal areas. Brain magnetic resonance imaging (MRI, 3T) was performed the next day, revealing diffuse T2 hyperintensity with mild diffusion restriction in the right parietal, right occipital, and right lateral temporal areas. T2 hyperintensity with diffusion restriction was also shown in the pons. In addition, T2 hyperintensity without diffusion restriction was shown in the caudate nucleus, putamen, right thalamus, pulvinar, and middle cerebellar peduncle (Figure 2A, 2B). An electroencephalogram (EEG) showed persisting repetitive spikes in the right temporo-parieto-occipital region. In the Korean version of the Mini-Mental State Examination (K-MMSE), his score was 26 out of 30. He was unable to draw interlocking pentagrams.

There are many causes of seizure, including hyponatremia and alcohol abuse. However, these can be the causes of ODS as well. After discussion among clinicians, radiologists, and nuclear physicians, we concluded that the seizure was due to ODS. Under the impression of seizure induced by osmotic demyelination, he was treated by electrolytes correction, hydration and vitamin supplement. The patient had not had second seizure attack after the first one. Also, he had been alert and without headache.

Follow-up imaging studies (brain perfusion SPECT and brain MRI of 1.5T) were done after 2 months during an out-patient clinic visit schedule on SPECT, showing decreased asymmetric hyperperfusion. Only mild hypoperfusion in the bilateral frontal lobe remained (Figure 1B). On MRI, the cortical hyperintensity and swelling of the right-side area on previous imaging was resolved completely, without atrophic change. In the medial pons, encephalographic change was first shown. At 7-month follow-up, he had not had any further seizures.
Discussion

To the best of our knowledge, this is the first case report describing changes in brain perfusion SPECT and MRI in an alcoholic patient with seizure due to ODS with central-pontine and extrapontine myelinolysis. On follow-up images with brain perfusion SPECT and MR, the seizure-related changes had resolved. There has not been a previous report showing seizure-related changes on brain perfusion SPECT in a patient with both CPM and EPM. In 2015, Chatterjee et al. reported brain perfusion SPECT in a patient with alcoholism and CPM [8]. However, in their case, brain SPECT was done after the seizure event. In contrast to our case, there was only hypoperfusion in the bilateral frontal lobes, which also was thought to be due to chronic alcoholism [8]. In our case, prominent hyperperfusion in the right cortex was shown on the brain perfusion SPECT, which was done on the day of the seizure (Figure 1). Hyperperfusion in the seizure focus on brain perfusion SPECT on the ictal timing is a well-known feature [11]. In our case, hyperperfused lesions on brain SPECT were strongly correlated with the lesions shown by MRI changes – T2 hyperintensity with mild diffusion.

Figure 1. Brain perfusion SPECT (Tc-99m HMPAO) images. 3D Talairach cortical perfusion images of the perfusion SPECTs (A) on the day of seizure (4 hours after seizure onset), and (B) after 2 months. (A) On the day of the seizure, hyperperfusion was observed in the right lateral parietal and in the right occipital and right lateral temporal lobes. The patient was sedated during the scan. (B) On follow-up after 2 months, except for mild hypoperfusion in the frontal lobes, the previous perfusion abnormality had disappeared. There was not second seizure attack.
In this patient, demyelination was also shown in the thalamus, basal ganglia, and pons. However, on brain perfusion SPECT, the perfusion in the thalamus and basal ganglia was within normal range in this case, but the reason for this is unclear. There has not been a report of scintigraphic findings on ODS.

The typical MR finding of ODS is hyperintense on T2-weighted images and fluid attenuated inversion recovery sequences, with corresponding hypointensity on T1-weighted images in the central pons or associated extrapontine structures [9,5,10,12]. However, Singh et al. reported that restricted diffusion was found only 45.5% of reported patients. There has been no clear explanation of the MR signal changes related to the seizure [13–15]. However, restricted diffusion on MR is usually associated with acute cerebral infarction, neoplasms, intracranial infections, traumatic brain injury, and status with demyelination processes [16,17]. In our case, osmotic demyelination seemed to induce the diffusion change on MRI. In this case, there were other possibilities of MR changes suggested by radiologists, such as seizure-related MRI signal change and unusual Wernicke’s encephalopathy. Wernicke’s encephalopathy was excluded after clinical expressions and laboratory findings. Still, there remain the seizure-related MR signal changes.

Figure 2. Magnetic resonance images on the day of seizure (A, B) and after 2 months (B, C). T2 flair image on the day after the seizure episode showing T2 hyperintensity without diffusion restriction in both caudate nucleus (arrows), putamen (arrows), right thalamus (arrows), pulvinar, and both middle cerebellar peduncles. Osmotic demyelination syndrome was suggested. 

(B) Diffuse T2 hyperintensity with mild diffusion restriction in right cerebral cortical area (arrows). Cortical signal change was predominantly in the insular temporal and parieto-occipital lobe. (C, D) After 2 months, follow-up MR showed that T2 hyperintensity was normalized (T2-weighted images).

In our case, the hyperperfusion and signal change in the right cerebral cortical area (predominantly insular temporal and parieto-occipital lobe) disappeared on the follow-up images after 2 months. The patient did not have any further seizures. This clinical feature was correlated with the image findings. However, there remained some changes on brain perfusion SPECT, showing mild hypoperfusion in bilateral frontal lobes. This hypoperfusion was thought to be related to chronic alcoholism. It is known that independent of brain atrophy, two-thirds of chronic alcoholics show frontal lobe changes [18], and hypoperfusion in frontal areas is reported in chronic alcoholics [18–20].

Due to greater susceptibility of white-matter tracts of the pontine, CPM is the classical presentation [5]. However, a recent article assessing studies published between 1959 and 2013 revealed CPM only (56.1%), EPM only (12.8%), and both CPM and EPM (31.1%) based on radiological diagnosis [5]. Our patient had both CPM and EPM.

Not all patients with hyponatremia develop ODS [3]. As mentioned above, rapid correction of chronic hyponatremia is the most frequent predisposing factor [3,5–7]. Our patient was thought to have chronic hyponatremia status on admission. However, the correction of hyponatremia was done slowly enough, at less than 10 mmol/day. Singh et al. found that ODS can occur in patients with sodium >135 mmol/l (20.1% of enrolled patients) [5].

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Conclusions

Brain perfusion SPECT with MRI can show changes related to seizure due to ODS. To the best of our knowledge, this is the first case report demonstrating changes in brain perfusion SPECT and MRI in an alcoholic patient with seizure due to ODS with both central-pontine and extrapontine myelinolysis. This case report may be helpful to neurologists, radiologists, and nuclear physicians who read images of patients with this uncommon disease.

Conflict of interest

None.

References:

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